UNIWERSYTET MEDYCZNY
IM. PIASTÓW ŚLĄSKICH WE WROCŁAWIU

ROZPRAWA DOKTORSKA

„Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci”

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Chciałem złożyć podziękowania rodzinie, przyjaciołom i współpracownikom, którzy mieli ogromny wpływ na kształt tej pracy. Zawsze mogłem liczyć na Wasze wsparcie, pomoc i doświadczenie.

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Spis treści

1. Wykaz publikacji stanowiących rozprawę doktorską

1. **Sobieski M**., Sobieska A., Sekułowicz M., Bujnowska-Fedak M.M. Tools for early screening of autism spectrum disorders in primary health care – a scoping review. *BMC Primary Care* **23**, 46 (2022). https://doi.org/10.1186/s12875-022-01645-7
IF = 2,9; Pkt. MNiSW/KBN: 100 pkt

2. **Sobieski M**., Wrona S., Flakus M., Pierchała K., Sobieska A., Podgórska K., Wołowicz A., Sekułowicz M., Bujnowska-Fedak MM. Reliability and validity of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist. *Research in Autism Spectrum Disorders* **117**, 102454 (2024). https://doi.org/10.1016/j.rasd.2024.102454
IF = 2,2; Pkt. MNiSW/KBN: 140 pkt

3. **Sobieski M**., Kopszak A., Wrona S., Bujnowska-Fedak M.M. Screening accuracy and cut-offs of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist. *PLOS One* **19**(8): e0299618 (2024)
IF = 2,9; Pkt. MNiSW/KBN: 100 pkt

4. **Sobieski, M**.; Grata-Borkowska, U.; Bujnowska-Fedak, M.M. Implementing an Early Detection Program for Autism Spectrum Disorders in the Polish Primary Healthcare Setting—Possible Obstacles and Experiences from Online ASD Screening. *Brain Sciences* **14**, 388 (2024). https://doi.org/10.3390/brainsci14040388
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Oświadczenia współautorów określający ich wkład w powstanie poszczególnych prac stanowi załącznik 3 niniejszej rozprawy doktorskiej.

2. Wykaz stosowanych skrótów

90%CI – 90% przedział ufności (ang. 90% *confidence interval)*

95%CI – 95% przedział ufności (ang. 95% *confidence interval)*

ADOS-2 – zestaw diagnostyczny *Autism Diagnostic Observation Schedule 2nd-Version*

ASD – zaburzenia ze spektrum autyzmu (ang. autism spectrum disorders)

ASRS – kwestionariusz *Autism Spectrum Rating Scales*

CFA – konfirmacyjna analiza czynnikowa (ang. *confirmatory factor analysis*)

CFI – porównawczy wskaźnik dopasowania (ang. *comparative fit index*)

CI – przedział ufności (ang. *confidence interval)*

CSBS-DP ITC – kwestionariusz Communication and Symbolic Behaviour Scales – Infant-Toddler Checklist

DD – zaburzenia rozwojowe (ang. developmental disorders)

df – ilość stopni swobody (ang. *degrees of freedom*)

DSM - Podręcznik Diagnostyczny i Statystyczny Zaburzeń Psychicznych (ang. *Diagnostic and Statistical Manual of Mental Disorders*)

DWLS – metoda estymacji z użyciem metody najmniejszych kwadratów przekątnych między dwoma badanymi zmiennymi (ang. *diagonally weighted least squares*)

H – wynik statystyki H testu Kruskala-Wallisa

ICC3,B – współczynnik korelacji wewnątrzklasowej (ang. *intraclass correlation coefficient*), wariant 3 (typ B)

ICD-10 - Międzynarodowa Statystyczna Klasyfikacja Chorób i Problemów Zdrowotnych, wydanie 10 (ang. *International Statistical Classification of Diseases and Related Health Problems 10th Revision*)

IT – technologie informatyczne (ang. *information technology*)

LD – opóźnienie rozwoju mowy (ang. *language delay*)

LR+ - iloraz wiarygodności wyniku dodatniego (ang. *likehood ratio positive*)

LR- - iloraz wiarygodności wyniku ujemnego (ang. *likehood ratio negative*)

M – średnia (ang. *mean*)

Me - mediana

NPV – negatywna wartość predykcyjna (ang. *negative predictive value*)

PPV – pozytywna wartość predykcyjna (ang. *positive predictive value*)

PRISMA – organizacja odpowiedzialna za wytyczne do tworzenia metaanaliz oraz przeglądów systematycznych (ang. *Preferred Reporting Items for Systematic reviews and Meta-Analyses*)

POZ – podstawowa opieka zdrowotna

r – wartość statystyki *r* testu korelacji Pearsona

RMSEA - pierwiastek ze średniego błędu aproksymacji próby (ang. *root mean square error of approximation*)

ROC – charakterystyka operacyjna odbiornika (ang. *receiver operating characteristic*)

TLI – wskaźnik Tuckera-Lewisa (ang. *Tucker-Lewis Index*)

WHO – Światowa Organizacja Zdrowia (ang. World Health Organization)

3. Streszczenie w języku polskim

Wstęp: Zaburzenia ze spektrum autyzmu (ang. *autism spectrum disorders,* ASD) to grupa zaburzeń neurorozwojowych o nieokreślonej etiologii, charakteryzująca się przede wszystkim występowaniem trudności w zakresie komunikacji, mowy, zachowań społecznych oraz zainteresowań, występująca ze średnią częstością 1:100 dzieci na świecie. Wczesna diagnostyka jest jedną z metod mającą na celu przeciwdziałanie negatywnym skutkom ASD, które obejmują m.in. obniżenie jakości życia dziecka oraz jego rodziny, trudności w zakresie edukacji i nawiązywaniu relacji międzyludzkich oraz w późniejszym życiu – możliwości podjęcia odpowiedniej pracy. Wczesne postawienie diagnozy ASD u dziecka pozwala na wczesne rozpoczęcie zindywidualizowanej terapii, która ma na celu kompensować deficyty dziecka oraz zwiększać szansę na normalne współuczestniczenie w społeczeństwie. Im wcześniej rozpocznie się taką terapię u dziecka obciążonego ASD, tym lepsze efekty można uzyskać – szczególnie w zakresie rozwoju mowy i komunikacji, zdolności poznawczych, a także zachowania odpowiedniego do sytuacji.

Fakt, iż lekarze rodzinni oraz pediatrzy są specjalistami z zakresu ochrony zdrowia, którzy statystycznie najczęściej mają kontakt z dzieckiem w wieku niemowlęcym i wczesnym dzieciństwie, umożliwia baczną obserwację dziecka i w ten sposób zauważenie wczesnych objawów ASD. W celu ułatwienia wczesnej diagnostyki ASD stworzono wiele kwestionariuszy diagnostycznych – jednym z nich jest Communication and Symbolic Behaviour Scales-Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC), który jest efektywnym narzędziem do przeprowadzenia badań przesiewowych w krajach anglojęzycznych. Poza krajami anglojęzycznymi narzędzia te zazwyczaj wykorzystywane są bez pełnej walidacji – tj. tłumaczone „słowo-w-słowo", bez przeprowadzenia działań mających dopasować je kulturowo i językowo celem zwiększenia ich efektywności i skuteczności.

Cel badania: Celem niniejszego badania było przeprowadzenie pełnego procesu walidacyjnego polskiej wersji kwestionariusza przesiewowego CSBS-DP ITC – z oceną właściwości psychometrycznych (trafność, rzetelność), diagnostycznych (czułość, swoistość) oraz ewentualnych różnic międzypłciowych, a także ocena ewentualnych potencjalnych trudności we wdrożeniu metod przesiewowych w kierunku ASD w codziennej praktyce lekarzy w Polsce.

Metodologia: Badanie było badaniem podłużnym, prowadzonym w większości drogą elektroniczną za pomocą kwestionariuszy CSBS-DP ITC, Autism Spectrum Rating Scales

(ASRS) oraz stacjonarnie – w przypadku procedur obserwacyjnych w ramach badania Autism Diagnostic Observation Schedule oraz oceny stabilności wyniku w przypadku ewaluacji jednego dziecka przez dwóch niezależnych oceniających (tj. stabilności *interrater*). W pierwszej części badania polegającej na wypełnieniu metryki osobowej i kwestionariusza CSBS-DP ITC wzięło udział 1461 dzieci w wieku od 6 do 24 miesiąca życia i ich rodziców, którzy tworzyli grupę normalizacyjną; w badaniu kontrolnym z użyciem CSBS-DP ITC wzięło udział 490 dzieci z grupy normalizacyjnej. Po ukończeniu przez dane dziecko 30 miesiąca życia kontaktowano się ponownie z rodzicami celem oceny rozwoju dziecka za pomocą kwestionariusza ASRS – w tym etapie wzięło udział 602 dzieci. W przypadku uzyskania dodatniego wyniku badania lub obaw rodziców co do rozwoju dziecka przeprowadzano ewaluację w kierunku ASD z użyciem baterii obserwacyjnej Autism Diagnostic Observation Schedule celem weryfikacji podejrzeń o występowaniu ASD u dziecka. Ponadto rodzice biorący udział w badaniu zostali poproszeni o udział w ocenie screeningu on-line za pomocą specjalnie przygotowanej ankiety – w tej części badania wzięło udział 418 rodziców. Informacje te uzupełniono danymi zebranymi wśród 95 lekarzy podstawowej opieki zdrowotnej na temat ich opinii nt. elektronicznego screeningu ASD oraz potencjalnych trudności we wprowadzeniu narzędzi przesiewowych w kierunku zaburzeń rozwojowych do codziennej praktyki lekarskiej.

Wyniki: Polska wersja kwestionariusza CSBS-DP ITC cechuje się wysoką stabilnością wyniku (α Cronbacha = 0,92), dobrym dopasowaniem modeli jednoczynnikowych dla wyniku ogólnego, jak i trójczynnikowych dla podskal narzędzia (odpowiednio: χ^2 = 1188.19, χ^2/df = 4,72, CFI = 0,98, TL = 0,98, RMSEA = 0,050, 90%CI RMSEA = 0,048-0,053, p < 0.001 oraz χ^2 = 973,72, χ^2/df = 3,91, CFI = 0,99, TL = 0,99, RMSEA = 0,045, 90%CI RMSEA = 0,042-0,048, p < 0.001), równoważnością międzypłciową oraz stabilnością w pomiarach metodami *test-retest* (dla wyniku całkowitego r = 0,83, p < 0,001) i *interrater* (ICC3 od 0,916 do 0,969). Kwestionariusz CSBS-DP ITC w zależności od grupy wiekowej cechuje się wysokimi wartościami czułości (od 0,667 do 0,750), swoistości (od 0,854 do 0,939), zadowalającą wartością predykcyjną dodatnią (od 0,261 do 0,400) oraz wysoką negatywną wartością dodatnią (od 0,980 do 0,981). Ponadto wynik w kwestionariuszu CSBS-DP ITC koreluje w stopniu słabym lub umiarkowanym z późniejszym wynikiem ASRS (siła efektu rośnie z wiekiem dziecka; maksymalnie r = -0,431, p < 0,001).

Zarówno rodzice jak i lekarze podstawowej opieki zdrowotnej dostrzegają konieczność wdrożenia metod przesiewowych w kierunku ASD do codziennej praktyki lekarskiej bez

widocznej jasnej preferencji co do metody badania (online vs stacjonarnie). Według lekarzy, największe trudności dotyczące wdrożenia takich działań do codziennej praktyki to brak wystarczającej ilości czasu oraz trudności z dostępem do specjalistów – psychiatrów, psychologów i pedagogów.

Wnioski: Kwestionariusz CSBS-DP ITC w wersji polskiej jest skutecznym i efektywnym narzędziem, mogącym wykrywać wczesne objawy zaburzeń ze spektrum autyzmu w grupie wiekowej od 9 do 24 miesięcy życia. Polska wersja CSBS-DP ITC jest pierwszym w pełni zwalidowanym narzędziem, które może zostać użyte w ramach badań przesiewowych w populacji polskich dzieci i cechuje się wysokimi wartościami psychometrycznymi.

Użycie kwestionariusza CSBS-DP ITC w wersji elektronicznej w ramach screeningu online może okazać się częściowym remedium na liczne trudności systemowe, które stoją na przeszkodzie we wdrożeniu stacjonarnego programu badań przesiewowych.

4. Streszczenie w języku angielskim

Introduction: Autism spectrum disorders (ASD) are a group of neurodevelopmental disorders of unspecified etiology, characterized primarily by observed difficulties in communication, speech, social behavior and interests, occurring with an average frequency of 1:100 children worldwide. Early diagnosis is one of the methods aimed at counteracting the negative effects of ASD, which include, among others: lower quality of life of the child and his family, difficulties in education and interpersonal relations, and later in life - the possibility of taking up appropriate work. Early diagnosis of ASD in a child allows for early initiation of individualized therapy, which aims to develop the child's deficits and increase the chance of normal participation in society. The earlier such therapy is introduced in a child with ASD, the better the results can be achieved - especially in the areas of development of speech and communication, cognitive abilities, and behavior appropriate to the situation.

The fact that family doctors and pediatricians are health care specialists who most often interact with children in infancy and early childhood makes it possible to closely observe the child and thus notice early symptoms of ASD. In order to facilitate the early diagnosis of ASD, many diagnostic questionnaires have been created - one of them is the Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC), which is an effective tool for screening in English-speaking countries. Outside English-speaking countries, these tools are usually used without full validation - i.e. translated "word-for-word", without any measures to adapt them culturally and linguistically to increase their effectiveness and efficiency.

Aim of the study: The aim of this study was to conduct a full validation process of the Polish version of the CSBS-DP ITC screening questionnaire - with the assessment of psychometric properties (validity, reliability), diagnostic properties (sensitivity, specificity) and possible sex differences, as well as to assess any potential difficulties in implementing ASD screening methods in the everyday practice of physicians in Poland.

Methodology: The study was a longitudinal study, conducted mostly online using the CSBS-DP ITC and Autism Spectrum Rating Scales (ASRS) questionnaires, and on-site - in the case of observation procedures as part of the Autism Diagnostic Observation Schedule examination and assessment of the stability of the result in the case of evaluation of one child by two independent evaluators (interrater stability). The first part of the study, consisting in filling out the personal metrics and the CSBS-DP ITC questionnaire, involved 1,461 children

aged 6 to 24 months and their parents, who formed the normalization group; 490 children from the standardization group participated in a control study using CSBS-DP ITC. After the child turned 30 months of age, parents were contacted again to assess the child's development using the ASRS questionnaire - 602 children took part in this stage. In the event of a positive test result or parents' concerns about the child's development, an evaluation for ASD was carried out using the Autism Diagnostic Observation Schedule battery to verify the suspicion of ASD in the child. In addition, parents participating in the study were asked to participate in the online screening evaluation using a specially prepared survey - 418 parents took part in this part of the study. This information was supplemented with data collected from 95 primary care physicians regarding their opinions on electronic ASD screening and potential difficulties in introducing screening tools for developmental disorders into everyday medical practice.

Results: The Polish version of the CSBS-DP ITC questionnaire is characterized by high stability (Cronbach's $\alpha = 0.92$), good fit of one-factor models for the overall result and three-factor models for the subscales of the tool ($\chi^2 = 1188{,}19$, $\chi^2/df = 4{,}72$, CFI = 0,98, TL = 0,98, RMSEA = 0,050, 90%CI RMSEA = 0,048-0,053, $p < 0.001$ and $\chi^2 = 973{,}72$, $\chi^2/df = 3{,}91$, CFI = 0,99, TL = 0,99, RMSEA = 0,045, 90%CI RMSEA = 0,042-0,048, $p < 0.001$, respectively), intersex equivalence and stability in measurements using test-retest method (for the total result $r = 0.83$, $p < 0.001$) and interrater method (ICC3,B from 0.916 to 0.969). The CSBS-DP ITC questionnaire, depending on the age group, is characterized by high values of sensitivity (from 0,667 to 0,750), specificity (from 0,854 to 0,939), a satisfactory positive predictive value (from 0,261 to 0,400) and a high negative positive value (from 0,980 to 0,981). Moreover, the score in the CSBS-DP ITC questionnaire correlates weakly or moderately with the later ASRS score (the strength of the effect increases with the child's age; maximum $r = -0{,}431$, $p < 0{,}001$).

Both parents and primary care physicians recognize the need to implement ASD screening methods into everyday medical practice, with no apparent clear preference for the screening method (online vs. stationary). According to doctors, the greatest difficulties in implementing such actions into everyday practice are the lack of sufficient time during visit and difficulties with access to other specialists - psychiatrists, psychologists and special educators.

Conclusions: The Polish version of the CSBS-DP ITC questionnaire is an effective and efficient tool that can detect early symptoms of autism spectrum disorders in the age group from 9 to 24 months of age. The Polish version of CSBS-DP ITC is the first fully validated tool that can be used as part of screening tests in the population of Polish children and is characterized by high psychometric and diagnostic properties.

Usage of the electronic version of the CSBS-DP ITC questionnaire as part of online screening may prove to be a partial remedy for numerous systemic difficulties that prevent the implementation of a stationary screening program.

5. Wstęp

Zaburzenia ze spektrum autyzmu (ang. autism spectrum disorders, ASD) są szeroką grupą zaburzeń neurorozwojowych, charakteryzujących się występowaniem trudności w zakresie:

- występowania sztywnych, repetetywnych oraz dysfunkcjonalnych zachowań o wzorcu typowym dla danej osoby,
- występowania specyficznych, zazwyczaj wąskich i bardzo nasilonych zainteresowań,
- integracji sensorycznej – nadwrażliwości lub niedoczulicy w zakresie dźwięków, dotyku, smaku lub zapachu,
- rozwoju mowy – zarówno braku funkcjonalnej mowy, stosowania języka własnego, obecności echolalii, opóźnienia rozwoju mowy i innych,
- relacji międzyludzkich i komunikacji społecznej – występowania trudności z empatyzowaniem, pozornego braku potrzeby wchodzenia w interakcje międzyludzkie, problemów z odczytywaniem sygnałów niewerbalnych,

a także wielu innych, mniej specyficznych objawów (tj. trudności z motoryką małą, przetwarzaniem informacji czy występowaniem zachowań autoagresywnych) [1]. Nie wszystkie wymienione wyżej objawy muszą występować, by możliwe było stwierdzenie u dziecka zaburzeń ze spektrum autyzmu; podobnie nasilenie tych objawów może się znacząco różnić u poszczególnych dzieci, niemniej składają się one na całość obrazu klinicznego. Objawy te zazwyczaj pojawiają się we wczesnym dzieciństwie, lecz w niektórych przypadkach stają się one wyraźne dopiero w momencie, kiedy dziecko nie jest w stanie sprostać rosnącym wymaganiom społecznym środowiska. Etiologia ASD nie jest znana – sugeruje się potencjalną wieloczynnikowość obejmującą czynniki genetyczne, infekcyjne, metaboliczne, związane z przebiegiem ciąży, a nawet socjalne [2–4].

ASD występują we wszystkich grupach socjoekonomicznych, etnicznych i rasowych, a częstość ich występowania w populacji nie jest dokładnie określona – szacunki wahają się, w zależności od przyjętej metodologii i badanej populacji [5]. Według Światowej Organizacji Zdrowia (WHO) częstość występowania ASD w populacji ogólnoświatowej wynosi 1:160 dzieci, jednakże nowe metaanalizy podają wartości wyższe, sięgające 1:100 [6, 7]. Raportowana częstość występowania ASD jest większa w krajach, w których metody diagnostyczne są łatwiej dostępne – przykładowo, w Stanach Zjednoczonych prewalencję określa się na poziomie 1:64 [8]. Dane dotyczące częstości występowania w Polsce są niepełne

– na podstawie informacji podanych przez Narodowy Fundusz Zdrowia oraz wybranych ośrodków edukacji specjalnej zebranych z województw zachodniopomorskiego i pomorskiego szacuje się ją na 1:286 dzieci [9].

Występowanie ASD wiąże się z dużym obciążeniem psychicznym, społecznym, finansowym i czasowym – zarówno dla osoby obciążonej ASD, jak i jego rodziny. Występowanie ASD zazwyczaj wiąże się z trudnościami w zakresie tworzenia i utrzymania relacji koleżeńskich i romantycznych, zdobycia odpowiedniej edukacji, koniecznością uczęszczania do placówek edukacji specjalnej, radykalnym zmniejszeniem jakości życia, większym ryzykiem zachorowania na choroby somatyczne (tj. otyłość, nadciśnienie tętnicze, cukrzyca, zespół jelita drażliwego), występowania zaburzeń psychicznych (tj. depresja, zespół nadpobudliwości psychoruchowej) czy trudnościami ze znalezieniem odpowiedniej pracy [10–14]. W przypadku rodzin z dzieckiem z ASD obserwowane są m.in. obniżenie jakości życia rodziców, zwiększenie odczuwalnego stresu rodzicielskiego, obniżenie jakości interakcji pomiędzy rodzicem a dzieckiem, obniżenie statusu majątkowego rodziny, a także większe obciążenie czasowe związane np. z koniecznością uczestniczenia w zajęciach dodatkowych, działaniach terapeutycznych czy wizytach lekarskich [15, 16]. Szacuje się, że występowanie u dziecka zaburzeń ze spektrum autyzmu ma większy negatywny wpływ na życie rodzin niż występowanie cukrzycy typu 1 czy zespołu Downa [17].

Wczesna diagnostyka zaburzeń ze spektrum autyzmu jest jedną z metod przeciwdziałania negatywnego wpływowi ASD na jakość życia osób dotkniętych ASD oraz ich rodzin. Wczesne postawienie diagnozy ASD umożliwia szybsze rozpoczęcie odpowiedniej, dopasowanej do potrzeb dziecka terapii i rehabilitacji, która ma za zadanie skompensować deficyty obserwowane u dziecka [18]. Im dziecko jest młodsze, tym można uzyskać lepsze efekty w obszarze komunikacji i interakcji społecznej, zdolności poznawczych, rozwoju mowy czy zachowania odpowiedniego do sytuacji [19, 20]. Wynika to przede wszystkim z większej podatności dopiero rozwijającego się układu nerwowego dziecka na stymulację z zewnątrz – we wczesnych latach życia łatwiej tworzone są nowe sieci nerwowe, a także sprawniej są one wzmacniane i integrowane z dotychczas istniejącymi pod wpływem stymulacji środowiskowej [21]. Pierwsze oznaki behawioralne ASD można wykryć u dziecka pomiędzy 6 a 14 miesiącem życia oraz wiarygodnie zdiagnozować w 24 miesiącu życia; niestety, z uwagi na liczne trudności związane z diagnozą ASD czas ten jest znacząco dłuższy – w danych dotyczących średniego wieku postawienia diagnozy ASD u dzieci określono go na 60,5 miesiąca (zakres w różnych krajach świata wynosił od 30,9 do 234,6 miesięcy) [22–24].

Największą trudnością w diagnostyce ASD jest wychwycenie zaburzeń we wczesnym etapie, kiedy zmiany w rozwoju są szybkie, a objawy – często bardzo subtelne [25]. Sytuacja, iż lekarze rodzinni lub pediatrzy pracujący w przychodniach podstawowej opieki zdrowotnej są specjalistami, którzy najczęściej obserwują dziecko w okresie niemowlęcym i wczesnym dzieciństwie (np. podczas bilansów, kwalifikacji do szczepień czy wizyt z powodu błahych infekcji) sprawia, że to oni powinni bacznie obserwować rozwój i zachowanie dziecka w okresie krytycznym dla rozpoznania ASD [26–28]. Wczesne wychwycenie niepokojących objawów przez lekarzy POZ – a następnie weryfikacja podejrzeń umożliwia szybsze postawienie diagnozy. W takim przypadku możliwe jest wcześniejsze rozpoczęcie zindywidualizowanej terapii, co znacząco poprawia funkcjonowanie dziecka względem rówieśników o podobnych trudnościach, u których diagnozę ASD postawiono później. Uważa się, że włączenie do procesu diagnostycznego lekarzy POZ na pierwszym etapie wykrywania zaburzeń ze spektrum jest najważniejszym pojedynczym czynnikiem wpływającym na zmniejszenie średniego wieku, w jakim stawiana jest diagnoza ASD. Wdrożenie badań populacyjnych w kierunku ASD w ramach podstawowej opieki zdrowotnej w Stanach Zjednoczonych pozwoliło na zwiększenie odsetka dzieci otrzymujących finalną diagnozę ASD przed 4 rokiem życia z 58% na 71% [8, 28, 29].

Potencjalnie idealną sytuacją z punktu widzenia pacjenta i jego rodziców jest przyjęcie przez lekarza POZ roli „stróża” – osoby, która jako pierwsza dostrzeże alarmujące objawy w zachowaniu dziecka oraz te zgłaszane przez rodziców, podejmie decyzję o konieczności uczestnictwa w dalszych konsultacjach specjalistycznych oraz będzie nadzorować proces diagnostyczny i terapeutyczny [30, 31]. W praktyce jednak opieka nad dzieckiem podejrzanym o występowanie ASD odbiega od ideału – zdarzają się sytuacje, iż lekarze marginalizują lub ignorują objawy zgłaszane przez rodziców lub nie zauważają subtelnych objawów zaburzeń rozwojowych [32]. Prawdopodobnie wynika to z przyczyn organizacyjnych w ramach POZ oraz trudności systemowych w zakresie dalszej opieki nad dzieckiem z grupy ryzyka – należą do nich m. in. ograniczony czas na przeprowadzenie wizyty lekarskiej, nadmierne obciążenie pracą czy trudność w dostępie do opieki specjalistycznej – psychologicznej czy psychiatrycznej [30, 33]. Ponadto, dostrzeżenie niektórych symptomów ASD wymaga – poza posiadaniem doświadczenia w zakresie diagnostyki i objawów zaburzeń rozwojowych – poświęcenia większej ilości czasu na danego pacjenta, niż jest to zwykle przewidziane w ramach jednej wizyty lekarskiej [34].

W celu zwiększenia skuteczności pracowników POZ w wykrywaniu wczesnych objawów zaburzeń ze spektrum autyzmu, opracowano liczne kwestionariusze służące do screeningu w kierunku ASD. Dowody z badań naukowych wskazują, iż włączenie takich narzędzi przesiewowych do rutynowych wizyt lekarskich (szczególnie wizyt bilansowych) może skutkować wcześniejszą i skuteczniejszą identyfikacją dzieci z grupy ryzyka, które potrzebują dalszej pomocy aniżeli opieranie się jedynie na doświadczeniu klinicznym i pobieżnej obserwacji zachowania dziecka [35, 36]. W związku z powyższym, od 2006 roku Amerykańska Akademia Pediatryczna (ang. *American Academy of Pediatrics*) zaleca rutynową diagnostykę w kierunku objawów ASD u wszystkich dzieci w wieku 18 i 24 miesięcy życia podczas wizyt bilansowych [27]. Pilotaże podobnych programów screeningowych uruchomiono m. in. w Szwecji, Islandii, Holandii i Hiszpanii [37–40].

Z drugiej strony, wykorzystanie kwestionariuszy do przesiewu w kierunku ASD w codziennej praktyce lekarskiej spotyka się z krytyką niektórych środowisk (np. w ramach raportu organizacji rządowej *US Preventive Services Task Force*) – podnoszony jest temat wysokich kosztów związanych z organizacją screeningu, ograniczeniami czasowymi oraz niskimi właściwościami psychometrycznych testów, zwłaszcza u bardzo małych dzieci (tj. poniżej 12 miesiąca życia) [41–44]. Niemniej, wydaje się, iż krytyka idei badań przesiewowych w kierunku ASD wynika z trudności w dostępności do narzędzi przesiewowych, ich niepełnej lub niepoprawnej walidacji czy braku jasnych wskazówek co do dalszego postępowania z dzieckiem z grupy ryzyka [27]. Kolejnym problemem jest fakt, iż pozytywnych doświadczeń ze Stanów Zjednoczonych nie można przenieść bezpośrednio do innych krajów. Z uwagi na kwestie badanych zmiennych (np. odpowiedniego zachowania, komunikacji czy zainteresowań) konieczne jest uwzględnienie kontekstu kulturowego i językowego, a nawet lokalnego postrzegania pojęcia niepełnosprawności, która wiąże się z ww. trudnościami. Przykładowo, odsetek rodziców zgłaszających, że ich dziecko wykazuje opóźnienia w rozwoju w porównaniu z rówieśnikami, jest znacznie wyższy na Jamajce niż w Bangladeszu czy Pakistanie [45]. Różnica wynikała jedynie z różnicy postrzegania pytania dotyczącego „powolności" dziecka – rozumianego na Jamajce jako bycie „wolniejszym" w sportach biegowych od innych rówieśników, które są popularne w tym kraju, a nie „powolnego rozumienia" czy „spowolnienia procesów myślowych". Stąd stosowanie nieprzystosowanych narzędzi poddanych jedynie prostej translacji może okazać się niewłaściwe [5].

Krytycy wdrożenia kwestionariuszy przesiewowych jako pierwszego etapu procesu diagnostycznego w kierunku ASD często powołują się na obserwowany fakt, iż dziewczynki

potencjalnie rzadziej otrzymują pozytywny wynik screeningu. Uważa się, iż przyczyną takiego stanu jest niedopasowanie płciowe narzędzi diagnostycznych – zawarte w nich pytania dotyczą objawów, które w opinii publicznej są mocno związane z ASD, i które zazwyczaj są mocniej wyrażone u chłopców (np. trudności w komunikacji interpersonalnej) [46]. Doniesienia te nie zostały jednak potwierdzone w badaniach naukowych, a zauważalny efekt nadreprezentacji wśród chłopców wynikał przede wszystkim z częstszego uczestnictwa w screeningu dzieci płci męskiej oraz mniejszego odsetka dziewczynek z grupy wysokiego ryzyka ASD, które finalnie otrzymywały diagnozę ASD [47–49]. Niemniej, z uwagi na potencjalne niedoszacowanie ryzyka występowania ASD u dziewczynek, pełna walidacja winna obejmować również analizy międzypłciowe.

Z uwagi na brak dostępnych w Polsce w pełni zwalidowanych narzędzi diagnostycznych do badań przesiewowych w kierunku ASD, celem pracy była pełna walidacja oraz przystosowanie do warunków polskich kwestionariusza Communication and Symbolic Behaviour-Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC) autorstwa A. Wetherby oraz B. Priznanta. Narzędzie jest wypełniane przez rodziców lub opiekunów dziecka i w wersji oryginalnej cechuje się wysokimi wartościami czułości oraz swoistości [50]. CSBS-DP ITC jest jednym z kwestionariuszy zalecanych przez Amerykańską Akademię Pediatryczną do użytku w celach badań przesiewowych [27].

W okresie realizacji niniejszego badania pandemia COVID-19 rozprzestrzeniła się na cały świat, co uniemożliwiło sprawną realizację badania stacjonarnego w przychodniach podstawowej opieki zdrowotnej. Pandemia wymusiła konieczność wprowadzenia rozwiązań, które umożliwiłyby dostęp do zdalnych usług medycznych, co miało zmniejszyć ryzyko zakażenia wirusem SARS-CoV-2. Z tego powodu konieczne było stworzenie elektronicznej wersji kwestionariusza CSBS-DP ITC do wypełnienia przez rodziców lub opiekunów w domu. Sytuacja epidemiologiczna wymusiła przeprowadzenie badania przesiewowego z wykorzystaniem wersji on-line, bez bezpośredniego kontaktu z osobą badaną. Zastosowanie metod używanych w telemedycynie w prowadzonym badaniu umożliwiło kontakt z pacjentem, wykonanie badań przesiewowych, kontrolnych, a w wielu przypadkach rozwianie wątpliwości i pytań rodziców. Taka forma prowadzenia badania umożliwiała szerszy dostęp do programu screeningowego oraz specjalistów z dziedziny zdrowia psychicznego mającego doświadczenie w zakresie zaburzeń rozwojowych. Pomimo wszystkich opisanych powyżej zalet badań profilaktycznych online, prowadzenie takich działań z wykorzystaniem technologii internetowych nie jest jeszcze popularne [51, 52]. Z uwagi na tę wymuszoną sytuacją

epidemiologiczną nową metodę prowadzenia badań przesiewowych w kierunku ASD i brak dotychczasowych badań w tym zakresie, oceniono potencjalne korzyści i trudności płynące z prowadzenia screeningu on-line.

6. Założenia i cele pracy

Główne cele pracy to:

1. Analiza dostępności narzędzi do przesiewowej diagnostyki zaburzeń ze spektrum autyzmu z uwzględnieniem ich właściwości psychometrycznych i diagnostycznych
2. Ocena wartości psychometrycznych (rzetelność, trafność predykcyjna, trafność konwergentna, stabilność wyniku) polskiej wersji kwestionariusza CSBS-DP ITC.
3. Ocena wartości diagnostycznych (czułość, swoistość, wartość predykcyjna dodatnia, ujemna, skuteczność) polskiej wersji kwestionariusza CSBS-DP ITC.
4. Ocena przydatności i efektywności screeningu ASD z użyciem technologii online.
5. Analiza potencjalnych trudności we wdrożeniu metod przesiewowych w kierunku ASD w codziennej praktyce lekarskiej w Polsce.

Dodatkowe cele badawcze to:

1. Określenie punktów odcięcia dla poszczególnych badanych grup wiekowych dla polskiej wersji kwestionariusza CSBS-DP ITC.
2. Ocena potencjalnych różnic międzypłciowych w zakresie osiąganych wyników w polskiej wersji kwestionariusza CSBS-DP ITC.

7. Materiał i metody pracy

7.1. Metodologia

W pierwszej części rozprawy - badaniu typu *scoping review* – użyto podejścia wg Arksey i O'Malley opierającego się na określeniu problemu badawczego, identyfikacji odpowiednich prac naukowych, selekcji badań, zestawieniu zebranych z nich danych oraz finalnie – podsumowaniu i raportowaniu wyników z uwzględnieniem wytycznych PRISMA (ang. *Preferred Reporting Items for Systematic reviews and Meta-Analyses*) dla artykułów typu *scoping review* (PRISMA-ScR) [53, 54]. W tym celu przeszukano cztery bazy naukowe (PubMED, EBSCO, Scopus, Web of Science) celem znalezienia publikacji odnoszących się do wczesnej diagnostyki i badań przesiewowych w kierunku ASD. Kryteriami włączenia były: 1) tematyka prac dotycząca narzędzi diagnostycznych do użytku w grupie dzieci od 0 do 3 lat życia, 2) dostępność informacji nt. danego narzędzia w języku angielskim oraz 3) możliwość zastosowania danego narzędzia w celu szybkiej oceny rozwoju dziecka w ramach badania przesiewowego. Kryteriami wyłączenia były: 1) tematyka prac dotycząca narzędzi do diagnostyki formalnej (pełnej) lub wymagających uczestnictwa w dodatkowym szkoleniu z uwagi na ich zawiłości psychometryczne, 2) tematyka prac skupiona na narzędziach diagnostycznych dla dzieci powyżej trzeciego roku życia, 3) prace dotyczące narzędzi screeningowych dotyczących innych zaburzeń rozwojowych niż ASD.

Uczestnikami głównej części badania były dzieci w wieku od 6 do 24 miesięcy oraz ich rodzice lub opiekunowie, mieszkający w Polsce oraz używający języka polskiego jako dominującego języka, którzy wyrazili poinformowaną zgodę na udział w badaniu. Z uwagi na wpływ pandemii COVID-19 i trudności w przeprowadzeniu badania w formie stacjonarnej, przygotowano stronę internetową projektu (dostępną pod adresem www.spojrzecwoczy.pl), gdzie umieszczono elektroniczne wersje kwestionariuszy wykorzystanych w badaniu, informacje o badaniu oraz dodatkowe informacje dla rodziców nt. ASD. Informacje na temat projektu udostępniono za pomocą mediów społecznościowych oraz kanałów Polskiego Towarzystwa Medycyny Rodzinnej. Jedną z części badania przeprowadzono stacjonarnie, we współpracujących żłobkach zlokalizowanych we Wrocławiu

W przypadku części rozprawy, dotyczącej oceny badań przesiewowych przeprowadzonych on-line w kierunku ASD oraz oceny ewentualnych trudności we wdrożeniu takich badań do codziennej praktyki lekarskiej uczestnikami byli rodzice biorący udział w głównej części badania oraz lekarze podstawowej opieki zdrowotnej (N = 95 osób).

Główna część badania była badaniem podłużnym, składającym się z kilku etapów, opisanych poniżej.

7.1.1. Badanie przesiewowe dzieci

W pierwszym etapie badania wykorzystano kwestionariusz osobowy oraz polską wersję kwestionariusza CSBS-DP ITC celem wykonania pomiarów rzetelności narzędzia, ustalenia wyników uzyskanych przez całą populację badanych (grupa normalizacyjna). Do tego etapu zakwalifikowano 1461 dzieci (625 dziewczynek, 42,78%) oraz ich rodziców lub opiekunów. W badaniu wzięło udział 1449 matek, 7 ojców i 5 innych członków rodziny.

7.1.2. Badanie kontrolne

Po trzech miesiącach od udziału w etapie pierwszym zaproszono rodziców, których dzieci w momencie badania przesiewowego miały od 6 do 21 miesięcy do ponownego badania za pomocą kwestionariusza CSBS-DP ITC (wg założeń kwestionariusza CSBS-DP ITC za jego pomocą nie można badać dzieci powyżej 24 miesiąca życia). Celem etapu była ocena trafności *test-retest* badającej stabilność pomiaru w czasie. W tym etapie wzięło udział 490 dzieci z 1313 zakwalifikowanych dzieci.

7.1.3. Pomiar rater-interrater

Celem oceny trafności *rater-interrater* (mierzącej zbieżność wyników uzyskanych w kwestionariuszu przez dane dziecko w przypadku użycia go przez różnych oceniających – w tym przypadku obu rodziców dziecka) 112 par rodziców wypełniło osobno kwestionariusz CSBS-DP ITC dla jednego ze swoich dzieci (tego samego), spełniającego kryteria włączenia do całego badania. Tę część badania przeprowadzono stacjonarnie, wśród rodziców dzieci, których dzieci należały do wrocławskich żłobków.

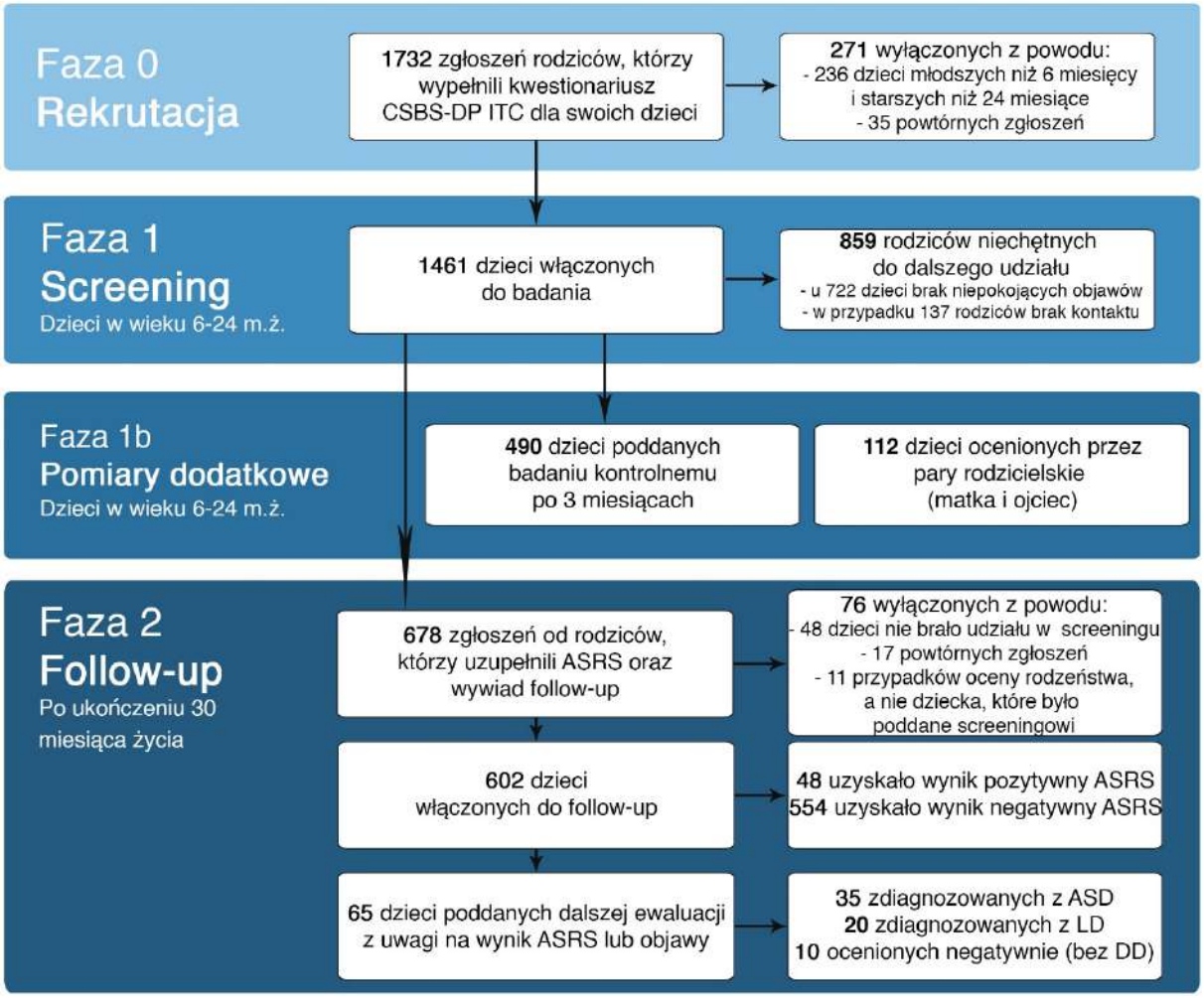
7.1.4. Badanie z użyciem kwestionariusza Autism Spectrum Rating Scales i ocena końcowa funkcjonowania dziecka

W momencie ukończenia przez dziecko 30 miesiąca życia rodzice zostali zaproszeni do udziału w ostatniej części badania, podczas której wypełniali oni polską wersję kwestionariusza Autism Spectrum Rating Scales (ASRS), a także przekazywali informację w zakresie ewentualnych uwag co do rozwoju dziecka oraz otrzymania diagnozy ASD lub innych zaburzeń rozwojowych. W przypadku pozytywnego wyniku w ASRS lub dalszego występowania uwag co do rozwoju dziecka u rodzica, rodzic wraz z dzieckiem byli zachęcani do udziału stacjonarnego w badaniu z użyciem baterii diagnostycznej Autism Diagnostic Observation

Schedule-2nd Version (ADOS-2) w Cieszynie (gdzie znajdował się ośrodek diagnostyczny, którego pracownicy współpracowali przy badaniu) lub wykonaniu tego badania w miejscu zamieszkania celem uzyskania finalnej diagnozy ASD. Finalnie, z rodzicem każdego dziecka kontaktowano się ponownie, by uzyskać ostateczną odpowiedź na temat potwierdzenia lub wykluczenia diagnozy ASD u dziecka za pomocą protokołu ADOS-2 lub nozologicznej diagnozy psychiatrycznej (punkty końcowe badania).

Pełen przebieg badania przedstawia Ryc. 1.

Rycina 1. Diagram faz projektu badawczego.



7.1.5. Ewaluacja udziału w badaniu przesiewowym prowadzonym on-line

Rodziców, którzy brali udział w badaniu z użyciem strony projektu „Spojrzeć w oczy" zaproszono do wypełnienia autorskiej ankiety mającej na celu ocenę ich satysfakcji i odczuć względem prowadzenia screeningu online. W tej części badania, trwającej od 9 do 18 stycznia 2023 r. wzięło udział 418 rodziców (odsetek zgłoszeń = 28,6%).

7.1.6. Ocena potencjalnych barier we wprowadzeniu narzędzi przesiewowych do codziennej praktyki lekarskiej oraz ewaluacja opinii i działań w zakresie wczesnej diagnostyki zaburzeń rozwojowych wśród lekarzy POZ

Celem uzyskania opinii specjalistów nt. potencjalnych przeszkód utrudniających wdrożenie programów przesiewowych w kierunku ASD w codziennej praktyce lekarzy w Polsce przesłano elektroniczne zaproszenia do udziału w badaniu do lekarzy zrzeszonych w Polskim Towarzystwie Medycyny Rodzinnej oraz na grupie na portalu Facebook zrzeszającej młodych lekarzy rodzinnych. Lekarze zostali zweryfikowani za pomocą numeru Prawa Wykonywania Zawodu. Zgłoszenia (N = 95) były zbierane od 31 czerwca 2023 do 1 lutego 2024 roku.

7.2. Narzędzia diagnostyczne

7.2.1. Metryka uczestnika badania

Metryka wypełniana przez rodziców biorących udział w badaniu składała się z pytań demograficznych (dotyczących płci badanego dziecka, stosunku osoby wypełniającej do dziecka, miejsca zamieszkania) oraz dotyczących historii chorobowej dziecka i jego rodziny celem oceny czynników ryzyka występowania ASD (zdiagnozowanych chorób genetycznych, problemów ze słuchem, wzrokiem, rozwojem ruchowym lub innych problemów zdrowotnych, kwestii wycofania się dziecka ze zdobytych umiejętności lub diagnozy ASD lub innych zaburzeń rozwojowych w rodzinie.

7.2.2. Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist

Kwestionariusz Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist (CSBS-DP ITC) został stworzony w 2002 roku przez Amy Wetherby oraz Barry'ego Priznanta jako narzędzie do przesiewowego wykrywania zaburzeń ze spektrum autyzmu u dzieci w wieku od 6 do 24 miesięcy [55]. Kwestionariusz jest wypełniany przez rodziców lub opiekunów dziecka. Składa się z 24 pytań, które dotyczą siedmiu predyktorów rozwoju dziecka (emocje i kontakt wzrokowy, komunikacja, gesty, dźwięki, słowa, rozumienie, użycie przedmiotów), tworzących łącznie trzy podskale (Umiejętności społeczne, Umiejętności symboliczne oraz Mowa). Analiza wyniku kwestionariusza opiera się na sumarycznej wartości punktowej, przy czym im wyższy wynik, tym na wyższym poziomie oceniany jest rozwój dziecka (mniejsze jest ryzyko zaburzeń ze spektrum autyzmu). W wersji oryginalnej przygotowano progi odcięcia dla każdego miesięcznego zakresu wiekowego jako 1.25 odchylenia standardowego od wyniku w populacji normalizacyjnej. Maksymalny wynik wynosi 57 punktów, a analizie podlega nie tylko wynik ogólny, ale również wynik w podskalach, które mogą stanowić informację dla terapeutów odpowiedzialnych za realizację wczesnych interwencji u dziecka podejrzanego o ASD, ale również być przesłanką do wdrożenia diagnostyki różnicowej (np. izolowane obniżenie wyniku w podskali Mowa może sugerować konstytucyjne opóźnienie rozwoju mowy). W wersji oryginalnej (amerykańskiej) cechuje się czułością i swoistością rzędu 0,89, pozytywną wartością predykcyjną równą 0,94 oraz negatywną wartością predykcyjna rzędu 0,80. Rzetelność narzędzia mierzona za pomocą alfy Cronbacha wynosi 0,87.

Polska adaptacja CSBS-DP ITC przygotowana w ramach badania została zaadaptowana do polskich warunków – zarówno kulturowo, jak i lingwistycznie. Ze względu na znaczne różnice w budowie fonemicznej języka polskiego (będącego językiem z grupy języków słowiańskich) i angielskiego (języka z grupy języków germańskich) zdecydowano, po konsultacji z logopedami, o zmianie brzmienia dwóch pytań – w pytaniu 15 usunięto jeden z fonemów łączonych („*uh oh”*), a „*bye bye*” zmieniono na polską wersję pożegnania tj. „*pa pa*”. Podobnie w pytaniu 16 zamieniono wymawiane głoski na te, których polskie dzieci uczą się jako pierwsze (tj. „*ma, na, ba, da, ga, ka, la, ja, ta, pa*”). Zwroty występujące w pytaniach 17 i 18 dostosowano do najczęściej spotykanych wśród polskich dzieci [56]. Pozostałe pytania zostały przetłumaczone możliwie najwierniej, dostosowując ich składnię do gramatyki języka polskiego. Eksperci oceniający rozwój dzieci zostali poproszeni o ocenę zrozumiałości pytań w języku polskim i nie zgłosili żadnych zastrzeżeń.

7.2.3. Autism Spectrum Rating Scales

Zestaw kwestionariuszy Autism Spectrum Rating Scales (ASRS) to narzędzie poziomu drugiego (tj. pomocnicze w procesie diagnostycznym lub zaawansowane narzędzie przesiewowe lub monitorujące postępy terapeutyczne) do diagnostyki zaburzeń ze spektrum autyzmu [57]. Zestaw składa się z pełnych i skróconych wersji testów dla dzieci młodszych (2–5 lat) oraz starszych dzieci i młodzieży (6–18 lat), zarówno dla rodziców, jak i nauczycieli. ASRS został opracowany w 2013 roku, a jego struktura odpowiada kryteriom diagnostycznym Podręcznika Diagnostycznego i Statystycznego Zaburzeń Psychicznych (wydanie czwarte, zrewidowane) (ang. Diagnostic and Statistical Manual of Mental Disorders, 4th Version, Text Revised, DSM-IV-TR) oraz Międzynarodowej Statystycznej Klasyfikacji Chorób i Problemów Zdrowotnych, wydanie 10 (ang. *International Statistical Classification of Diseases and Related Health Problems 10th Revision*, ICD-10) Polska wersja została opracowana w 2016 roku i charakteryzuje się dużą wiarygodnością w wersji dla rodziców oraz ma potwierdzoną trafność dyskryminacyjną, zbieżną i różnicującą, a ponadto jest zbieżna z kryteriami dla ASD zawartymi w nowej wersji podręcznika DSM-5 [58]. W badaniu zastosowano skróconą wersję dla dzieci młodszych, zawierającą 15 pozycji, które najlepiej różnicują dzieci ze zdiagnozowanym ASD od grupy porównawczej z populacji niewykazującej cech ASD. Analiza funkcji dyskryminacyjnej dla wyników surowych wykazała, że wskaźniki prawidłowej klasyfikacji wahały się od 88,2% do 91,4%. Alfa Cronbacha dla zastosowanej w badaniu wersji ASRS wynosi 0,85. Czułość i swoistość wynoszą odpowiednio 87,8% i 83,5%.

7.2.4. Autism Diagnostic Observation Schedule – Wersja Druga

Autism Diagnostic Observation Schedule – 2nd Version (ADOS-2) to częściowo ustrukturyzowany, wystandaryzowany protokół obserwacji wykorzystywany do diagnostyki osób z ASD i w połączeniu z oceną kliniczną dziecka przez specjalistę i wywiadem z opiekunem stanowi tzw. „*złoty standard*" w zakresie stawiania diagnozy ASD [59]. Obejmuje on zestaw prób prowokujących osobę badaną do uczestnictwa w określonych zachowaniach społecznych, co umożliwia obserwowanie jej w trakcie naturalnie przebiegającej interakcji z badającym. W przypadku dzieci powyżej 31 miesiąca życia pozwala określić, czy zachowanie badanego wskazuje na ASD; w przypadku dzieci młodszych – określa stopień, w jakim zachowanie dziecka budzi niepokój. Wykorzystanie zestawu ADOS-2 wymaga ukończenia studiów psychologicznych lub dodatkowego szkolenia. Badanie z użyciem ADOS-2 trwa ok. 40-60 minut. Badania prowadzone w ramach polskiej standaryzacji ADOS-2 wykazały dobrą rzetelność narzędzia, ocenianą na podstawie zgodności ocen sędziów, stabilności i zgodności wewnętrznej [60]. Ponadto polska wersja ADOS-2 cechuje się występowaniem wysokich wartości korelacji wyników z wynikami innych narzędzi do diagnozy autyzmu oraz istotnych i dużych różnic między grupą z rozpoznaniem autyzmu dziecięcego lub innych zaburzeń ze spektrum autyzmu a grupą kontrolną, obejmującą osoby z zaburzeniami spoza spektrum autyzmu oraz osoby rozwijające się typowo.

W przypadku konieczności przeprowadzenia badania ADOS u uczestników badania, przeprowadzającym badanie była współpracująca podczas projektu psycholog lub – w przypadku chęci rodziców do wykonania badania w okolicy miejsca zamieszkania – psycholog lub certyfikowany pedagog. W takim przypadku, rodzice zostali poproszeni o przekazanie informacji nt. osoby wykonującej badanie celem weryfikacji posiadanych uprawień do wykonania takiego badania.

7.2.5. Kwestionariusze oceniające screening on-line ASD przez rodziców oraz screening on-line ASD i potencjalne bariery we wdrożeniu screeningu ASD przez lekarzy

Celem zebrania opinii rodziców i lekarzy na temat screeningu on-line w kierunku ASD oraz opinii lekarzy nt. potencjalnych barier we wdrożeniu programu powszechnego screeningu przygotowano osobne autorskie kwestionariusze – dla rodziców i dla lekarzy. Rodzice zostali poproszeni o wypełnienie ankiety składającej się z dziewięciu pytań metrykalnych – dotyczących wieku, płci, miejsca zamieszkania, wykształcenia, subiektywnej oceny umiejętności korzystania z technologii informatycznych (w dziesięciopunktowej skali Likerta)

oraz dotychczasowego udziału w badaniu oraz dziesięciu pytań dotyczących zadowolenia z udziału w badaniu przesiewowym on-line (w pięciopunktowej skali Likerta) oraz chęci udziału i zasadności wdrożenia populacyjnego programu screeningowego w kierunku ASD. Rodzice byli pytani również o preferencje dotyczące metody badań przesiewowych – on-line lub stacjonarnie.

Ankieta dla lekarzy składała się z siedmiu pytań metrykalnych (wiek, płeć, główne miejsce praktykowania zawodu, subiektywna ocena umiejętności korzystania z technologii informatycznych, odsetek pacjentów pediatrycznych wśród wszystkich pacjentów, ilość pacjentów pediatrycznych z zaburzeniami psychicznymi lub rozwojowymi pod opieką) oraz dziesięciu pytań dotyczących potencjalnych ograniczeń screeningu on-line, zalet lub wad screeningu on-line, chęci do wdrożenia screeningu w swojej codziennej praktyce, a także dotychczasowych działań w celu diagnozowania i dalszej ewaluacji zaburzeń rozwojowych u swoich pacjentów. Obie ankiety stanowią załącznik do niniejszej rozprawy doktorskiej.

7.3. Analiza statystyczna

W pierwszej publikacji użyto metod jakościowych zgodnie z wytycznych PRISMA (ang. Preferred Reporting Items for Systematic reviews and Meta-Analyses) dla artykułów typu scoping review (PRISMA-ScR) [54].

We wszystkich publikacjach w celu oceny normalności rozkładu zmiennych użyto testów Kołomogorowa-Smirnowa oraz Shapiro-Wilka. W drugiej i trzeciej publikacji z uwagi na niewielką ilość badanych w poszczególnych miesiącach życia zostali oni złączeni w większe grupy wiekowe zgodnie z założeniami teorii koncepcji psychomotorycznej rozwoju intelektualnego wg Jeana Piageta z późniejszymi zmianami [61].

W drugiej publikacji do analiz statystycznych wykorzystano oprogramowanie JASP 0.1.11. Wykonano testy *chi-kwadrat* oraz konfirmacyjną analizę czynnikową (ang. confirmatory factor analyses, CFAs) dla kwestionariusza za pomocą dopasowania modeli jedno- i trzyczynnikowych (czyli dla wyniku ogólnego i podskal kwestionariusza CSBS-DP ITC) z użyciem metody estymacji DWLS (ang. *diagonally weighted least squares*) z uwagi na jej przydatność dla zmiennych porządkowych w przypadku braku rozkładu normalnego [62, 63]. Wykładnikami dopasowania modelu były porównawczy wskaźnik dopasowania (ang. *comparative fit index*, CFI), będący wskaźnikiem badającym rozbieżności między danymi a hipotetycznym idealnym modelem, wskaźnik Tuckera-Lewisa (ang. *Tucker-Lewis Index*, TLI), będący wskaźnikiem rozbieżności między wartością χ^2 hipotetycznego modelu, a χ^2 wartością

modelu badanego (oba im bliżej 1, tym lepsze dopasowanie modelu) oraz pierwiastek ze średniego błędu aproksymacji próby (ang. *root mean square error of approximation,* RMSEA) będący miarą szacowanej rozbieżności między populacją a macierzą kowariancji populacji implikowaną z idealnego modelu na każdy stopień swobody; w tym przypadku im bliżej 0, tym lepsze dopasowanie modelu) [64]. Tożsamych analiz dokonano dla obu podgrup płciowych celem oceny równoważności pomiaru.

W celu oceny rzetelności narzędzia, obliczono wartości współczynników α Cronbacha (będącego stosunkiem wariancji dla wszystkich pozycji testowych lub inaczej - sumą wszystkich testów połówkowych) oraz ω McDonalda (będący współczynnikiem badającym również wewnętrzną spójność testu i posiadającym przedziały ufności). Do celów analizy zmienności wyniku w czasie (tzw. rzetelności *test-retest*) oraz w zależności od oceniającego (tzw. stabilność *interrater*) użyto analizy korelacji Pearsona oraz analizy współczynników korelacji wewnątrzklasowych – z uwagi na charakter zmiennych zastosowano wariant 3 (typ B) tej metody (ang. *intraclass coefficient* ICC,3(B)). Ponadto, porównano średnie wyniki osiągane w podgrupach płciowych celem analizy trafności, co zostało umotywowane potencjalną "stronniczością diagnostyczną" na korzyść płci męskiej. Wcześniejsze badania wskazywały, iż dziewczynki rzadziej otrzymywały pozytywne wyniki badań przesiewowych w kierunku ASD z uwagi na prawdopodobne „mniej wyrażone" objawy niż u chłopców. Doniesienia te nie zostały jednak wiarygodnie potwierdzone w badaniach naukowych [46–48]. Niemniej, u dziewczynek z wysokim prawdopodobieństwem ASD rzadziej diagnozowano te zaburzenia, co zmniejszało dokładność narzędzi i prowadziło do opóźnień w diagnozowaniu tej grupy [49]. W celu porównań między polską a amerykańską (oryginalną) grupą normalizacyjną z uwagi na brak dostępnych danych użyto porównania median za pomocą metody testu Wilcoxona z jednej próby.

W trzeciej publikacji dokonano analizy korelacji pomiędzy wynikami uzyskanymi przez badanych w kwestionariuszach ASRS oraz CSBS-DP ITC za pomocą metody korelacji rang Spearmana. Celem oceny zdolności różnicującej oraz określenia punktów odcięcia dla CSBS-DP ITC użyto analizy krzywej charakterystyki operacyjnej odbiornika (ang. *receiver operating characteristic,* ROC). W tej analizie wykorzystano dwie metody. Pierwszą z nich jest metoda Youdena (z określeniem punktów odcięcia dla tej wartości punktowej, która cechuje się największą efektywnością, tj. pozwala odnaleźć punkt krzywej ROC, który maksymalizuje funkcję:

$$J(x) = Se(x) – [1-(Sp(x)]$$

gdzie J jest punktem na krzywej tzw. najlepszego możliwego klasyfikatora zmiennej, Se(x) jest czułością przy progu x, a Sp(x) jest specyficznością przy progu x.

Drugą metodą jest metoda minimalizacji potencjalnych kosztów z użyciem stycznej, w której przyjęto arbitralne uwzględnienie tzw. "kosztu podjęcia błędnej klasyfikacji" przy konstruowaniu stycznych określających stosunki czułości do swoistości w przypadku poszczególnych punktów odcięcia. Zostało to umotywowane faktem, że zaklasyfikowanie dziecka z ASD jako osoby zdrowej wiąże się z wyższymi kosztami (w zakresie opóźnienia leczenia) niż błąd odwrotny [65].

W czwartej publikacji do analiz użyto nieparametrycznych testów Kruskala-Wallisa (w przypadku zmiennych ilościowych) lub chi kwadrat (w przypadku zmiennych jakościowych lub przyjmujących dwie wartości). W przypadku oceny korelacji między zmiennymi użyto testu korelacji rang Spearmana. W przypadku niewielkiej liczby respondentów w podgrupach przy użyciu testu chi kwadrat zastosowano poprawkę Yatesa. W przypadku części badanych cech możliwa była jedynie interpretacja opisowa z uwagi na brak możliwości sprawdzenia wartości zmiennych pod kątem homogeniczności – badani mogli wskazać więcej odpowiedzi w zakresie jednej badanej zmiennej (np. w przypadku określenia potencjalnych barier we wprowadzeniu populacyjnego screeningu ASD).

W publikacjach trzeciej i czwartej do analiz użyto oprogramowania Statistica 13.3 oraz pakietu statystycznego R. Dla wszystkich testów we wszystkich publikacjach przyjęto poziom istotności $p < 0,05$.

8. Wyniki

I. Sobieski M., Sobieska A., Sekułowicz M., Bujnowska-Fedak M.M., **Tools for early screening of autism spectrum disorders in primary health care – a scoping review.** *BMC Primary Care* 23, 46 (2022)

W pierwszej pracy z cyklu, będącej pracą typu *scoping review* dotyczącej dostępności kwestionariuszy do wczesnej diagnostyki zaburzeń ze spektrum autyzmu, użycie zapytania w czterech bazach prac naukowych wygenerowało łącznie 330225 rezultatów, z których aż 227371 było duplikatami. Po pierwszym przeglądzie tytułów i abstraktów wybrano 154 manuskrypty do dalszej analizy, spośród których ostatecznie 81 spełniło kryteria włączenia. Trzy dodatkowe źródła zostały dodane spoza przeszukiwanych baz danych.

Wyekstrahowane publikacje w znacznej większości były przeprowadzone na grupach badawczych pochodzących ze Stanów Zjednoczonych (N = 18), Australii (N = 5) oraz Korei Południowej (N = 4). Wyekstrahowano 26 badań naukowych nad indywidualnie zaprojektowanymi narzędziami (tzw. wersjami oryginalnymi kwestionariuszy), z czego 35% pochodziło z USA, a ponad połowa została przygotowana w języku angielskim. W przypadku adaptacji kwestionariuszy przesiewowych najpopularniejszym narzędziem na świecie jest *Modified-Checklist for Autism in Toddlers* (dostępny w 35 różnych językach; w kilku przypadkach dostosowany dialektycznie np. do dialektu języka hiszpańskiego używanego w Argentynie). Większość adaptacji to jednak bezpośrednie tłumaczenia językowe bez adaptacji kulturowej o zmniejszonych wartościach diagnostycznych tych kwestionariuszy względem wersji oryginalnych.

Dane dotyczące rzetelności narzędzi zostały opisane w 46 pracach, czułości – w 53, swoistości w 51, pozytywnej wartości predykcyjnej (ang. *positive predictive value*, PPV) w 47, a negatywnej wartości predykcyjnej (ang. *negative predictive value*, NPV) w 36. Wartości te znacząco różniły się w zależności od badania i użytych kwestionariuszy. Pomiary rzetelności w przypadku poszczególnych narzędzi diagnostycznych oraz ich adaptacji wahały się od 0,53 do 1,00; czułości – od 0,18 do 1,00, swoistości od 0,51 do 1,00, PPV od 0,01 do 1,00, NPV od 0,48 do 1,00.

Jedynie w 20 adaptacjach lub badaniach oryginalnych opisano wszystkie poszukiwane wartości psychometryczne. Badane populacje zazwyczaj dotyczyły populacji ogólnej (N = 46), w ośmiu – wykorzystano jedynie dzieci z grupy ryzyka. Ilość badanych wahała się od 13 do

52026 dzieci; przy czym w 34 badaniach wykorzystano próbkę powyżej 1000 dzieci, a w 6 – powyżej 10000 dzieci.

II. Sobieski M., Wrona S., Flakus M., Pierchała K., Sobieska A., Podgórska K., Wołowicz A., Sekułowicz M., Bujnowska-Fedak MM. **Reliability and validity of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist.** *Research in Autism Spectrum Disorders* 117, 102454 (2024)

W drugiej publikacji z cyklu sprawdzono wartości psychometryczne polskiej wersji kwestionariusza CSBS-DP ITC. Wykonana CFA dla dwóch modeli (jednoczynnikowego, dla wyniku ogólnego w kwestionariuszu CSBS-DP ITC oraz trójczynnikowego, dla podskal CSBS-DP ITC) wykazała dobre dopasowanie obu modeli. Lepsze dopasowanie uzyskał model jednoczynnikowy dla wyniku całkowitego (χ^2 = 1188.19, χ^2/df = 4,72, CFI = 0,98, TL = 0,98, RMSEA = 0,050, 90%CI RMSEA = 0,048-0,053, p < 0.001) niż trójczynnikowy (χ^2 = 973,72, χ^2/df = 3,91, CFI = 0,99, TL = 0,99, RMSEA = 0,045, 90%CI RMSEA = 0,042-0,048, p < 0.001).

Ponadto analiza ładunków poszczególnych czynników również wykazała statystyczną istotność wszystkich 24 badanych elementów w obu modelach (we wszystkich przypadkach p <0.001), a ich ładunki wynoszą od 0,146 do 0,894 w modelu jednoczynnikowym oraz od 0,152 do 0,926 w modelu trójczynnikowym.

Celem oceny równoważności pomiaru w podgrupach płciowych w teście replikacji dokonano czterech analiz – niezmienności konfiguracyjnej (dopasowania ogólnej struktury między podgrupami), metrycznej (równoważności wartości ładunków między podgrupami), skalarnej (równoważności przecięć wartości danych czynników między podgrupami) oraz ścisłej (równoważności ładunków, przecięć wartości danych czynników oraz wariancji poszczególnych czynników). Wszystkie analizy wykazały istnienie równoważności w obu modelach (wartości χ^2 = od 1041,23 do 1628,47, CFI = od 0,980 do 0,990, RMSEA = od <0.039 do 0.053, ΔRMSEA od <0,001 do 0,006, ΔCFI od <0,001 do 0,004). Niemniej, jedynie w przypadku modelu jednoczynnikowego można było założyć równoważność modelu we wszystkich badanych analizach (w przypadku równoważności metrycznej modelu trójczynnikowego ΔRMSEA wynosi 0,006).

Polska wersja CSBS-DP ITC cechuje się wysoką stabilnością wewnętrzną - określona wartością alfa Cronbacha wynosi 0,92 (95%CI = 0,912 – 0,924); podobną wartość przyjmuje omega McDonalda = 0,92. Średnia korelacja między poszczególnymi czynnikami wynosi r = 0,32, a między poszczególnym czynnikiem a wynikiem całkowitym = 0,54, co jest wartością satysfakcjonującą.

W analizie stabilności pomiaru metodą *test-retest* wskaźniki korelacji Pearsona pomiędzy pierwszym a drugim pomiarem (przeprowadzonym trzy miesiące później) przyjmują wartości w większości powyżej 0,70, co świadczy o wysokiej stabilności pomiaru (dla wyniku całkowitego r = 0,83, $p < 0,001$). Analiza korelacji wewnątrzklasowych stabilności pomiaru metodą *interrater* wskazuje na wysoki stopień stabilności między dwoma osobami oceniającymi (wartości ICC3,B dla podskali Umiejętności społeczne 0,916, 95%CI = 0,880-0,941, dla podskali Umiejętności symboliczne 0,969, 95%CI = 0,955-0,979, dla podskali Mowa 0,943, 95%CI = 0,918-0,960, dla wyniku całkowitego 0,968, 95%CI = 0,954-0,978.

Wykazano występowanie różnic międzypłciowych w zakresie uzyskiwanych wyników w CSBS-DP ITC. W najmłodszej grupie wiekowej nie wykazano istotnych różnic między podgrupami płciowymi. W grupie wiekowej dzieci w wieku 9-12 miesięcy dziewczynki uzyskiwały statystycznie istotnie wyższe wyniki w przypadku ocenianych predyktorów (gesty oraz wytwarzane dźwięki) i wyniku całkowitego. Wśród dzieci w wieku 13-18 miesięcy dziewczynki osiągały statystycznie istotnie wyższe wyniki we wszystkich ocenianych aspektach z wyjątkiem predyktorów emocje i komunikacja. Po 19 miesiącu życia, różnice statystyczne w zakresie podskali Umiejętności społeczne oraz predyktora użycie obiektów zanikają.

Ponadto porównano średnie wyniki osiągane w polskiej i amerykańskiej grupie walidacyjnej w grupach wiekowych od 12 do 24 miesiąca życia (dane dotyczące młodszych dzieci nie były dostępne). Polskie dzieci osiągnęły statystycznie istotnie niższe wyniki w 6 podgrupach wiekowych w zakresie podskali Umiejętności społeczne oraz w 7 podgrupach w podskali Mowa. Istotnie wyższe wyniki odnotowano wśród polskich dzieci w 11 kategoriach wiekowych w podskali Umiejętności symboliczne. Mediana wyniku całkowitego różniła się istotnie statystycznie jedynie w grupach wiekowych 14 i 16 miesięcy życia.

III. Sobieski M., Kopszak A., Wrona S., Bujnowska-Fedak M.M. **Screening accuracy and cut-offs of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist.** *PLOS One* 19(8): e0299618 (2024)

W trzeciej publikacji z cyklu dokonano sprawdzenia trafności predykcyjnej polskiej wersji kwestionariusza CSBS-DP ITC i ustalono najbardziej efektywne punkty odcięcia dla populacji generalnej polskich dzieci i niemowląt. Za pomocą analizy korelacji rang Spearmana dokonano pomiaru trafności konwergentnej (dokładności predykcyjnej) pomiędzy kwestionariuszami CSBS-DP ITC i ASRS. Wykazano słabe lub umiarkowane korelacje między wynikami obu kwestionariuszy oraz fakt, że siła tej korelacji rośnie wraz z wiekiem dziecka. Wyniki statystycznie istotne wykazano w grupach wiekowych 9-12 miesięcy (r = -0,280, p = 0,001), 13-18 miesięcy (r = -0,364, $p < 0,001$) oraz 19-24 miesięcy (r = -0,431, $p < 0,001$).

Z wykorzystaniem metody Youdena dokonano analizy ROC celem określenia punktów odcięcia w poszczególnych podgrupach wiekowych, a także czułości, swoistości, wartości predykcyjnej dodatniej (PPV), ujemnej (NPV), wskaźnika wiarygodności wyniku dodatniego (LR+) i ujemnego (LR-) oraz dokładności kwestionariusza CSBS-DP ITC. Analizy udało się przeprowadzić jedynie dla podgrup powyżej 9 miesiąca życia – w podgrupie dzieci w wieku od 6 do 8 miesięcy nie było wystarczająco dużo przypadków dzieci z ASD by dokonać tego typu analiz. Podobnie, nie udało się przeprowadzić tożsamych analiz dla podskal kwestionariusza. Pole pod krzywą dla badanych podgrup wynosiło od 0,782 do 0,856, p <0,001. Dokładne wyniki dla punktów odcięcia cechujących się najwyższymi wskaźnikami indeksu Youdena (wynoszącym odpowiednio w poszczególnych podgrupach 9-12 miesięcy 0,612, 13-18 miesięcy 0,604 oraz 19-24 miesięcy 0,604) zaprezentowano w Tabeli 1.

Tabela 1. Wyniki analizy ROC z użyciem metody Youdena dla polskiej wersji kwestionariusza CSBS-DP ITC.

Grupa wiekowa	Przyjęty punkt odcięcia	Czułość	Swoistość	Dokładność	PPV	NPV	LR(+)	LR(-)
9-12 miesięcy	21 pkt	0.750	0.862	0.855	0.261	0.981	5.426	0.290
13-18 miesięcy	36 pkt	0.750	0.854	0.847	0.267	0.980	5.136	0.293
19-24 miesięcy	39 pkt	0.667	0.939	0.923	0.400	0.979	10.889	0.355

Uwaga. LR(+) - iloraz wiarygodności wyniku dodatniego (ang. *likehood ratio positive*), LR(-) - iloraz wiarygodności wyniku ujemnego (ang. *likehood ratio negative*), NPV – negatywna wartość predykcyjna (ang. *negative predictive value*), PPV – pozytywna wartość predykcyjna (ang. *positive predictive value*)

Rezultaty analizy krzywej ROC z użyciem metody cięciw z założeniem minimalizacji oczekiwanych kosztów w przypadku kwestionariusza CSBS-DP ITC wskazują na niższą dokładność narzędzia w przypadku zastosowania takiego założenia; w przypadku przyjęcia prewalencji oszacowanej z próbki badanej, przy przyjętych wg tej metody punktów odcięcia czułość narzędzia wyniosłaby od 0.188 do 0.667 a swoistość – od 0.939 do 0.984. W związku z niską czułością tej metody – a co za tym idzie – dużą ilością dzieci wysyłaną nadmiarowo na dalszą ewaluację, jako zalecane punkty odcięcia przyjęto te uzyskane za pomocą analizy metodą Youdena.

IV. Sobieski, M.; Grata-Borkowska, U.; Bujnowska-Fedak, M.M. **Implementing an Early Detection Program for Autism Spectrum Disorders in the Polish Primary Healthcare Setting—Possible Obstacles and Experiences from Online ASD Screening.** *Brain Sciences* 14, 388 (2024)

Ostatnia publikacja z cyklu miała na celu ocenę potrzeby wprowadzenia badań przesiewowych w kierunku ASD, zalet i wad prowadzenia takiego programu za pomocą technologii teleinformatycznych, a także ustalenie potencjalnych przeszkód we wprowadzeniu screeningu ASD w ramach podstawowej opieki zdrowotnej. Praktycznie wszyscy rodzice (99,04%) są świadomi problematyki ASD, niemniej istnieje znacząca różnica w poziomie świadomości na temat ASD wśród rodziców z niższym wykształceniem, mieszkających na wsi i mających niższą biegłość w technologiach informatycznych w porównaniu do pozostałych rodziców (odpowiednio $\chi^2 = 13{,}013$, $p = 0{,}005$; $\chi^2 = 13{,}398$, $p = <0{,}001$; $H = 14{,}801$, $p = 0{,}011$, gdzie H – wartość statystyki H testu Kruskala-Wallisa). 56% rodziców przynajmniej rozważała występowanie u swoich dzieci jakichkolwiek zaburzeń rozwojowych. Intrygującą kwestią jest fakt, że rodzice, u których ostatecznie zdiagnozowano u dzieci zaburzenia rozwojowe, znacznie częściej niż pozostali zgłaszają, że pojawiała się u nich myśl, że ich dziecko może być zagrożone występowaniem ASD ($\chi^2 = 12{,}350$, $p = < 0{,}001$).

98,09% rodziców (N = 410) uważa, że badania przesiewowe w kierunku ASD powinny być obowiązkowe, 98,56% (N = 412) ponownie wzięłoby udział w badaniach przesiewowych w kierunku zaburzeń rozwojowych u swojego kolejnego dziecka, a 97,13% (N = 406) wzięłoby udział w badaniach przesiewowych online. Odsetek rodziców chcących ponownie uczestniczyć w programie dla kolejnego dziecka jest niższy w przypadku rodziców mieszkających w mniejszych miejscowościach. Zdania rodziców są mocno rozbieżne w przypadku preferencji dotyczących formy badania przesiewowego (online lub stacjonarnie) – niewielka większość preferuje wersję internetową (52,15%, N = 218), a jedynym czynnikiem, który statystycznie istotnie koreluje z preferencją screeningu online nad stacjonarnym jest wyższa subiektywna umiejętność obsługi technologii informatycznych przez rodzica ($H = 16{,}212$; $p = 0{,}006$).

Zebrane dane wskazują na bardzo wysoką ogólną ocenę screeningu elektronicznego przeprowadzonego w ramach projektu przez rodziców biorących udział w badaniu. Średnie ocen na poszczególne kwestie – dotyczące dostępności informacji, możliwości uzyskania pomocy, zadania dodatkowych pytań i ogólnego zadowolenia z udziału, wahają się od 3,877 do 4,925 w pięciopunktowej skali Likerta (1-5). Opinie, z kilkoma wyjątkami, charakteryzują się dużą jednorodnością pod względem badanych cech. Rodzice mieszkający w małych

miejscowościach najgorzej ocenili możliwość kontaktu i możliwość otrzymania pomocy (odpowiednio H = 10,794, p = 0,013 i H = 21,323, p = < 0,001). Osoby z wykształceniem wyższym niżej niż rodzice z wykształceniem średnim oceniały możliwość uzyskania odpowiedniej pomocy w ramach badania online (H = 6,043, p = 0,048). Ponadto istnieje słaba ujemna korelacja wskazująca, że starsi rodzice gorzej niż młodsi rodzice oceniają możliwość uzyskania dalszych informacji i odpowiedzi od badających podczas screeningu online.

Większość z badanych lekarzy wykorzystuje w swojej codziennej praktyce jedynie podstawowe metody wykrywania objawów zaburzeń rozwojowych - do najpopularniejszych zalicza się obserwację dziecka w trakcie badania przedmiotowego (94,74%), próbę porozumiewania się i nawiązania kontaktu z dzieckiem (93,68%) oraz ocenę tempa osiągania kamieni milowych (89,47%). Ustandaryzowane narzędzia diagnostyczne wypełniane przez opiekunów lub pracowników służby zdrowia są wykorzystywane znacznie rzadziej (odpowiednio 21,05 i 15,79% ankietowanych). Ponadto aktywne wykorzystanie powyższych metod częściej występuje jedynie u dzieci z podejrzeniem występowania zaburzeń rozwojowych; rzadziej w całej populacji pacjentów pediatrycznych.

Dość znaczna część ankietowanych lekarzy ma postawę wyczekującą w przypadku występowania subtelnych cech zaburzeń rozwojowych u dziecka do drugiego roku życia (37,89%); w przypadku dzieci powyżej drugiego roku życia zdecydowana większość lekarzy kieruje dzieci z objawami zaburzeń do dalszej diagnostyki (94,74%). Pacjenci pediatryczni podejrzani o występowanie zaburzeń rozwojowych częściej kierowani są przez lekarzy bieglejszych w technologiach IT oraz dłużej pracujących w podstawowej opiece zdrowotnej.

Podobnie jak w przypadku rodziców, preferencje co do metody przeprowadzania badań screeningowych (online vs stacjonarne) są mocno podzielone - 53,68% lekarzy opowiada się za badaniami on-line. Statystycznie istotne zależności zaobserwowano jedynie pod względem wieku i stażu pracy lekarzy (lekarze młodsi oraz z mniejszym stażem w podstawowej opiece zdrowotnej częściej preferowali wersję internetową (odpowiednio H = 27,876, p = 0,046 i H = 24,384, p = 0,041). Odpowiedzi respondentów wskazują na wyraźną, istotną statystycznie różnicę w chęci skorzystania z kwestionariuszy przesiewowych w różnych sytuacjach w gabinecie lekarskim (H = 46,069, p < 0,001); lekarze istotnie chętniej skorzystaliby z narzędzi przesiewowych w celu potwierdzenia zauważonych objawów (M = 4,362, gdzie M – średnia (ang. *mean*)) oraz potwierdzenia lub rozwiania wątpliwości rodziców (M = 4,362). Chęć stosowania tych metod w ramach populacyjnego screeningu jest znacznie niższa (w przypadku

wizyty okołoszczepiennej w 18 miesiącu życia – M = 3,512; w przypadku bilansu 2-latka – M = 3,828).

W przypadku zalet badań online, rodzice najczęściej doceniają wygodę badania (oszczędność czasu i możliwość przeprowadzenia badania z domu – odpowiednio 68,18% i 78,23% rodziców) oraz łatwiejszy dostęp do screeningu. Wśród negatywów rodzice najczęściej wskazują na brak możliwości weryfikacji wyniku i oceny rozwoju dziecka bezpośrednio przez specjalistę (57,89%) oraz brak osobistego kontaktu z badającym (46,65%). Podobne wyniki uzyskano wśród lekarzy – również najczęściej zwracano uwagę na wygodę badań przesiewowych online; efekt ten był jednak istotnie niższy niż u rodziców (p = 0,025 i 0,002). Jako główną wadę badań przesiewowych on-line lekarze również najczęściej wskazują na brak bezpośredniego kontaktu z osobą przeprowadzającą badanie (55,79%).

Badani lekarze najczęściej zwracają uwagę na to, że podczas wizyty brak jest wystarczającej ilości czasu, by móc wykonać badanie przesiewowe w kierunku zaburzeń rozwojowych – takiej odpowiedzi udzieliło prawie 92% lekarzy. Jest to także najpoważniejsza przeszkoda we wdrożeniu screeningu populacyjnego wg badanych. Mniejszy odsetek lekarzy zgłasza trudności w dostępie do specjalistów oraz brak jednoznacznych zaleceń klinicznych dotyczących badań przesiewowych w kierunku zaburzeń rozwojowych.

9. Wnioski

1. Mimo istnienia licznych kwestionariuszy diagnostycznych do wykrywania zaburzeń ze spektrum autyzmu, wiele z nich jest trudno dostępnych, nie przeprowadzono dla nich pełnej walidacji, bądź cechują się niskimi wartościami psychometrycznymi oraz diagnostycznymi.

2. Niewiele z istniejących adaptacji kwestionariuszy przesiewowych zostało poddanych pełnemu dopasowaniu kulturowemu i lingwistycznemu, co mogłoby zwiększyć ich przydatność diagnostyczną.

3. Polska wersja kwestionariusza CSBS-DP ITC cechuje się wysoką rzetelnością (α Cronbacha 0,92 oraz ω McDonalda 0,92), dobrym dopasowaniem w zakresie modeli zarówno jednoczynnikowych jak i trzyczynnikowych w przeprowadzonej konfirmacyjnej analizie czynnikowej,

4. Wykazano także satysfakcjonujące dopasowanie podmodeli zależnych od płci w konfirmacyjnej analizie czynnikowej polskiej kwestionariusza CSBS-DP ITC i nie wykazano istotnych różnic w osiąganych wynikach pomiędzy podgrupami dziewczynek i chłopców.

5. Polska wersja kwestionariusza CSBS-DP ITC cechuje się stabilnością pomiaru (zarówno w zakresie upływu czasu, jak i oceny przez innego obserwatora).

6. Wyniki uzyskane za pomocą polskiej wersji kwestionariusza CSBS-DP ITC wykazują odwrotną korelację z wynikami uzyskanymi w kwestionariuszu ASRS, a siła tego efektu rośnie wraz z wiekiem badanego, co wskazuje na rosnącą trafność predykcyjną polskiej wersji kwestionariusza CSBS-DP ITC wraz z wiekiem dziecka.

7. W poszczególnych grupach wiekowych przy wyznaczonych punktach odcięcia z użyciem metody Youdena czułość kwestionariusza waha się od 0,667 do 0,750, swoistość od 0,854 do 0,939, dodatnia wartość predykcyjna od 0,261 do 0,4, a ujemna wartość predykcyjna – od 0,979 do 0,981. Dokładność badania przesiewowego waha się w zależności od grupy wiekowej od 0,847 do 0,923.

8. Kwestionariusz CSBS-DP ITC może być wykorzystany w populacyjnych badaniach przesiewowych wśród polskich dzieci w wieku od 9 do 24 miesiąca życia jako skuteczne narzędzie diagnostyczne.

9. Częstość występowania ASD w grupie normalizacyjnej okazuje się być znacząco wyższa niż dotychczasowe szacunki w polskiej populacji i znacznie bliższa do wartości osiąganych w

badaniach w krajach sąsiednich, co wskazuje na potrzebę poprawy w zakresie dostępności do metod diagnostycznych w kierunku ASD w Polsce.

10. Zarówno rodzice, jak i lekarze zauważają potrzebę wdrożenia badań przesiewowych w kierunku ASD do codziennej praktyki, jednakże na drodze stoją liczne ograniczenia systemowe – szczególnie dotyczące ograniczeń czasowych oraz trudności z dostępami do specjalistów.

11. Screening elektroniczny ASD jest wysoko oceniany przez rodziców i może w dużej części zastąpić badanie przesiewowe prowadzone stacjonarnie w placówkach podstawowej opieki zdrowotnej. Niemniej, zarówno rodzice, jak i lekarze zwracają uwagę na brak możliwości weryfikacji wyniku badania przesiewowego uzyskanego w ten sposób za pomocą bezpośredniej obserwacji.

10. Cykl publikacji stanowiących rozprawę doktorską

10.1. Tools for early screening of autism spectrum disorders in primary health care – a scoping review

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Tools for early screening of autism spectrum disorders in primary health care – a scoping review

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Abstract

Background: Autism spectrum disorder (ASD) is a neurodevelopmental disorder that manifests itself in early childhood. Early diagnosis of these disorders allows for the initiation of early therapy, which is crucial for the child's further functioning in society.

Objectives: This review aims to gather and present the existing ASD screening tools that can be used in primary care and adapted to different countries conditions linguistically and culturally.

Eligibility criteria: We searched for English-language publications on ASD screening tools for children aged 0–3 years suitable for use in primary care (i.e. free, requiring no additional training or qualifications).

Sources of evidence: Four databases were explored to find English studies on ASD screening tools intended for the rapid assessment of children aged 0–3.

Charting methods: The information sought (specific features of the questionnaires relevant to primary health care workers, psychometric and diagnostic values of a given cultural adaptation of screening tools, and the linguistic and cultural changes made) were extracted and collected to create profiles of these tools.

Results: We found 81 studies which met inclusion criteria and underwent full data extraction. Three additional data sources were included. These allowed to create 75 profiles of adaptations for 26 different screening tools and collect data on their psychometric values and characteristic features.

Conclusions: The results of our study indicate the availability of several diagnostic tools for early ASD screening in primary care setting concordant culturally and linguistically with a given population. They could be an effective method of accelerating the diagnostic process and starting personalized therapy faster. However, most tools have significant limitations – some are only available for research purposes, while others do not have scientific evidence to prove their effectiveness.

Keywords: Autism spectrum disorder, Diagnostic screening programs, Primary health care

Introduction

Autism spectrum disorder (ASD) is a category of neurodevelopmental disorders characterized by challenges concerning social skills, speech development and behavior [1]. The cause of ASD is not known—it was suggested that the etiology includes many factors, including genetic, infectious or metabolic ones [2]. These disorders

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occur in all racial, ethnic and socioeconomic groups [3]. The prevalence is yet to be clearly defined; however, the World Health Organization (WHO) estimates that ASD occurs in 1 in 160 children worldwide [4]. However, this estimate varies considerably depending on the research method and country. For example, in Israel, it is 4.8%; in Iceland – 3.13%; in the United States – 1.7%; in Qatar – 1.14%; in Iran – 0.06% [5–10]. Thus, the percentage of individuals with ASD in the population depends primarily on diagnosis methods. The growing number of registered cases of ASD in recent years probably results from a greater number of diagnosed adults and children than changes in the frequency of the autism spectrum phenotype in the population [11].

Diagnosis of ASD is a long-term and multi-stage process aimed at recognizing existing disorders and assessing a child's functioning on many levels. It begins with observing the child by parents, guardians, or other people who have contact with the child. It is also necessary to exclude other diseases that may cause symptoms similar to ASD. For this reason, consultations with other specialists (e.g., audiologists, laryngologists, geneticists) are necessary. The final stage is the definitive diagnosis by a team of specialists (psychiatrists, psychologists, special educators, or speech therapists) [12].

The role of family doctors and pediatricians in early diagnosis of ASD

Family doctors or pediatricians working in a primary care clinic most often observe a child during infancy and early childhood, especially as part of well-child care visits, qualifications for vaccinations, or visits due to common infectious diseases. This fact enables careful observation of the child's development and behavior in the critical period for diagnosing ASD, which means that the general practitioner (GP) may be first to notice the behavioral signs of disorders [13–15].

A desirable situation from the patients' point of view is the GP taking the role of a "gatekeeper," i.e., a person who notices the first "red flags" in the child's behavior, analyzes the concerns raised by parents, and decides about the need for further specialist consultations [16]. During the aforementioned visits, parents ask questions about the symptoms they notice and express concerns about their child's development [13]. Unfortunately, there are still frequent situations when doctors marginalize, minimize, or ignore the concerns raised by parents [17]. This may be due to organizational reasons related to primary health care structure (e.g., limited consultation time, excessive workload) [16, 18]. Moreover, identifying some ASD-specific features (e.g., sensory disorders) requires – apart from experience in this matter – devoting more time to patients than is generally provided for a visit in primary care clinics [19]. Another problem that hinders early diagnosis in primary care is the insufficient knowledge of doctors about ASD. A study conducted in 2020 showed that only 23% of primary care physicians (PCPs) had sufficient knowledge about ASD, and the percentage of such doctors was higher in countries with higher income [20]. For example, in Pakistan, only 44% of GPs knew the concept of autism, and only 42% of them had any further knowledge about it [21]. The driving force to improve the knowledge and skills of PCPs in the field of ASD may be the growing public awareness of the issue. Unfortunately, the spread of the term "autism" in society produced mixed results. On the one hand, greater awareness of the problem allowed many families to get help and additional financial resources; on the other hand, it also led to an uncontrolled public debate and spread of unfavorable stereotypes and untruths about ASD and its etiology [22–24]. A better method of spreading knowledge about ASD is special training for doctors by experts [25].

Possibilities of early detection of ASD

Identification of autism spectrum disorders is challenging in the early stages of life when changes in development are rapid and symptoms – often subtle [26]. However, early diagnosis is a necessary first step to implement effective therapy appropriate to the child's needs at a critical time of development – the younger the child at the time of ASD diagnosis, the better therapy results [27–29].

In order to increase the effectiveness of PCPs in the early diagnosis of ASD, numerous screening questionnaires have been developed, which their proponents claim to be some of the most beneficial health policy innovations ever created for children with ASD [30]. On the other hand, ASD screening is criticized in terms of cost-effectiveness or time constraints and the low psychometric properties of tests, especially in very young children [31–33]. However, there is evidence suggesting that including screening tools in routine medical appointments may result in earlier and more accurate identification of children who need further help than relying solely on clinical impressions, which is particularly important when care providers are less experienced in diagnosing ASD [34, 35]. Since the effectiveness of detecting ASD using various questionnaires (understood as the percentage of true positive results) increases with age, very early diagnosis of the youngest children is one of the major therapeutic problems. For such patients in whom screening is associated with tests of low psychometric properties, developmental follow-up is essential later in life. A solution to these problems may be developing novel and better diagnostic methods that take into account both the age and gender of the child [36].

Since 2006, the American Academy of Pediatrics (AAP) has recommended routine diagnosis of ASD at 18 and 24 months of age during well-child care visits [37]. Children who receive a positive screening result should be sent for further ASD evaluation to an early intervention center and referred to an audiologist to rule out hearing impairment, as recommended by the AAP [38, 39]. Over 14 years, these activities significantly increased the prevalence of ASD and made primary care facilities the main places of early diagnosis of ASD [40]. Following these recommendations resulted in more than 50% of American children undergoing screening for autism spectrum disorder [41–43]. In addition, the increasing availability of screening significantly lowered the age of ASD diagnosis in the US, with diagnosis before the age of 4 made in 71% of children (2018) compared to 58% in 2014 [40, 44].

In turn, the recommendations of the US Preventive Services Task Force indicate the lack of sufficient evidence in favor or disadvantage of performing ASD screening in children, for whom no concerns of ASD have been raised by their parents or a clinician [45].

Aim of the study

The main aim of this scoping review was to demonstrate available, culture-specific and language-adapted tools for the early screening of autism spectrum disorders in children from 0 to 3 years of age, that can be used by healthcare professionals working in primary care. We were interested in gaining better insight into their psychometric properties and cultural adaptations, which is particularly important due to the social diversity of cultures [46]. Our final goal is to identify the most relevant tools for screening for ASD in primary care.

The collected data can be used by primary care professionals to select the best tool for the early diagnosis of ASD in their daily practice to accelerate the therapeutic process and for specialists in this field to highlight existing gaps.

Materials and methods

In this research we used the five-step approach described by Arksey and O'Malley to conduct a scoping review: 1) identifying the research question, 2) identifying relevant studies, 3) selecting the studies, 4) charting the data, 5) collating, summarizing and reporting the results [47]. The whole process was dynamic and iterative, with each step discussed with a group of investigators. The Arksey and O'Malley's framework is the primary method of conducting a scoping review which synthesizes the knowledge from the previous literature and allows to adapt the data for the purposes of the study. As the ambiguity of concepts remains the main disadvantage of this approach, when designing the study we also relied on the recommendations that appeared later e.g. Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews (PRISMA-ScR) (Additional file 1) [48]. We do not have a published protocol for this study.

Identifying the research question

Our scoping review focused on answering the question: What are the suitable ASD screening questionnaires available that can be used in primary health care, and what are their characteristics? By suitability we mean a free (available in the public domain or after contact with the authors), short screening questionnaire, completed by a parent or clinician, characterized by good psychometric values and requiring no additional training in order to use it.

Identifying relevant studies

The primary search strategy was developed collaboratively by all authors. We conducted an online search using four different scientific databases containing articles concerning medical and psychological sciences (PubMed, EBSCO, Scopus, and Web of Science) to find publications related to the early diagnosis of children with ASD. We used Mendeley to collect and organize the references. The search began in March 2021 and included all publications written in English and released from January 1980 to May 2021.

The initial search results included a large number of studies related to developmental screening processes and provided guidance and recommendations for the use of screening tools (e.g. AAP guidelines) [37]. The results also included research describing the development and validation of tools, the adaptation of screening tools, and comparisons between individual instruments. Using previously published scientific research on ASD screening tools and our literature search, we compiled a list of tools used for this purpose [49, 50].

As names of screening tools were not mentioned in the title or keywords of many peer-reviewed papers, we also performed individual searches to identify them. Therefore, at each stage of the search for screening tools (step 1), an individual search (step 2) was performed using the name of each instrument indicated in the general search results. In addition, we adapted the search string to the thesaurus of three other databases. Finally, using a snowball approach, we added articles of the reference lists if they met the inclusion criteria mentioned below but were not listed in the initial search. The exact terms we used in the searches can be found in S1 File.

Unfortunately, not all the information sought by us was available in peer-reviewed scientific publications. Therefore, we collected information about screening

instruments from several sources. For example, we checked test reviews and articles describing psychometric properties in peer-reviewed journals, manuals, technical papers, doctoral dissertations, and information from test publishers or distributors.

Inclusion criteria

1. Studies on tools intended for diagnosing children from 0 to 3 years of age;
2. Research describing the use of the tool published in English (or at least an abstract providing the necessary information);
3. Research on the tool intended for screening or rapid assessment, not a formal diagnosis of ASD.

Exclusion criteria

1. Studies on tools intended for formal diagnosis (for this reason, instruments such as the Autism Diagnostic Observation Schedule (ADOS) or Bayley Scales of Infant Development (BSID) were excluded from the study). We also excluded more complex tools beyond the competencies of family doctors, requiring additional training or completion of training authorizing to use them (e.g., Ages and Stages Questionnaire (ASQ), Social Responsiveness Scale (SRS-2), Achenbach System of Empirically Based Assessment (ASEBA), Parents' Evaluation of Developmental Status (PEDS), or Autism Spectrum Rating Scales (ASRS));
2. Studies on tools intended for screening children older than three years. For this reason, the publication omits, for example, the Social Communication Questionnaire (SCQ), which, according to the authors of the tool, is intended for screening of children over four years of age;
3. Research on diagnostic tools used in screening for other developmental disorders.

Information on screening tools was not always readily available; therefore, the decision to include a particular instrument was made based on the best current knowledge. After individual searches, some tools were excluded as they were replaced with a newer, improved version.

Selecting the studies

We imported all titles of our search into Rayyan software and deleted duplicates [51]. Reviewers in pairs (MSo and MBF, AS and MSe) read the titles and abstracts of the studies found following the search strategy to determine their eligibility. Then, studies were categorized as "include" or "exclude." In the event of contradictory information or disagreement, all the authors responsible for the publication made a final decision after a discussion. Finally, full texts of the selected studies were retrieved for a final review and distributed among the researchers in the same pairs. As before, authors jointly decided to include or exclude given publication for this scoping review in case of doubt.

Charting the data

Data from all studies included in the review were extracted and collected in an Excel spreadsheet to create an appropriate profile for each tool and determine its suitability for use in a primary care setting. The spreadsheet presents information about the purpose of the instrument, children age range, required time to complete the questionnaire, information whether an assessment report (e.g., filled in by a parent or guardian) or a direct assessment was used (e.g., observation of a child's behavior), and its psychometric and diagnostic properties. We were also interested in knowing whether any cultural changes were made in a given questionnaire adaptation. The same pairs of reviewers involved in the study selection extracted data from selected studies using an Excel sheet and discussed the discrepancies. To calibrate our data extraction, MSo prepared a calibration exercise on five studies, which improved data extraction.

Collating, summarizing and reporting the results

After extracting the data, we created tool profiles to standardize the available information about their characteristics, properties, and application in primary care. Each tool that met the inclusion criteria for the study received its profile with data on the name, abbreviation, time of completing the questionnaire, and the person responsible for completing it. In addition, each adaptation of the questionnaire received its line on the spreadsheet for the country for which the validation was prepared, the language into which the text was translated, psychometric and diagnostic data (i.e., reliability, sensitivity, specificity, positive and negative predictive value), and the population in which the study was conducted (with an indication of the specific features of this population). Additionally, we marked in the spreadsheet whether a given version of the questionnaire is the original version and whether the adaptations were subject to linguistic and cultural changes. Figures were rounded to the second decimal point.

Results

The initial search yielded 330,225 titles, of which 227,371 were duplicates. After the first screening of titles and abstracts, we assessed 154 full text studies and finally

identified 81 studies, which met inclusion criteria and underwent full data extraction. Three additional data sources were included outside of database searches, e.g. test manuals available on-line (see Fig. 1). All collected data are presented in Table 1.

Study characteristics

The studies described research from 37 countries; most studies originated from the US ($N=18$), Australia ($N=5$), and South Korea ($N=4$). In addition, one article reported a study conducted in nine Arabic countries (Egypt, Kuwait, Jordan, Oman, Qatar, Saudi Arabia, Syria, Tunisia, and Lebanon), and one from the US conducted on a group of Nepalese refugees from Bhutan [92, 102]. The number of scientific papers published during the period under review was relatively stable, with an increase over the last five years (2016–2021).

Study objectives

The studies included in the review had varied purposes; however, a significant majority focused on determining the psychometric values of the tools. Reliability (defined as Cronbach's alpha) was provided in 46 of all studies (one study reported only the factor analysis of the instrument), sensitivity was assessed in 53 studies, specificity in 51 studies, positive predictive value (PPV) in 47, and negative predictive value (NPV) in 36 studies. Two studies aimed to determine the cut-off points for the study population for a given tool [77, 78]. One study aimed to demonstrate the need for further research on the cultural and linguistic adaptation of screening questionnaires and simplifying the wording used in them [102]. Finally, one study was designed to test the stability of the cross-cultural measurement, and one aimed to identify possible difficulties related to translating the ASD screening

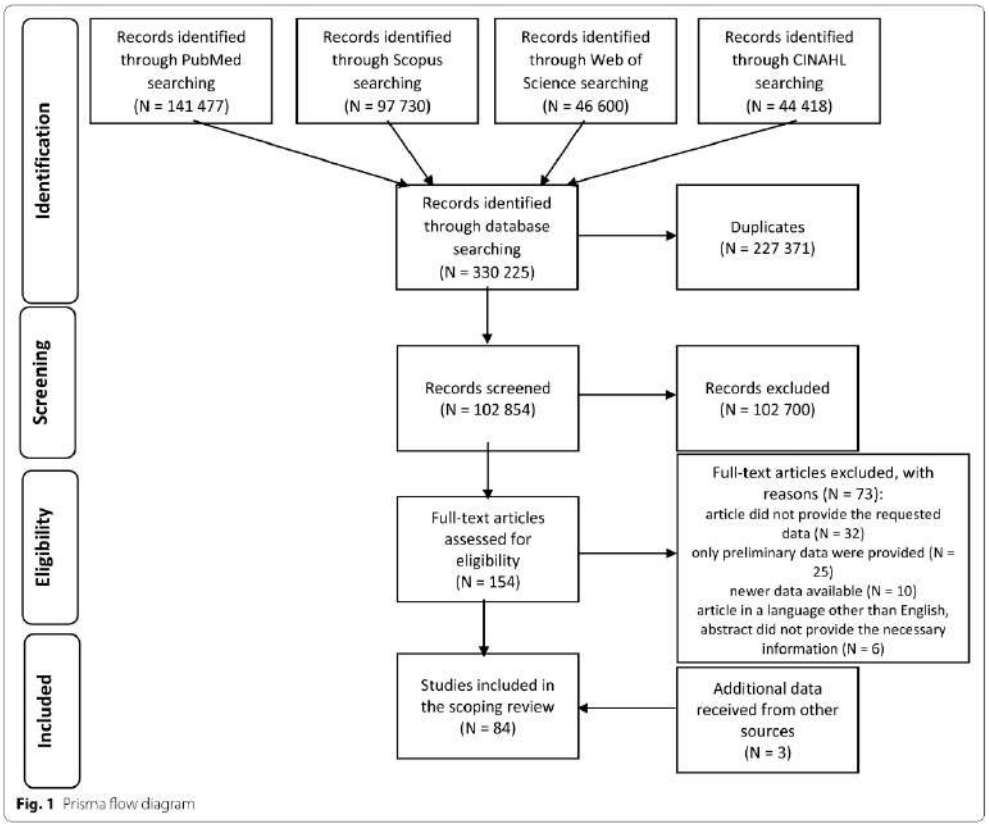

Fig. 1 Prisma flow diagram

Table 1 Overview of available tools for the screening of autism spectrum disorders that met the study criteria

Full name of the tool	Abbreviation	Administer time (in minutes)	Age of the tested child (in months)	Person completing the questionnaire	Country	Language	Reliability (Cronbach's alpha)	Sensitivity
23-Item Screener	**23Q**	NDA	24–108	parents	Uganda	Luganda	NDA	0.8
Autism Observation Scale for Infants	**AOSI**	20	6–18	health professionals	Canada	English	0.68–0.94	0.84
Autism Parent Screen for Infants	**APSI**	10–15	6–24	parents	Canada	English	0.77–0.92	0.67
Baby and Infant Screen for Children with aUtism Traits	**BISCUIT**	30	17–37	parents or caregivers	USA	English	NDA	0.67–0.94
Behavior Development Screening for Toddlers	**BeDevel**	18–42	10–15	parents or primary caregivers	South Korea	Korean	0.87–0.96	0.83
Brief Autism Detection in Early Childhood	**BADEC**	10–25	12–36	observers	Australia	English	NDA	0.81
Brief Infant Toddler Social Emotional Assessment	**BITSEA**	15	12–36	parents	Finland	Finnish	0.75	NDA
					France	French	0.65–0.79	NDA
					Netherlands	Dutch	0.61–0.76	NDA
					Turkey	Turkish	0.72–0.82	0.83–0.90
					USA	English	0.79	0.67–0.95
Chandigarh Autism Screening Instrument	**CASI**	15–20	18–120	parents	India	Hindi	0.86	0.89
Checklist for Autism Spectrum Disorders	**CASD**	15	12–204	observers	USA	English, Spanish	0.97	0.86
Checklist for Early Signs of Developmental Disorders	**CESDD**	NDA	3–39	observers	Belgium	Dutch	NDA	0.80
Communication and Symbolic Behavior Scale-Infant and Toddlers Checklist	**CSBS-DP**	5–10	6–24	parents	Australia	English	0.82	NDA
					South Korea	Korean	0.90	NDA
					Taiwan	Chinese	0.77	NDA
					USA	English	0.87	0.89
Developmental Behavior Checklist-Early Screen	**DBC-ES**	5–10	18–48	parents or teachers	Australia	English	0.87	0.75
Early Screening Autistic Traits Questionnaire	**ESAT**	10	14–15	parents	Netherlands	Dutch	NDA	0.68
					Norway	Norwegian	NDA	NDA

Table 1 (continued)

Full name of the tool	Abbreviation	Administer time (in minutes)	Age of the tested child (in months)	Person completing the questionnaire	Country	Language	Reliability (Cronbach's alpha)	Sensitivity
First Year Inventory	**FYI**	30	12	parents	China	Chinese	The study was aimed at determining the cut-off points for the study population	
					Israel	Hebrew	0.69	The study was aimed at determining the cut-off points for the study population
					Italy	Italian	The study was designed to test the stability of the cross-cultural measurement between American and Italian children	
					USA	English	0.81	0.44 (0.41 in sample of high risk children)
					USA	Spanish	The study aimed to identify possible difficulties related to translating the ASD screening questionnaires to adapt them to other languages and cultures	
INCLEN Diagnostic Tool for Autism Spectrum Disorder	**INCLEN-ASD**	45–60	24–108	observers	India	English, Gujarati, Hindi, Khasi, Konkani, Malayalam, Odia, Telugu, Urdu	0.96	0.98
Joint attention-observation schedule	**JA-OBS**	5–10	30	nurses	Sweden	English, Swedish	0.93	0.86
Modified-Checklist for Autism in Toddlers (Revised)	**M-CHAT (R/F)**	5–10	16–30	parents	Albania	Albanian	0.737	NDA
					Argentina	Spanish	0.76	NDA
					Brazil	Portugese	0.95	0.94
					Chile	Spanish	0.89	1.0
					China	Chinese (Mandarin)	0.57	0.96
					Egypt	Arabic	Version of the questionnaire during the assessment of psychometric values—only preliminary results are presented	
					Egypt, Jordan, Kuwait, Lebanon, Oman, Qatar, Saudi Arabia, Syria, Tunisia,	Arabic	NDA	0.87–0.95
					France	French	NDA	0.67
					Iceland	Icelandic	NDA	0.62
					Indonesia	Indonesian	NDA	0.89

Table 1 (continued)

Full name of the tool	Abbreviation	Administer time (in minutes)	Age of the tested child (in months)	Person completing the questionnaire	Country	Language	Reliability (Cronbach's alpha)	Sensitivity
					Iran	Kurdish, Persian	0.61	0.90
					Israel	Hebrew	NDA	0.70
					Japan	Japanese	0.56	0.72–0.77
					Malaysia	Chinese, English, Malay	NDA	0.18–0.64
					Mexico	Spanish	0.76	NDA
					Nepal (Bhutan/USA)	Nepali	The study aimed to demonstrate the need for further research on the cultural and linguistic adaptation of the screening questionnaires and the simplification of the wording used in	
					Netherlands	Dutch	NDA	NDA
					Norway	Norwegian	NDA	0.34
					Serbia	Serbian	0.91	NDA
					Spain	Spanish	0.59 (without FU), 0.62 (with FU)	0.79
					South Korea	Korean	Only item factor analysis was made	NDA
					Sri Lanka	Sinhala	NDA	0.25
					Sweden	Swedish	NDA	0.77
					Taiwan	Chinese	0.8	0.77–0.88
					Thailand	Thai	NDA	0.91
					Turkey	Turkish	0.67	1.0
					USA	English	0.85	0.91
					Vietnam	Vietnamese	NDA	NDA
Pictorial Autism Assessment Schedule	**PAAS**	15–20	18–40	parents	Sri Lanka	Sinhala, Tamil	0.96	0.89
Quantitative Checklist for Autism in Toddlers	**Q-CHAT**	5	18–24	parents	Iran	Persian	0.89	0.96
					Italy	Italian	0.68	NDA
					Serbia	Serbian	> 0.81	0.96
					Singapore	English	0.53–0.60	NDA
					South Korea	Korean	0.66	NDA
					United Kingdom	English	0.67–0.83	0.44

Table 1 (continued)

Full name of the tool	Abbreviation	Administer time (in minutes)	Age of the tested child (in months)	Person completing the questionnaire	Country	Language	Reliability (Cronbach's alpha)	Sensitivity
Quantitative Checklist for Autism in Toddlers – 10 items	**Q-CHAT-10**	< 5	18–24	parents	Chile	Spanish	0.85	0.93
					Serbia	Serbian	> 0.81	0.39
					United Kingdom	English	0.88	0.91
Rapid Interactive Screening Test for Autism in Toddlers	**RITA-T**	10	18–36	doctors	Lebanon	Arabic	0.91	0.96
					USA	English	NDA	1.00
Social Attention and Communication Study	**SACS**	5	12–24	nurses and other health professionals	Australia	English	0.88	0.84
					China	Chinese	NDA	0.53
Screen for Social Interaction	**SSI**	15	24–61	parents or caregivers	USA	English	0.756	0.58–0.94; 0.87–0.81
Screening Tool for Autism in Two-Year-Olds	**STAT (T-STAT in Taiwan)**	20	24 (12–24) (18–24 in T-STAT)	examiner	Taiwan	Chinese	0.90	0.93
					USA	English	1.00	1.00 (0.95 for children < 24 months)
Three-Item Direct Observation Screen	**TIDOS**	15–20	18–60	trained pediatric-oriented professionals	Turkey	Turkish	NDA	0.8
Young autism and other developmental disorders checkup tool	**YACHT-18**	10	18	nurses	Japan	Japanese	NDA	0.80

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
23-Item Screener	0.77	0.23	0.98	1169 from general population	+			[52]
Autism Observation Scale for Infants	0.98	NDA	NDA	In first study 92 infants siblings of children with ASD for the assessment of reliability, in second—150 infants siblings of children with ASD	+			[53, 54]
Autism Parent Screen for Infants	0.87	0.43–0.79	0.87–0.99	283 in total (79 low risk, 204 high risk)	+			[55]
Baby and Infant Screen for Children with aUtism Traits	0.74–0.89 depending on cut-offs	NDA	NDA	3062 in total (499 with ASD, 383 with PDD-NOS and 2180 with non ASD related atypical development)	+			[56]
Behavior Development Screening for Toddlers	0.81	0.80	0.83	155 in total (75 ASD, 55 TD, 25 DD)	+			[57]
Brief Autism Detection in Early Childhood	0.78	0.81	0.78	270 in total (95 with ASD, 28 with PDD-NOS, 69 ODD, 78 TD)	+			[58]
Brief Infant Toddler Social Emotional Assessment	NDA	NDA	NDA	50 from general population		Translated with some language changes		[59, 60]
	NDA	NDA	NDA	589 from general population		Translated according to international guidelines		[61]
	NDA	NDA	NDA	3170 from general population		Translated according to international guidelines		[62]
	0.88–0.91	NDA	NDA	462 from general population		Translated with "minimal corrections"		[63]
	0.68–0.95	0.74–0.93	0.75–0.93	1788 from general population	+			[64]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
Chandigarh Autism Screening Instrument	0.89	0.67	0.96	405 in total (75 with intellectual disability, 83 with ASD, 87 with DD and 160 TD)	+			[65]
Checklist for Autism Spectrum Disorders	1.00	1.00	1.00	2469 (1052 with ASD, 925 TD, 55 with typical autism and 437 nonautistic clinical children)	+			[66]
Checklist for Early Signs of Developmental Disorders	0.94	0.7	0.99	6808 from general population	+			[67]
Communication and Symbolic Behavior Scale-Infant and Toddlers Checklist	NDA	NDA	NDA	1725 infants already participating in a longitudinal study of language development				[68]
	NDA	NDA	NDA	219 of TD children				[69]
	NDA	NDA	NDA	171 from general population		Translation was made	Substitution of English phonemes with suitable Chinese phonemes; replacement of frequently used words (for example "uh/oh" was replaced with "thank you")	[70]
	0.89	0.94	0.80	2454 in the reliability study, 3026 (3021 of children not previously identified, 5 with DD) in the study of diagnostic properties	+			[71–73]
Developmental Behavior Checklist-Early Screen	0.51	0.77	0.48	207 children with or suspected of DD	+			[74]
Early Screening Autistic Traits Questionnaire	0.96	0.10	0.99	31,724 from general population	+			[75]
	NDA	0.07–0.3	NDA	12,666 from general population		Translation only		[76]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
First Year Inventory				518 from general population		Translation only		[77]
				471 from general population		Items were culturally and linguistically adapted		[78]
				657 from general population		Translation only		[79]
	0.99 (0.81 in sample of high-risk children)	0.31 (0.52 in sample of high-risk children)	0.99 (0.73 in sample of high-risk children)	1496 from general population for study of test reliability; 699 from general population for other properties; 121 in the study on high-risk sample	+			[80–82]
				25		Items were culturally and linguistically adapted		[83]
INCLEN Diagnostic Tool for Autism Spectrum Disorder	0.95	0.91	0.99	In first round of the study – 266 in total (81 with ASD, 120 NDDs, 65 TD); in second round – 154 (90 with average and 64 with subnormal intelligence)	+			[84]
Joint attention-observation schedule	NDA	0.93	NDA	3999 from general population	+			[85]
Modified-Checklist for Autism in Toddlers (Revised)	NDA	0.895	NDA	2594 from general population		Translation only		[86]
	NDA	NDA	NDA	420 from general population		New translation was completed to adapt it to the Spanish used in Argentina, making slight changes to idiomatic turns of phrases and different expressions closer to Argentine vocabulary, for example: item "plaza" (square) was changed into "parque"	Throughout the entire questionnaire, the words "hijo/hija" (son/daughter) were replaced by "nino/nina" (boy/girl), so that the questionnaire could be administered in the case of another relative taking the child to a pediatric check-up	[87]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
	0.91	0.86	0.97	303 from general population		The older version of the M-CHAT test was used in the study, no data available on cultural changes		[88]
	0.98	NDA	NDA	120 in total (20 with suspected ASD, 100 TD)		Semantic changes in 15 out of 20 items, grammatical changes		[89]
	0.86	0.07	1.0	7928 from general population		Translation in accordance with the principles semantic and linguistic of equivalence	Adapting children's behavior to culture: for example—"vacuum the rug" and "mow lawn" are not common activities in China, so we use "wipe the table" instead	[90]
				5546 from general population		Translation only		[91]
	0.76–0.89	0.82–0.9	0.86–0.93	228 in total (122 with ASD, 106 TD)		Translators added the specific dialect of some words to the classical Arabic to clarify the meaning of a number of items of the questionnaire		[92]
	0.94	0.14	0.99	1250 from general population		Translation only		[93]
	0.99	0.72	0.99	1585 from general population		Translation with minor changes	Minor cultural adaptation in follow-up interview – "an action figure" was specified as a "Lego or Playmobil figure"	[94]
	0.95	0.76	0.98	168 from general population without severe sensory and communication disability or ASD diagnosed before		Translation only		[95]
	0.82	0.05 (without FU)	NDA	2941 from general population		Translation only		[96]
	0.98	0.20	1.00	1591 from general population		In this study the original version of M-CHAT was used		[97]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
	0.84	0.08–0.12	NDA	1851 from general population; in reliability study – 24 children		NDA	Authors added some illustrations in order to encourage caregivers to notice negative symptoms	[98, 99]
	1.00	0.26–0.78	0.99	19,297 from general population		Translation only		[100]
	NDA	NDA	NDA	456 in total (117 high-risk, 339 TD)		Translation with minor cultural adjustments	Describing the "peek-a-boo" game (Mexican parents do not have a specific name for it)	[101]
				13 pediatric Nepali refugee patients living formerly in Bhutan (from general population)		Translation only		[102]
	NDA	0.01–0.1	> 0.98	12,102 from general population		Translation only		[76]
	0.93	0.02	NDA	52,026 from general population		Translation only		[103]
	NDA	NDA	NDA	148 in total (128 from general population, 20 high-risk)		Slight adaptation of wording was required due to languages differences		[104]
	0.99	0.39	0.99	6625 from general population		Translation only		[105]
	NDA	NDA	NDA	2300 from general population		Translation with revision of ten Korean mothers living in Sothern California		[106]
	0.71	0.13	0.85	374 from general population (28 with "red flags")		Translation only		[107]
	NDA	0.92	NDA	3999 from general population		A few minor adjustments of the Swedish language were done		[85]
	0.53–0.72	0.63–0.72	0.77–0.82	236 of ASD high-risk sample		Translation only		[108]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
	1.0	0.96	0.99	841 in total (109 high-risk, 732 low-risk)			To increase the suitability of the M-CHAT in a Thai cultural context, authors hypothesized that a screening process that includes both a parent-completed questionnaire followed by a semi-structured interview by trained clinicians, only for cases that initially screen positive that could improve overall sensitivity, specificity, PPV and NPV	[109]
	0.91	0.09	1.0	6712 from general population		Translation only		[110]
	0.99	0.11 (without follow-up interview (FUI))/ 0.65 (with FUI)	0.99	3793 in total (3309 low-risk, 484 high-risk)	+			[111]
	NDA	0.763	NDA	6583 from general population				[112]
Pictorial Autism Assessment Schedule	0.61	0.78	0.77	105 in total (45 with ASD, 30 DD, 30 TD)	+			[113]
Quantitative Checklist for Autism in Toddlers	0.90	NDA	NDA	100 in total (50 TD, 50 ASD)		Translation only		[114]
	NDA	NDA	NDA	2400 from general population		Translation only		[115]
	0.82	NDA	NDA	220 in total		No access to the full content of the article		[116]
	NDA	NDA	NDA	514 from general population		NDA		[117]
	NDA	NDA	NDA	104 in total (24 ASD, 80 unselected)		Translation only		[118]
	0.98	0.28	NDA	In first study – 795 in total (754 from unselected group, 41 ASD); in second study 3770 from general population	+			[119, 120]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
Quantitative Checklist for Autism in Toddlers – 10-items	0.77	0.48	NDA	287 in total (125 TD, 149 DD, 13 ASD)		Translation only		[121]
	0.99	1.00	1.00	220 total		No access to the full content of the article		[116]
	0.89	0.58	NDA	880 in total (754 control, 162 ASD)	+			[122]
Rapid Interactive Screening Test for Autism in Toddlers	1.00	1.00	0.96	48 in total (19 TD, 29 high-risk)		Translation only		[123]
	0.84	0.88	NDA	61 in total (23 ASD, 19 DD/non-ASD, 19 TD)	+			[124, 125]
Social Attention and Communication Study	0.99	NDA	NDA	First study—20,770 from general population, second study – 99 identified as "at risk" in first study	+			[126, 127]
	1.00	0.42	1.00	10,514 from general population		Translated and evaluated with English version to be comparable in meaning		[128]
Screen for Social Interaction	0.61–0.87; 0.70–0.71	NDA	NDA	In first study – 111 in total (51 high-risk and 60 low-risk), in second study – 350 (168 from general population, 182 high-risk)	+			[129, 130]
Screening Tool for Autism in Two-Year-Olds	0.74	0.94	0.97	107 in total; in first stage—15 with ASD and 15 with DD or LI; in second – 77 with ASD, PDD-NOS or DD/LI		Two items of the questionnaire were changed	The toy that is shown to the child has been changed (from a dog to an elephant)	[131]

Table 1 (continued)

Full name of the tool	Specificity	Positive predictive value	Negative predictive value	Population and subgroups size	Changes made in the questionnaire			References
					Original version	Lingual changes	Cultural changes	
	0.85 (0.73 for children < 24 months)	0.86 (0.56 for children < 24 months)	0.92 (0.97 for children < 24 months)	In first research -104 in total (50 with ASD, 15 with PDD-NOS and 39 with DD/LI). In second research – 77 with older sibling with ASD or had been referred for evaluation for concerns about ASD	+			[132, 133]
Three-Item Direct Observation Screen	0.74	0.6	0.87	259 in total (86 with ASD, 76 with DD without ASD, 97 with typical development)	+			[134]
Young autism and other developmental disorders checkup tool	0.863	NDA	NDA	2814 from general population, without any recognized disease or disorder	+			[135]

DD Development delay, *LI* Language impairment, *NDA* No data available, *NDDs* Other neuro-developmental disorders, *PDD-NOS* Pervasive developmental disorder not otherwise specified, *TD* Typical development

questionnaires to adapt them to other languages and cultures [79, 83].

Study populations

The number of participants included in the studies differed significantly, ranging from 13 to 52,026 [102, 103]. 34 studies included more than 1,000 children, while six had more than 10,000 participants.

Children from the general population were included in 46 studies. In eight papers, the research was based only on a group of children at risk. One study was conducted in a group of typically developing children [69]. In the case of three publications, the characteristics of the studied population were not specified. The remaining publications concerned both children with a low and high risk of ASD. It is worth noting the different understanding of the term "high-risk children" in individual papers, as risk groups, for example, included siblings of children diagnosed with ASD, children already diagnosed with ASD or other developmental disorders, or suspected of developmental delay, etc.

Tools characteristics

In the course of the study, we were able to identify 26 different autism spectrum disorder screening tools that met our study criteria.

We would like to point out that while researching the information about tools, we found mixed data on the availability of the Checklist for Autism Spectrum Disorder (CASD) for professionals who are not psychologists or have not completed the appropriate training. Nevertheless, we decided to include CASD in this publication as a tool available to PCPs.

Original versions of questionnaires

The original versions of questionnaires come from 13 countries. Most of them (as much as 35%, $N=9$) were created in the US. Only two questionnaires were developed in low- and low-middle-income countries (Uganda and Sri Lanka) [52, 113, 136]. An even greater disproportion could be observed in the languages in which the original versions of the tools are available. Of the 35 original language versions (some questionnaires such as CASD, JA-OBS, and PAAS were prepared in two languages, and INCLEN-ASD even in nine), almost half ($N=17$) were in English.

Number of language versions and cultural adaptations of ASD screening tools

Data from selected publications allowed us to create 75 profiles of different versions of the adaptations or original versions of ASD screening questionnaires. Most tools were prepared in one country in one language version. At least one questionnaire was tested in a total of 45 different countries. The largest number of various questionnaires was available in the US (11), Australia and South Korea (4 each), China, the Netherlands, and Turkey (3 each).

Some questionnaires in one study were translated into multiple languages simultaneously; however, at least one tool was available in 35 different languages. In some countries, the questionnaires were adapted to the local dialect (e.g., the Spanish versions of M-CHAT were adapted to Spanish, Mexican, Chilean, and Argentinian respondents) [87, 89, 101, 105]. Most of the questionnaires were available in English ($N=21$), Spanish ($N=7$), Chinese ($N=6$), Dutch ($N=4$) and Korean ($N=4$).

At this point, it is worth mentioning that there are many translations of the questionnaires, such as M-CHAT or Q-CHAT, available on the websites of organizations involved in developing them. For example, the most popular M-CHAT is available in 73 versions, but most lack research published in international journals [137, 138]. The situation is similar with the Japanese and Spanish BITSEA versions [139].

Most language versions of the individual questionnaires were translated directly into the language of the surveyed population, sometimes with minor changes. However, for example, in the Argentinian version of the M-CHAT questionnaire, the dialect was changed to match better Spanish used in Argentina. Likewise, in the Taiwanese version of STAT, two items were changed to suit the Taiwanese population better [87, 131].

In addition, cultural changes were made in nine adaptations. For example, phonemes were adapted to the language, and the type of assessed play or the type of toy shown to children was changed to capture their interest.

Psychometric values

When searching for information on different versions of questionnaires, we focused primarily on reliability, sensitivity, specificity, PPV, and NPV. We made the decision not to include validity data in our review due to the considerable variation in the methodology used across studies (different types of validity measured by various means) or other psychometric values (such as positive or negative likelihood ratio) due to the small number of studies containing these data and the desire to simplify the table as much as possible to facilitate its use by practitioners.

Out of all 75 profiles, we were only able to complete 20 of them containing all the five values sought.

Reliability

Internal reliability of the test is a measure defining the consistency of items included in a given scale, i.e., it

determines to what extent the items included in a given factor or scale are similar to each other or whether they test the same phenomenon. The most common measure of reliability is Cronbach's alpha (α) [140]. In the profiles we created, this measure ranges from 0.53 to 1.00. Using the rule of thumb and other different qualitative descriptors methods, 6 of the studies had excellent reliability (α > 0.93), 2 – strong (0.91–0.93), 12 – reliable (0.84–0.90), 14 – relatively high (0.70–0.83), and 13 had reliability below 0.70 [141].

Sensitivity

Test sensitivity is the ratio of the true positives to the sum of the true positives and the false negatives. A sensitivity of 100% would mean that all individuals with existing disorders would be diagnosed. Values of reported sensitivity in 53 profiles varied from 0.18 to 1.00. Most of the tests ($N = 42$) scored above 0.70. There is a significant discrepancy between the sensitivity values between linguistic adaptations of the same type of questionnaire (e.g., M-CHAT used in the US and Sri Lanka), resulting potentially from an inadequate cultural adaptation of the tool [107, 111].

Specificity

Test specificity is the ratio of the true negatives to the sum of the true negatives and false positives. A specificity of 100% would mean that all healthy individuals in the test performed would be marked as healthy. Specificity was calculated for 51 of the above-mentioned versions of questionnaires and ranged from 0.51 to 1.00. In 37, specificity exceeded 0.80.

Positive predicting value (PPV)

PPV is equal to the proportion of true positives out of all positives and determines the probability that a positive test result is accurate. PPV of the questionnaires in the studies included in the review ranged from 0.01 to 1.00, showing a significant variety. Noteworthy is the considerable increase in PPV after the follow-up interview was used in the American version of M-CHAT, showing an increase from 0.11 to 0.65 [111].

Negative predicting value (NPV)

NPV is the proportion of true negatives out of all negatives; it determines the probability that a negative test result is accurate. All versions of questionnaires, except one (DBC-ES with NPV = 0.48), for which NPV was calculated, had NPV greater than 0.73 [74].

Person completing the questionnaire

ASD screening questionnaires can generally be divided into questionnaires filled in by people who have constant contact with the child (parents or guardians) or independent observers – specialists (e.g., doctors, nurses, psychologists, etc.). Most (15 out of 26) tools were intended to be filled by parents, and specialists only dealt with possible doubts arising while filling in the questionnaire and calculated the result of the test. These also tools underwent cultural adaptation much more often than those in which a specialist assessed the child. Some instruments were by definition predisposed to a given professional group, e.g., the assessment of a child's development using the JA-OBS test is performed by nurses [85].

Time of completing the questionnaire

Most of the questionnaires listed above should not take more than 10–20 min for parents or specialists to complete, and some only take 5 min. For example, according to the authors, the shortened version of Q-CHAT (Q-CHAT-10) takes less time than 5 min [122]. On the other hand, BeDevel can take over 40 min to complete, and INCLEN-ASD takes 45–60 min [57, 84].

Discussion

Our research revealed many tools for early ASD screening that can be employed in primary care (26 different instruments in 75 adaptations). An ideal tool for ASD screening seems to be a free and short instrument with items suitable for assessing development, with good psychometric properties, corresponding to the entire studied population, using plain language (low-reading level), easy to assess by people with no experience in psychometrics (easily score-able), providing simple and clear guidance on what to do after screening [142]. Unfortunately, it is unclear which existing tools are best suited for this, so further development of both instruments and research into their use is necessary. Furthermore, there is a possibility that it will be appropriate to create an entirely new tool, which will be much more effective than the existing ones. The problem is further exacerbated by the small number of meta-analyses and systematic reports on the effectiveness of given screening tools [143–145]. Compared to previous studies, we were able to collect data on a much larger number of ASD screening tools available, however our results confirm the previous findings that screening tools for ASD are adequate to detect autism at the early stages of life. The APSI, BITSEA, CESDD, CSBS-DP, M-CHAT, SACS, and STAT deserve recognition, as the studies examining these tools had large sample sizes, and they found these tools in particular to have high psychometric and diagnostic values. For this reason, it seems that these mentioned tools can be used in the population-based ASD screening. The new questionnaires (e.g. BeDevel) look promising as effective tools, but more

research is required. Furthermore, among all included tools CSBS-DP, M-CHAT, and STAT are recommended by the Centers for Disease Control and Prevention (CDC) for ASD screening in the United States [146]. An additional positive in favor of M-CHAT is the multitude of language versions that were at least partially validated; the second such questionnaire is Q-CHAT. Another issue is that evaluating the usefulness of some of the questionnaires mentioned above is based on studies conducted a long time ago.

From the perspective of primary care workers, it is also important to reduce the occupational encumbrance of implementing another examination tool which is the responsibility of the PCPs. Hence, it seems that it would be favorable for PCPs to implement screening questionnaires filled out by a parent. On the other hand, questionnaires in which a neutral observer assesses the child are slightly more effective in detecting early symptoms of autism spectrum disorders [147].

Still, the main problem for PCPs will be choosing the right tool to carry out ASD screening. Positive experiences from the United States, where a mass ASD screening system was implemented successfully, indicate the suitability of using ASD screening tests in primary health care [40, 44]. Unfortunately, experiences from the US cannot be transferred directly to other countries. Furthermore, it is crucial for early diagnosis of ASD to have tools that respond to cultural and linguistic differences (as well as the local perception of "disability"). Hence, the use of mismatched tools may be inappropriate [46]. For example, in Jamaica, the percentage of parents reporting that their child shows developmental delays compared to peers is significantly higher than in Bangladesh or Pakistan [148]. The global application of ASD screening, especially in low- and middle-income countries (LMICs), is associated with many problems because most existing tools were developed in North America or Europe, but they are used – often without any significant modifications – in countries whose cultures differ significantly from those in which they were created. In particular, our study shows that there is a lack of tools to identify children with ASD in Africa and other LMICs [149, 150]. There are many possible causes of this state—the cultural maladjustment of the existing tools developed in Europe and North America, the lack of funds for research, a smaller number of psychiatrists and psychologists per capita than in Western countries, or less interest in the subject of ASD [150, 151]. The inability to diagnose ASD in LMICs leads to significant burden on quality of life and costs of medical care and special education that these communities are increasingly witnessing. Therefore, further steps (i.e. developing new culturally appropriate tools, increasing research funding) are needed to raise awareness of the early detection of ASD among the LMICs communities.

Another difficulty is the availability of some of the tools. Many of the instruments that met the study criteria are only available for scientific use. And even if access to them is free, it requires contact with researchers and the authors' consent for further use.

The lack of available screening tests for individual populations, incomplete validation, or limited availability is not the only difficulty in popularizing early diagnosis of ASD. Screening tests have limited sensitivity—some of the children who received negative screening will receive in subsequent years of their life diagnosis of ASD [152]. Hence, it is not only necessary to pay attention to the dissemination of screening but also to remember the necessity of further continuous monitoring (follow-up) of children's development [152]. The situation is further exacerbated by the fact that there are no readily available (e.g., in the public domain) rapid tests for older children (aged 30–60 months) as is the case with other psychiatric disorders, e.g., Vanderbilt ADHD Assessment Scales for attention-deficit hyperactivity disorder or Screen for Child Anxiety Related Disorders (SCARED) for anxiety disorders. This makes it necessary to decide whether the child should be referred for further tests based solely on the experience of the primary care worker, which may delay the diagnostic process. These limitations can have long-term adverse consequences (e.g. limited availability of screening tools to individual populations, their incomplete validation or the lack of existing guidelines on developmental disorders for GPs) that can lead to a delay in the diagnostic process of ASD, which can significantly increase the age of diagnosis.

This is not the only reason for delayed ASD diagnosis in children. The example of the United States demonstrates that causes for delay may be due to imperfections of the public health system and low predictive values of the tests (especially in children scoring close to the cut-off limits). Examples of such restrictions include the following:

1) not all children receive healthcare as infants,
2) not all children who are receiving healthcare are screened—only 8–28% of pediatricians in the United States use ASD screening tools in their daily practice [153, 154],
3) not all screened children undergo additional consultations in case of a positive result [42, 43] – only 31% of children with a positive screening test were referred for further diagnosis, 20% to an early intervention center and 36% to an audiologist [155]; these values are slightly higher in another study [42].

The data above show that even with the widespread of the idea of ASD screening, it may not be enough for a complete diagnosis of all affected children. In this case, the delay or lack of diagnosis is primarily due to omissions of the diagnostic process on the part of health care workers.

From an ethical point of view, it should be noted that lowering the age at which the diagnostic process begins in the population will result in an increased number of "false positive" cases, which entails a lot of stress experienced by the families of children that could be difficult to counteract in primary health care [33, 49]. Another problem is that it implies the rising cost of additional evaluation processes in children developing correctly.

The most controversial issue regarding universal ASD screening in children is the cost-effectiveness of ASD screening, primarily due to the moderate accuracy of current tools and the low prevalence of the disorder [45] Attempts to estimate the cost-effectiveness of ASD screening indicate that universal screening may not be financially sound mainly due to delays in further diagnostic and therapeutic steps [33]. Eliminating the waiting time for further consultations with simulation models showed that the initial high cost incurred for screening might be offset by future savings resulting from improved functioning of ASD patients in society. However, the same analyses conducted in high-risk children showed the cost-effectiveness of screening. Nevertheless, the significant benefits of early intervention justify attempts to further refine this strategy for the early detection of autism spectrum disorders [156].

Limitations of the study

There are several limitations to this review. This study includes only scientific publications whose full text was in English or had an abstract containing most of the necessary data to create a profile for the tool. Because the goal of researchers studying early diagnosis of ASD is the implementation of the instruments in a given country, some existing research may have been excluded due to the publication of results of the validation process in languages other than English in local peer-reviewed journals.

The review was carried out mainly by using search string for publications in four scientific databases, potentially limiting the results. We searched for publications over a broad period of time (1980–2021), which increased the number of available manuscripts. This may be a drawback of the research, because we could include in the study tools, the use of which in practice may prove difficult or ineffective.

It should also be noted that researchers carried out the measurements of psychometric and diagnostic properties of various tests in different ways, making it impossible to compare their parameters without taking into account the methodological details contained in the source texts.

During the review process of the article, research on a new, promising screening tool—Early Screening for Autism and Communication Disorders (ESAC) was published. ESAC consists of 46 items, covers children between 12–36 months and has a reliability ranged from 0.92 to 0.95, sensitivity between 0.86 and 0.92 and specificity between 0.74 and 0.85 in an American population [157].

Conclusions

The results of our review show that there are several diagnostic tools for early ASD screening that can be used in a primary care setting for which the full validation process was carried out and showed high psychometric and diagnostic values. These tools could effectively accelerate the diagnostic process and lead to a faster start of personalized therapy. As some examples show (e.g. Icelandic version of M-CHAT or Taiwanese version of STAT), they could also become the basis for preparing almost equally effective adaptations of screening tests for different populations, especially after introducing cultural and linguistic modifications [94, 131].

Unfortunately, for a large part of the tools, no changes other than accurate translation were made to fit the questionnaire to the characteristics of the particular population. Furthermore, only partial validation studies were carried out in many cases, which means that using them in everyday practice may be ineffective. Finally, the more culturally different two populations are, the more a tool designed for one will be less effective for the other.

Therefore, it appears necessary to continue research on adaptations of existing ASD screening methods and attempt to improve them and constantly increase the knowledge of health care professionals about ASD, improve the follow-up process, and further evaluate the cost-effectiveness of the ASD screening process.

Practical implications

This review highlights the available options for early diagnosis of ASD in primary care from a global perspective, indicating the importance of psychometric and diagnostic values in choosing the most suitable tool for everyday practice.

Abbreviations

23Q: 23-Item Screener; AAP: American Academy of Pediatric; ADHD: Attention-deficit hyperactivity disorder; AOSI: Autism Observation Scale for Infants; APSI: Autism Parent Screen for Infants; ASD: Autism spectrum disorder; BADEC: Brief Autism Detection in Early Childhood; BeDevel: Behavior Development Screening for Toddlers; BISCUIT: Baby and Infant Screen for Children with aUtism Traits; BITSEA: Brief Infant Toddler Social Emotional Assessment;

CASD: Checklist for Autism Spectrum Disorder; CASI: Chandigarh Autism Screening Instrument; CESDD: Checklist for Early Signs of Developmental Disorders; CSBS-DP: Communication and Symbolic Behavior Scale-Infant and Toddlers Checklist; DBC-ES: Developmental Behavior Checklist-Early Screen; DD: Development delay; ESAC: Early Screening for Autism and Communication Disorders; ESAT: Early Screening Autistic Traits Questionnaire; FYI: First Year Inventory; GP: General Practitioner; INCLEN-ASD: INCLEN Diagnostic Tool for Autism Spectrum Disorder; JA-OBS: Joint attention-observation schedule; LI: language impairment; LMICs: Low and middle income countries; M-CHAT (R/F): : Modified-Checklist for Autism in Toddlers (Revised); NDA: No data available; NDDs: Other neuro-developmental disorders; NPV: Negative predicting value; PAAS: Pictorial Autism Assessment Schedule; PCP: Primary care physician; PDD-NOS: Pervasive developmental disorder not otherwise specified; PPV: Positive predicting value; Q-CHAT: Quantitative Checklist for Autism in Toddlers; Q-CHAT-10: Quantitative Checklist for Autism in Toddlers – 10-items; RITA-T: Rapid Interactive Screening Test for Autism in Toddlers; SACS: Social Attention and Communication Study; SSI: Screen for Social Interaction; STAT : Screening Tool for Autism in Two-Year-Olds; TD: Typical development; TIDOS: Three-Item Direct Observation Screen; WHO: World Health Organization; YACHT-18: Young autism and other developmental disorders checkup tool.

Supplementary Information

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Additional file 1.

Additional file 2.

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Authors' contributions
MSo and MMBF were responsible for conceptualization of the study an orginal draft preparation. MSo was responsible for data curation. All authors were responsible for formal data analysis, investigation and methodology. MMBF and MSe supervised research. All authors read and approved the final manuscript.

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Declarations

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Not applicable.

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10.2. Reliability and validity of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist

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Reliability and validity of the Polish version of Communication and Symbolic Behaviour Scales-Developmental Profile - Infant-Toddler Checklist

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ABSTRACT

Background: Early diagnosis of autism spectrum disorders allows to start a therapy tailored to the child - the earlier it happens, the better the results. This study marked a preliminary attempt to prepare culturally and linguistically adapted Polish version of the Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist (CSBS-DP ITC).
Method: The study was conducted among 1471 Polish parents of children from the general population aged 6 to 24 months, 490 of them participated in the follow-up. An additional 122 pairs of parents completed the questionnaire to determine reliability using the inter-rater method. The collected data were statistically processed to calculate the reliability and validity of the Polish version of the CSBS-DP ITC questionnaire.

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Results: The results indicated a very good fit of the one-factor and three-factor models in confirmatory factor analysis. Both one-factor and three-factor model showed satisfactory fit in both sex subsamples. The total score of CSBS-DP ITC demonstrated satisfactory internal consistency, Cronbach's $\alpha = .92$ and McDonald's $\omega = .92$. In test-retest all the correlation coefficients between the first and second measurements were above.70, proving a satisfactory level of stability.
Conclusions: The Polish version of the CSBS-DP-ITC is a reliable tool for the early screening of autism spectrum disorder, but further studies on sensitivity and specificity are essential.

1. Introduction

Autism spectrum disorders (ASD) are neurodevelopmental disorders of undetermined etiology, most often manifested in early childhood, which affect daily activity and are characterized primarily by difficulties in communication and interpersonal interactions and limited interests or repetitive behaviors (American Psychiatric Association, 2013). Their prevalence is not clearly defined - the World Health Organization (WHO) estimates that ASD affects 1 in 160 children worldwide (Elsabbagh et al., 2012). More recent data from systematic review performed by Zeidan et al. determine the worldwide prevalence of ASD on 1/100 (Zeidan et al., 2022). According to official data from the Polish National Health Fund from 2012, the prevalence rate of ASD in individuals under the age of 18 in Poland is 3.4 cases per 10,000 children (Piskorz-Ogórek et al., 2020). More accurate and newer preliminary data from two Polish provinces estimate that ASD occurs in one in 286 children (Skonieczna-Zydecka et al., 2017).

The first symptoms of ASD usually appear during infancy or early childhood - prodromal symptoms may be observed as early as in the first six months of a child's life (Canu et al., 2021; Zwaigenbaum et al., 2013). A reliable diagnosis of ASD in a child can be reached as early as 2–3 years of age (Chawarska et al., 2007). However, due to delays associated with the diagnostic process (or its omissions), this diagnosis is made much later - in the 2019 meta-analysis, the average age of ASD diagnosis is 60.48 months (range 30.90–234.57); however, when only children $\leq$ 10 years of age were taken into account, this age was 43.18 months (range 30.90–74.70) (Hof et al., 2020).

Early diagnosis of ASD enables the initiation of an early, appropriately personalized therapy. The younger the child, the better the results of therapy can be achieved in the area of communication and social interaction, cognitive abilities, speech development, or behavior appropriate to the situation, which improves the quality of life of individuals with ASD, reduces the risk of mental disorders and significantly reduces the burden of ASD (Lai et al., 2019; MacDonald et al., 2014; Orinstein et al., 2014; Smith et al., 2015).

For this reason, the American Academy of Pediatrics recommends screening at 18 and 24 months of age as part of primary care during well-child care visits. (Hyman et al., 2020). There is evidence suggesting that including screening tools in routine medical appointments may result in earlier and more accurate identification of children who need further help than if solely clinical impressions are relied on, which is particularly important when care providers are less experienced in diagnosing ASD (Sheldrick et al., 2011). It appears that the increasing availability of screening significantly lowered the age of ASD diagnosis in the US, with diagnosis before the age of 4 made in 71 % of children (2018) compared to 58 % in 2014 (Maenner et al., 2020; Shaw et al., 2020). However, a US Preventive Services Task Force report shows insufficient evidence to recommend universal ASD screening (Siu et al., 2016). On the other hand, there is evidence suggesting that including screening tools in routine medical appointments may result in earlier and more accurate identification of children who need further help than if solely clinical impressions are relied on, which is particularly important when care providers are less experienced in diagnosing ASD (Sheldrick et al., 2011). Moreover, the use of public ASD screening may reduce social inequalities in terms of the age of diagnosis and access to further therapeutic activities (Coury, 2015; Fein et al., 2016). The conclusions of both reports indicate the need for further research on screening questionnaires and their effectiveness, and on the effectiveness of further proceedings after screening (Mandell & Mandy, 2015).

The Communication and Symbolic Behavior Scale-Developmental Profile (CSBS-DP) is one of the available tools for the early detection of symptoms of autism spectrum disorders. CSBS-DP was developed in the United States in 1993 and had good psychometric properties (i.e. reliability and validity) (Wetherby et al., 2002, 2004, 2008). One part of the CSBS-DP, the Infant-Toddler Checklist (ITC), is a 24-item questionnaire for parents or caregivers. The included questions are arranged into three composites (social, speech and symbolic composite) and seven development predictors (emotion and eye gaze, communication, gestures, sounds, words, understanding and object use). CSBS-DP ITC can be used in universal ASD screening of children aged 6 to 24 months in a primary care setting to select children who will need to take further diagnostic steps to confirm or exclude the diagnosis of ASD.

This study aims to validate the Polish version of the CSBS-DP ITC, primarily because cultural differences occur in different contexts in different populations. Therefore, it is vital to make cultural adaptations tailored to a particular population to increase the accuracy of screening tools (Soto et al., 2014). The purpose of the validation was to determine the validity, factor structure, invariance in sex groups, internal consistency and stability of Polish version of the CSBS-DP ITC.

Moreover, at the time of writing, no scientific publications on Polish versions of other ASD screening tools have been published. Due to the high psychometric properties of the original version, short time needed for caregivers to complete the questionnaire and versatile possibilities to conduct the examination (e.g. in physician's office, community setting or daycare setting and may be performed by nurses or trained paraprofessionals) we found CSBS-DP ITC as a supposedly useful and effective tool in everyday practice. In addition, it has been designed for use in children from 6 months of age, which potentially allows for a significant acceleration of the time of diagnosis of ASD in children.

Approval from the Bioethics Committee of the Wroclaw Medical University was obtained to conduct the study (number KB – 641/2020). All procedures were performed in accordance with the 1964 Helsinki declaration and its later amendments.

2. Methods

2.1. Measures

2.1.1. The original version of CSBS-DP ITC

As mentioned earlier, the CSBS-DP ITC questionnaire by Wetherby and Priznant is one of the ASD screening tools consisting of 24 questions that address seven predictors of child development (emotions and eye contact, communication, gestures, sounds, words, understanding, use of objects), combined in a total of three subscales (social skills, symbolic skills and speech). Assessment of child development using the CSBS-DP ITC is based on the total point result and the assessment of the result in three mentioned above subscales. The cut-off points for the need for further evaluation of the child were a score below 1.25 SD for the total, social or symbolic skills scores achieved comparing with the standardization group (3026 children from the general population participated in the original validation study) (Wetherby et al., 2008). A score below the cut-off point for the speech subscale indicates the need for close observation of the child and re-evaluation within three months. The original version of CSBS-DP ITC has a sensitivity and specificity of 0.89, a positive predictive value of 0.94 and a negative predictive value of 0.80. The reliability of the tool measured by Cronbach's alpha is 0.87 (Wetherby et al., 2002).

The research showed that the result obtained in the CSBS-DP ITC questionnaire could confidently predict the level of language development two years later and is an effective tool for the diagnosis of children with special needs (Wetherby et al., 2004). CSBS-DP ITC is one of five tools recommended by the Centers for Disease Control and Prevention for ASD screening in the United States (Centers for Disease Control and Prevention, 2020). So far, this questionnaire has also been adapted in Australia (Eadie et al., 2010), Croatia (Cepanec et al., 2012), South Korea (*only Behaviour Sample*) (Lee et al., 2018) and Taiwan (Lin et al., 2015).

2.1.2. Preparation of the Polish version of the CSBS-DP ITC

In order to prepare the Polish version of the CSBS-DP ITC questionnaire, the authors of the original version were contacted, and their consent was obtained for the adaptation to Polish conditions. Two English translators and one researcher were asked to translate the questionnaire into Polish, and then one version of the questionnaire was jointly created. Then, an English native speaker fluent in Polish was asked to translate the Polish version prepared in this way back into English to check if there were significant differences in content after the translation process.

Due to significant differences in the phonemic structure of Polish (which is a language from the Slavic language group) and English (a language from the Germanic language group), it was decided, after consultation with speech therapists, to change the wording of the two questions – in question 15 one of the combined phonemes was removed ("uh oh") and "bye bye" was changed to "pa pa" (Polish version of farewell). Similarly, in question 16, the pronounced sounds were matched with those that Polish children learn first (i.e. "ma, na, ba, da, ga, ka, la, ja, ta, pa"). Phrases mentioned in questions 17 and 18 were adjusted to the most frequently observed among Polish children (Boruta & Jastrzebska, 2012). The remaining questions were translated as faithfully as possible, adjusting their syntax

Table 1
Sociodemographic Characteristics of Participants.

Characteristic	Screening		Control	
	n	%	n	%
Sex				
Female	625	42.78	206	42.04
Male	836	57.22	284	57.96
Preterm born				
> 34 weeks	28	1.92	10	2.04
34-37 weeks	70	4.79	23	4.69
Medical conditions				
Serious genetic disorders	14	0.96	8	1.63
Serious health problems	39	2.67	22	4.49
Sight problems	23	1.57	5	1.02
Hearing problems	11	0.75	2	0.41
Muscle tone or other musculoskeletal system disorders	259	17.73	114	23.27
Physical rehabilitation in the past	568	38.88	214	43.67
ASD in closest family	42	2.87	26	5.31
Place of residence				
Village	332	22.7	98	20
Town inhabited by less than 20,000 people	95	6.5	37	7.55
City inhabited by 20,000-100,000 people	276	18.89	80	16.33
City inhabited by more than 100.000 people	758	51.88	275	56.12

to the Polish grammar. Experts assessing children's development were asked to evaluate the comprehensibility of the questions in Polish and did not raise any objections. The questionnaire was accompanied by a question regarding the understandability of the questions – only one parent raised an objection about question 23 (regarding the arrangement of blocks or rings).

2.2. Procedure and participants

2.2.1. Screening procedure

The participants were children aged 6 to 24 months and their parents or caregivers living in Poland and speaking Polish as their primary language, who gave their informed consent to participate in the study and completed the CSBS-DP ITC questionnaire and a personal questionnaire. No other exclusion criteria were used. Due to the COVID-19 pandemic and difficulties in conducting such a study in stationary health care clinics, an electronic version of the questionnaire was prepared and made available on the project's website examining the properties of the tool (*Spojrzeć w Oczy*, 2020). Study recruitment began on October 25, 2020 and ended on February 18, 2021. A total of 1732 parents expressed their willingness to participate in the study, of which 1461 children were enrolled after excluding repeated submissions and children who did not meet the age criteria. The number of participants significantly exceeds the minimum sample size to have a confidence level of 95 % and a margin of error of 5 % in the Polish population (the calculated minimum sample size was 139) (Schmidt et al., 2018).

The vast majority of questionnaire forms were filled in by the children's mothers (N = 1449, 99.18 %) – fathers filled in only 7 of them, and the rest were completed by another person from the family or a legal guardian. The mean age of children was 14.94 months (SD = 4.98), while the average age of mothers was 30.73 years (SD = 3.86), and of fathers – 32.81 years (SD = 4.68). Parents living in urban areas accounted for a greater percentage of the respondents – for example, almost 52 % of the surveyed parents came from large cities, whose inhabitants constitute approx. 28 % of the population in Poland (Polish Central Statistical Office, 2020). Sample characteristics are presented in Table 1.

2.2.2. Control Procedure

As recommended by the authors of the original version, three months after the first completion of the CSBS-DP ITC questionnaire, the parents of children aged 6 to 21 months at the time of the first measurement (9–24 months at the time of control measurement) were asked to re-complete the questionnaire (older children exceeded the recommended age). It was ensured that the person completing the questionnaire was the same parent who filled it in for the first time. Four hundred and ninety children out of 1313 participated in this part of the study, giving a response rate of 37.32 %. Data on study participants were displayed on a chart (Fig. 1).

Both in the first screening and the control, American cut-offs were used to determine the possible risk of ASD in the examined children due to the lack of scoring cut-offs suited to the Polish population. Parents were made aware of the use of cut-offs from a different population and were given the opportunity to consult their concerns about their children's development with those in charge of the project.

2.2.3. Interrater Reliability Evaluation Procedure

In November 2021, an additional 200 pairs of parents whose children attend nurseries in Wrocław, Poland, were asked to complete the CSBS-DP ITC questionnaires to obtain the results for the same child, but this time assessed by both parents/caregivers. Out of this group, 112 couples responded positively to the survey and returned questionnaires. In this part of the study, paper versions of the questionnaire were used.

2.2.4. Statistical Analyses

All the analyses presented in this paper were performed using JASP 0.1.11. First, the factor structure of the test was examined and

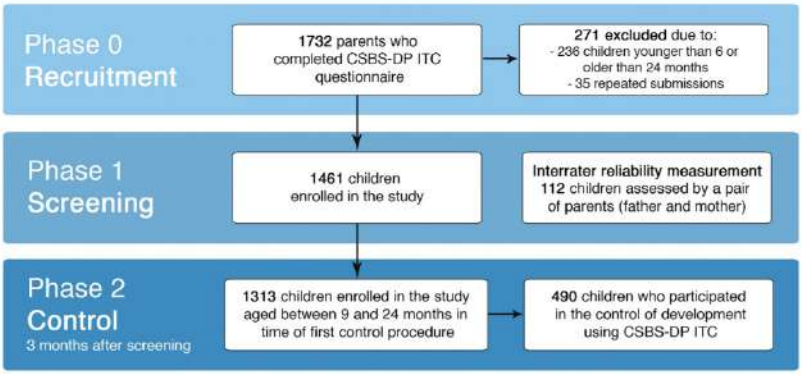

Fig. 1. Chart showing the number of study participants at each stage.

the goodness of fit of the one-factor and three-factor models was checked to determine which one had the best fit to the data in the Polish sample. Therefore, two separate confirmatory factor analyses (CFAs) were conducted. A diagonally weighted least squares (DWLS) estimation method was applied to compute the parameters of the models. This method is considered appropriate for ordinal and non-normal data (Christoffersson, 1975; Joreskog & Sorbom, 1982). As for indicators of goodness of fit, the following indicators were calculated: comparative fit index (CFI), Tucker–Lewis index (TLI; in both cases, values >.90 were considered acceptable) and root mean square error of approximation (RMSEA; with values <.08 considered as satisfactory; see Hu & Bentler, 1998). Also, the values of the chi-square test (together with their statistical significance) were reported.

After that, a multigroup CFA was run to determine the goodness of fit of the two models in samples of male and female subjects and to check whether the test was decoded similarly for distinct gender groups. Thus, we examined the measurement invariance of the scale. Testing measurement invariance is paramount in validating or adapting measures into new contexts. Without evidence of measurement invariance across different (primarily demographic - but not only) groups, the instrument may be sensitive to bias, as it is not interpreted equivalently in those groups. For example, if respondents react differently to the content of a given measure (e.g., items wording, concepts within items), the instrument will not be equivalent to individuals from diverse backgrounds (Holden et al., 2020).

The estimation method used in that analysis was identical to that described previously. Four levels of measurement invariance were tested: configural (identical factor structure), weak (equality of factor loadings), strong (equivalence between latent means) and strict invariance (invariance in residual variances) (Brown, 2015). It was assumed that configural invariance is supported if the same factor structure simultaneously presents a satisfactory fit for both groups. The adequacy of the following restricted models was compared in terms of their fit indices values. A change in both RMSEA and CFI coefficients was considered, as those criteria are less sensitive to sample size distortion (e.g., comparing to the $\Delta\ \chi 2$). If the drop in CFI of the constrained model relative to the unconstrained model does not exceed.002, and simultaneously the increase in RMSEA does not exceed.007, the constrained model is accepted (Meade et al., 2008).

To assess the questionnaire's reliability, Cronbach's α coefficients and McDonald's omega total coefficients were computed. Also, to provide more comprehensive information about the reliability of the test, Pearson's r correlation coefficients were computed to check the test-retest reliability of the given test. Also, to examine the interrater reliability of the results, intraclass correlation coefficients were calculated.

Furthermore, the mean differences between female and male participants were tested to assess validity. Earlier studies showed that females were less likely to be screen positive using ASD screening tools, which probably may result from a certain bias in relation to "more pronounced" ASD symptoms among boys - such reports, however, have not been credibly confirmed in scientific research (Giarelli et al., 2010; Santos et al., 2022; Wiggins et al., 2021). It was also associated with a lower frequency of receiving invitations to development evaluation compared to boys. Females at high likelihood for ASD were less likely to be diagnosed with autism, which reduced the accuracy of the tools and led to delays in the diagnosis of this group (Eldeeb et al., 2023). In order to compare sex differences, we used the measurement of invariance in sex groups and the analysis of the differences in the results achieved between boys and girls in different age groups.

The aim of this analysis was to show whether the Polish version of the CSBS-DP ITC questionnaire can be resistant to potential sex differences during early development, e.g. differences in speech development and possible specification of the age of children most at risk of omission.

Due to the insufficient number of participants to conduct analyzes in particular months of age, it was necessary to group children into larger age cohorts. The idea was to group the children in such a way that each cohort would collect children presumably at a similar level of development. For this purpose participants were divided into four age groups based on the substages of Piaget's Sensorimotor Stage. The first group, "Secondary circular reactions", included children aged from 6 to 8 months of age, the second group, "Coordination of secondary schemes", included children between 9 and 12 months, the third group, "Tertiary circular reactions", included children between 13 and 18 months, and the fourth group, "Beginning of mental representation", included children above 19 months. Although Piagetian theory has its limitations, it is a good conceptual tool (Lourenço, 2016) for this analysis. The assumption of normality was not met in most of the variables; therefore, mean differences were tested using the bootstrap method for the independent samples test (Field, 2017).

Based on the results from the collected questionnaires, the average results achieved by Polish children in the CSBS-DP ITC questionnaire were calculated, as well as the standard deviation and 1.25 SD (which includes 10 % of the most extreme results and is the cut-off point in the original version). Then, the obtained results were compared to the results achieved by 2000 children from the U.S. standardization group. Unfortunately, due to the lack of response from the authors of the original version regarding the disclosure of the results obtained in the U.S. standardization group, the results were compared only in the groups from 12 to 24 months of age using the only available values, i.e. mean and standard deviation. Due to the lack of a normal distribution in the Polish population, medians were used for comparisons. Since the medians in the U.S. standardization group are not known, the mean values were used as the medians (because if the distribution is normal or at least symmetrical, the mean is also the median). Then the one-sample median of the Wilcoxon test was used to compare both medians (in the Polish and U.S. groups). Unfortunately, the assumption that the medians in the American population are equal to the mean may be a limitation of this comparison.

3. Results

3.1. Factor structure

Confirmatory factor analysis was performed to examine the factor structure of the Polish adaptation of the test. Two models were

tested: the one-factor model (general scores) and the three-factor model (with the following sub-scales referring to the three different groups of skills: Social, Speech and Symbolic). The results of the analysis are presented in Table 2.

The results indicated a good fit for both proposed models. In the one-factor model, all items were significantly associated with the general latent factor (all $p < .001$; fully standardized regression weights ranging from.146 to.894 – for more information, see Table 3). Also, in the three-factor model, all items were significantly related to the given sub-factor latent factor (all $p < .001$; fully standardized regression weights ranging from.152 to.926 – for more information, see Table 3).

3.2. Measurement invariance in sex groups

The measurement invariance of the test was examined, comparing results obtained for males and females. In this case, measurement invariance was conducted as a replication test to ensure that the one-factor and three-factor models proposed previously work equivalently across different sex subsamples. The results of the analysis are presented in Table 4.

Both the one-factor and three-factor models showed an acceptable fit in both subsamples. Hence, configural invariance was supported for both proposed models. However, only regarding the one-factor model, all levels of measurement invariance could have been assumed across both groups. In this case, changes in both RMSEA and CFI were satisfactory and met the benchmarks assumed in the study. Hence, the results supported the stability of the proposed factor structure in male and female subsamples. On the other hand, in the case of the three-factor model, the metric level of measurement invariance could not have been assumed across both groups, as the changes in both RMSEA and CFI were significant, suggesting the high distortion in the goodness of fit. Hence, the results did not support the stability of the three-factor structure in male and female subsamples.

3.3. Internal consistency and stability

Descriptive statistics (mean, standard deviation), as well as reliability coefficients for CSBS-DP ITC, are given in Table 5. The total score of CSBS-DP ITC demonstrated satisfactory internal consistency, Cronbach's $\alpha = .92$ [95 % CI:.912;.924], McDonald's $\omega = .92$. The average inter-item correlation was $r = .32$, while the average item-rest correlation was $r = .54$; both were satisfactory. Composite scale reliability coefficients were also acceptable.

Test-retest correlations were also tested to assess the stability of the scores. Pearson's correlation coefficients are presented in Table 6. All the correlation coefficients between the first and second measurements were mostly above.70, proving a satisfactory level of stability.

3.4. Interrater reliability

Interrater reliability was assessed using intraclass correlation coefficients (ICC) based on ratings made by two different caregivers (P1 – primary caregiver, usually the mother; P2 – secondary caregiver, usually the father). The analysis utilized ICC*(3, B)* as referenced by Shrout and Fleiss (1979). The results presented in Table 7, demonstrate a high level of interrater reliability across all domains, confirming the strong consistency between primary and secondary caregiver evaluations.

3.5. Sex differences in the results of the Polish version of the CSBS-DP ITC questionnaire

To assess whether in the Polish version of the CSBS-DP ITC questionnaire there is an effect of gender differences in the results achieved by children (characterized by higher results achieved by girls on average) the mean differences between female and male participants were tested. The results for individual age groups based on the substages of Piaget's Sensorimotor Stage are presented in Tables 8–11.

Results presented in Tables 8–11 suggest that females between 9 and 12 months of age achieve higher results in the Social Composite (Gestures), Speech Composite (Sounds) and Total Score. Between 13 and 18 months, higher scores achieved by females in CSBS-DP ITC can be spotted in every domain, apart from Emotion and Communication. After 19 months of age, statistical differences in the social composite (total Social score) and Object use disappear.

3.6. Mean scores and comparison with the scores of the U.S. standardization group

Tables 12–15 with mean scores and their comparison with the results of the American standardization group were provided as

Table 2
Goodness of fit of one-factor and three-factor model: confirmatory factor analysis.

Measure	χ^2 (*df*)	χ^2 /*df*	CFI	TLI	RMSEA	90 % CI RMSEA
One-factor model	1188.19*** (252)	4.72	.98	.98	.050	.048;.053
Three-factor model	973.72*** (249)	3.91	.99	.99	.045	.042;.048

Note. TLI = Tucker-Lewis index, CFI = Comparative Fit Index; RMSEA = root mean square error of approximation; RMSEA 90 % CI = 90 % confidence interval of the RMSEA; χ2 (df) = chi-square with degrees of freedom.
* ** $p < .001$

Table 3
Factor loadings for the one-factor and three-factor model: confirmatory factor analysis.

Item	One factor model			Three factor model			
	Standardized estimate	Z	p	Factor	Standardized estimate	Z	p
CS1	0.15	15.02	< .001	Social	0.16	15.02	< .001
CS2	0.28	28.88	< .001		0.29	29.03	< .001
CS3	0.15	15.22	< .001		0.15	15.43	< .001
CS4	0.61	62.29	< .001		0.63	61.65	< .001
CS5	0.54	53.51	< .001		0.56	53.19	< .001
CS6	0.26	27.21	< .001		0.27	27.34	< .001
CS7	0.53	59.27	< .001		0.54	58.80	< .001
CS8	0.65	75.02	< .001		0.67	73.87	< .001
CS9	0.72	72.45	< .001		0.74	71.43	< .001
CS10	0.62	65.90	< .001		0.64	65.31	< .001
CS11	0.67	75.86	< .001		0.69	74.47	< .001
CS12	0.79	85.34	< .001		0.81	83.16	< .001
CS13	0.64	80.42	< .001		0.65	78.62	< .001
CS14	0.42	42.04	< .001	Speech	0.44	40.62	< .001
CS15	0.56	57.32	< .001		0.61	54.86	< .001
CS16	0.68	69.81	< .001		0.75	63.87	< .001
CS17	0.83	94.40	< .001		0.93	74.73	< .001
CS18	0.48	61.77	< .001		0.54	57.38	< .001
CS19	0.37	36.87	< .001	Symbolic	0.37	36.27	< .001
CS20	0.89	96.70	< .001		0.91	87.78	< .001
CS21	0.19	18.69	< .001		0.19	18.60	< .001
CS22	0.82	84.47	< .001		0.83	78.85	< .001
CS23	0.76	86.53	< .001		0.77	80.75	< .001
CS24	0.74	90.79	< .001		0.75	84.89	< .001

Note. All factor loadings are presented as fully standardized estimates.

Table 4
Measurement invariance of the one-factor and three-factor models across sex subsamples.

	Model	χ^2 (*df*)	RMSEA	RMSEA 90 % CI	Δ RMSEA	CFI	Δ CFI
One-factor model	Configural	1609.49*** (528)	.053	.050;.056	-	.980	-
	Metric	1534.11*** (527)	.051		.002	.981	.001
	Scalar	1571.15*** (550)	.050	.048;.053	.001	.981	.000
	Strict	1628.47*** (574)	.050	.047;.053	.000	.981	.000
Three-factor model	Configural	1041,23*** (498)	.039	.035;.042	-	.990	-
	Metric	1271.24*** (519)	.045	.041;.048	.006	.986	.004
	Scalar	1303.52*** (540)	.044	.041;.047	.001	.986	.000
	Strict	1350.43*** (564)	.044	.041;.047	.000	.986	.000

Note. CFI = Comparative Fit Index; RMSEA = root mean square error of approximation; RMSEA 90 % CI = 90 % confidence interval of the RMSEA; χ2 (df) = chi-square with degrees of freedom; Δ = change from previous model.
* p < .05; * * p < .01; * ** p < .001

Table 5
Descriptive statistics and reliability coefficients.

CSBS DP ITC			Reliability			
	M	*SD*	α	ω	Average interitem correlation	Average item-rest correlation
Social	18.16	5.26	.86	.87	.29	.50
Speech	8.42	3.27	.78	.86	.43	.60
Symbolic	11.67	4.27	.82	.90	.39	.59
Total	38.25	11.79	.92	.92	.32	.54

Supplementary File 1. Among the compared 13 age categories (from 12 to 24 months of age), Polish children achieved statistically significantly lower results in 6 of them in terms of composite "Social"; in 7, in composite "Speech" and higher results were recorded in 11 age categories in composite "Symbolic". The median of the total score differed statistically significantly only in the 14 and 16-month age groups.

4. Discussion

Worldwide, there are at least 26 questionnaires designed for the early detection of autism spectrum disorders in primary health care

Table 6
Test-retest stability - Pearson's correlation coefficients.

Variables	Pearson's r
Social 1st-Social 2nd	0.80 * **
Speech 1st-Speech 2nd	0.78 * **
Symbolic 1st-Symbolic 2nd	0.83 * **
Total 1st-Total 2nd	0.83 * **

* ** p < .001

Table 7
Interrater reliability - Intraclass correlation coefficients.

Type	Point Estimate	Lower 95 % CI	Upper 95 % CI
Speech	0.943	0.918	0.960
Social	0.916	0.880	0.941
Symbolic	0.969	0.955	0.979
Total	0.968	0.954	0.978

Note. 112 subjects and 2 raters/measurements. ICC type as referenced by Shrout and Fleiss (1979).

Table 8
Differences in the results achieved in the Polish version of CSBS-DP ITC questionnaire depending on the child's sex in the age subgroup from 6 to 8 months of age.

	Female (N = 75)		Male (N = 89)		*t*	*df*	Mean Difference	Bootstrap 95 % CI		Cohen's *d*	95 % CI	
	M	SD	M	SD				Lower	Upper		Lower	Upper
Emotion	6.29	1.35	6.01	1.32	-1.35	155.87	-.28	-.68	.13	-.21	-.52	.10
Communication	3.59	1.56	3.51	1.42	-.35	151.09	-.08	-.60	.38	-.05	-.36	.25
Gestures	.97	1.75	.64	1.10	-1.45	122.21	-.33	-.86	.10	-.24	-.54	.07
Social	10.85	3.65	10.16	2.95	-1.33	141.81	-.70	-1.80	.29	-.21	-.52	.10
Sounds	4.24	1.80	4.24	1.79	-.01	157.04	.00	-.53	.57	.00	-.31	.30
Words	.25	.72	.12	.39	-1.40	110.26	-1.13	-.33	.05	-.23	-.54	.08
Speech	4.49	2.18	4.36	1.98	-.41	151.04	-.13	-.76	.57	-.06	-.37	.24
Understanding	1.89	1.17	1.84	1.03	-.29	149.08	-.05	-.39	.29	-.05	-.35	.26
Object use	3.07	1.46	3.09	1.20	.11	143.69	.02	-.41	.43	.02	-.29	.32
Symbolic	4.96	2.35	4.93	1.82	.08	138.05	.03	-.75	.59	.01	-.32	.29
Total	20.31	7.22	19.45	5.21	.857	131.89	.86	2.93	1.06	.14	.45	.17

Note. Bootstrap results are based on 10 bootstrap samples. * p < .05; * * p < .01; * ** p < .001

Table 9
Differences in the results achieved in the Polish version of CSBS-DP ITC questionnaire depending on the child's sex in the age subgroup from 9 to 12 months of age.

	Female (N = 150)		Male (N = 181)		*t*	*df*	Mean Difference	Bootstrap 95 % CI		Cohen's *d*	95 % CI	
	M	SD	M	SD				Lower	Upper		Lower	Upper
Emotion	6.77	1.10	6.62	1.11	-1.17	318.71	-.14	-.38	.09	-.13	-.35	.09
Communication	4.77	1.76	4.43	1.86	-1.72	323.33	-.34	-.72	.04	-.19	-.41	.03
Gestures	4.46	2.72	3.82	2.48	-2.21 *	305.30	-.64	-1.21	-.10	-.25	-.46	-.03
Social	16.00	4.62	14.88	4.48	2.23 *	313.86	-1.12	2.09	-.19	.25	-.46	.03
Sounds	5.95	1.38	5.57	1.57	2.33 *	328.02	-.38	-.69	.07	.25	-.47	.04
Words	.93	.92	.79	.88	1.37	311.37	-.14	-.35	.06	-.15	-.37	.06
Speech	6.87	1.97	6.36	2.13	-2.28 *	325.09	-.51	-.95	-.10	-.25	-.47	-.03
Understanding	3.30	1.32	3.18	1.23	-.83	308.07	-.12	-.41	.15	-.09	-.31	.12
Object use	5.34	2.03	5.13	1.83	-.99	303.84	-.21	-.64	.18	-.11	-.33	.11
Symbolic	8.64	2.99	8.31	2.69	-1.05	303.28	-.33	-.98	.28	-.12	-.33	.10
Total	31.51	8.52	29.555	8.00	-2.15 *	309.43	-1.97	-3.80	-.26	-.24	-.46	-.02

Note. Bootstrap results are based on 10 bootstrap samples. * p < .05; * * p < .01; * ** p < .001

settings (Sobieski et al., 2022). However, none of them has been fully adapted to Polish conditions so far – in 2022, the first study on the Polish version of the Qualitative Checklist for Autism in Toddlers questionnaire was published (Niedźwiecka & Pisula, 2022). In Poland, the only validated questionnaire that could be used as a screening tool is the Autism Spectrum Rating Scales (ASRS), available only to psychologists or graduates of appropriate studies with additional training in psychometrics. Additionally, ASRS is not available

Table 10
Differences in the results achieved in the Polish version of CSBS-DP ITC questionnaire depending on the child's sex in the age subgroup from 13 to 18 months of age.

	Female (N = 225)		Male (N = 326)		*t*	*df*	Mean Difference	Bootstrap 95 % CI		Cohen's *d*	95 % CI	
	M	SD	M	SD				Lower	Upper		Lower	Upper
Emotion	7.14	.90	7.04	1.05	1.17	522.51	.10	-.27	.08	.10	-.27	.07
Communication	5.84	1.63	5.67	1.64	1.14	483.40	.16	-.46	.12	.10	-.27	.07
Gestures	7.40	1.85	6.71	2.22	4.00 * **	530.12	.70	1.04	.34	.34	.51	.16
Social	20.38	3.59	19.42	4.05	2.91 * *	515.58	.95	1.63	.30	.25	.42	.08
Sounds	6.71	1.33	6.42	1.44	2.42 *	505.02	.29	.52	.07	.21	.38	.04
Words	2.44	1.46	2.12	1.36	2.57 * *	459.12	.32	.57	.09	.23	.40	.06
Speech	9.14	2.52	8.54	2.53	2.77 * *	482.33	.61	1.04	.19	.24	.41	.07
Understanding	4.88	1.13	4.53	1.33	3.33 * **	526.35	.35	.55	.14	.28	.45	.11
Object use	8.57	1.88	8.05	1.93	3.18 * **	490.85	.52	.84	.20	.27	.45	.10
Symbolic	13.45	2.64	12.58	2.88	3.67 * **	507.69	.87	1.32	.42	.31	.48	.14
Total	42.97	7.22	40.54	7.80	3.72 * **	511.45	2.43	3.77	1.13	.32	.49	.15

Note. Bootstrap results are based on 10 bootstrap samples. * $p < .05$; * * $p < .01$; * ** $p < .001$

Table 11
Differences in the results achieved in the Polish version of CSBS-DP ITC questionnaire depending on the child's sex in the age subgroup from 19 to 24 months of age.

	Female (N = 175)		Male (N = 240)		*t*	*df*	Mean Difference	Bootstrap 95 % CI		Cohen's *d*	95 % CI	
	M	SD	M	SD				Lower	Upper		Lower	Upper
Emotion	7.13	.99	7.12	1.06	.05	389.08	.01	-.20	.19	.01	-.20	.19
Communication	6.19	1.39	6.26	1.45	.49	384.38	.07	-.22	.35	.05	-.15	.24
Gestures	8.16	1.70	7.61	2.12	2.92 * *	409.23	.55	.91	.19	.28	.48	.09
Social	21.48	3.45	21.00	3.96	1.33	399.97	.48	1.19	.22	.13	.32	.07
Sounds	7.29	1.28	6.94	1.44	2.61 * *	397.31	.35	.61	.08	.26	.45	.06
Words	4.29	1.63	3.62	1.74	3.98 * **	388.01	.67	.99	.35	.39	.59	.20
Speech	11.58	2.70	10.56	2.94	3.64 * **	392.22	1.02	1.56	.47	.36	.55	.16
Understanding	5.51	.81	5.24	1.13	2.81 * *	412.90	.27	.45	.09	.27	.46	.07
Object use	10.02	1.41	9.75	1.45	1.88	381.12	.27	.53	.01	.19	.38	.01
Symbolic	15.53	2.03	14.99	2.28	2.51 *	396.43	.53	.95	.12	.25	.44	.05
Total	48.58	7.06	46.55	7.97	2.74 * *	397.50	2.03	3.43	.61	.27	.46	.07

Note. Bootstrap results are based on 10 bootstrap samples. * $p < .05$; * * $p < .01$; * ** $p < .001$

in open access and a user fee has to be paid.

The lack of available tools for the early diagnosis of ASD is the main obstacle to starting a discussion on introducing the universal screening of developmental disorders in the Polish population of children and infants. Probably due to the lack of readily available tools for the screening and diagnosis of ASD, the reported prevalence of ASD in Poland is lower than in neighboring countries (e.g. in Germany - 0.38 %) and lower than assumed, based on newer data from European Union countries (Bachmann et al., 2018; Salari et al., 2022). It also leads to a delay in the diagnosis of developmental disorders in children and, consequently, to poorer outcomes. Lack of appropriate therapy leads to an increased indirect financial burden for families due to the necessity to provide further special education, problems with finding employment and the necessity to provide increased care by other family members. Early and appropriate access to early intervention is known to improve long-term outcomes and reduce lifetime costs on the part of the individual, family and society (Horlin et al., 2014; Sampaio et al., 2021).

Early detection of ASD symptoms allows the implementation of further, more detailed diagnostics in the risk group, as well as the initiation of early intervention. For this purpose, the authors aimed to provide Polish parents, but also their children's doctors, with such an opportunity. CSBS-DP ITC was chosen due to its good psychometric properties in the original research and the possibility of assessing three different components of a child's development. It does not require the use of additional items by the examiner and covers a wide age range (from 6 to 24 months of age). As a screening tool for one-year-old child screening, ITC was found to be a promising method for healthcare professionals to identify children needing further evaluation for ASD and those with other developmental delays (Pierce et al., 2011). Presumably, the Polish version of the CSBS-DP ITC questionnaire will be similarly effective in the detection of children who require further diagnosis due to obtaining even higher psychometric values than the original version.

The analysis of the collected data has proved that the one-factor structure of the questionnaire is a more reliable indicator than the three-factor structure based on the CSBS-DP ITC subscales nevertheless both models showed an acceptable fit in both subsamples. Hence, configural invariance was supported for both proposed models. However, only regarding the one-factor model, all levels of measurement invariance could have been assumed across both groups. In this case, changes in both RMSEA and CFI were satisfactory and met the benchmarks assumed in the study. The results supported the stability of the proposed factor structure in the male and female subsamples. On the other hand, in the case of the three-factor model, the metric level of measurement invariance could not have

been assumed across both groups, as the changes in both RMSEA and CFI were significant, suggesting a high distortion in the goodness of fit. Considering that, the results did not support the stability of the three-factor structure in the male and female subsamples.

This indicates that decisions as to further steps (i.e. either to refrain from further diagnosis or refer the child to further psychological or medical examinations) are safer to make based on the total score than based on lowering any subscale score. Nevertheless, the reliability coefficients for both the one- and three-factor models are very high and indicate a proper fit of the questionnaire to the diagnostic needs of the population of Polish children. The results of three-factor analyzes can therefore be used in early intervention to identify the first areas that require therapeutic action. Although the authors of the original version do not recommend using single-question responses to develop action plans based on the difficulties reported in the questionnaire by parents, due to the high rates in the conducted CFA, it seems that individual responses obtained in the CSBS-DP ITC could be helpful in developing therapeutic plans. When attempting to use the questionnaire in such a way, it should be borne in mind that items 1, 3, 6 and 21 have low values of standardized estimate and it may be a slight drawback of the questionnaire.

During the validity analysis, girls were observed to be achieving higher scores in the questionnaire. This may be due to the fact that some of the questions in the questionnaire refer to behavior significantly more often observed in boys. A study of sex differences and diagnosis from early parent concerns shows that males with ASD had a higher likelihood of repetitive behavior and speech and language concerns compared to females with ASD (Wallisch et al., 2021). On the other hand, females with developmental delays were more likely to have trouble with problem-solving. Data from another study conducted among toddlers and preschoolers showed that girls diagnosed with ASD had higher severity of symptoms of social skills than boys (Ros-Demarize et al., 2020). Similar differences are also visible in later age - among adolescents ASD girls and boys differ especially significantly in communication section (Rynkiewicz & Lucka, 2018). The imperfections of the existing questionnaires for early ASD screening (including CSBS-DP ITC) may under-identify ASD symptoms in toddler and preschool-aged girls.

When comparing the Polish and American populations, there is a slight difference in the number of points obtained by children at a particular age – children from the U.S. normative group achieved significantly higher results in social skills and speech than Polish children of the same age. On the other hand, children from the Polish normative group achieved higher scores in symbolic skills. Unfortunately, based on the data collected by the authors of this study, the reason for this difference could not be fully explained. One of the possible reasons for such an effect is the difference in time when the standardization studies were carried out. Data for the American normative group was collected in 2002, which is before smartphones and other handheld electronic devices became popular. Available studies suggest that excessive exposure of children to TV screens and telephones was highly prevalent and independently associated with poorer development outcomes among children (Rocha et al., 2021; Zimmerman et al., 2007). The second potential explanation is the impact of the COVID-19 pandemic on the deterioration of children's development – there are concerns that reducing interpersonal contact may adversely affect the development of social skills and speech (Charney et al., 2021; Marler & Ditton, 2021). Iverson et al. found that Italian children used gestures definitely more often than American, with the difference being inversely related to the size of the children's spoken vocabularies (Iverson et al., 2008). They also brought up the important point that children's gesture use reflected the environment where they were raised and input from their parents. Goldin-Meadow and Saltzman found that a difference in gesture rates between Taiwanese and American mothers could be accounted for by different child-rearing practices in the two cultures (Goldin-Meadow & Saltzman, 2000). Children raised in an environment with a high frequency of gestures tended to use more gestures compared to children raised in cultures where gesture use was less common.

Due to the lack of tools for very early diagnosis, there are only a few cases in Poland of children under 24 months of age who are diagnosed with autism spectrum disorders. Hence, it was difficult to reach a significant number of children with ASD in this age group and it was not feasible to estimate the criterion validity of the Polish CSBS-DP-ITC questionnaire.

4.1. Limitations of the study

The survey was largely carried out using the online version of the questionnaire –children were not recruited in primary care clinics, hence a group bias is possible (the project's website was visited mainly by concerned parents). However, every effort was made to convince also those parents who do not suspect any developmental disorders in their children to complete the questionnaire. It seems that these efforts were effective – 383 parents reported concerns about their child's development, which constitutes 25.7 % of all participants. This is perfectly in line with the data from a study conducted at the C.S. Mott Children's Hospital, where it was shown that a quarter of parents also report suspected developmental disorders in their children (Freed et al., 2021).

Unfortunately, the participants' percentages of the place of residence (cities vs. villages) could not be maintained in line with the data of the Polish Statistical Office. Most of the respondents were children and parents from large urban centers. As a result, children from rural areas are underrepresented in the study. Another limitation is still a relatively small group of respondents in extreme age groups (i.e. 6 and 24 months of age).

The study does not include measurements of the sensitivity and specificity of the questionnaire and established cut-off points – further observation of the study participants is necessary to assess their development.

5. Conclusions

The results of this research resonated with some of the results of previous original studies on the validity and reliability of the CSBS-DP ITC. The Polish version of CSBS-DP ITC is characterized with very high reliability results (both Cronbach's alpha and McDonald's omega), good fit of the factor structure and satisfactory test-retest stability results. Our preliminary results on psychometric values indicate the usefulness of the questionnaire in screening symptoms of autism spectrum disorders in the population of Polish children,

both males and females, specifically living in urban areas, and in highlighting children who require further diagnostic steps.

However, in order to confirm this hypothesis (and thus determine the sensitivity, specificity, positive and negative predictive values), further observation of the study participants is necessary. Nevertheless, the use of this questionnaire for screening in primary health care settings may contribute to a faster diagnosis, and thus – better outcomes.

CRediT authorship contribution statement

Aleksandra Sobieska: Writing – review & editing, Investigation, Formal analysis. **Kamila Pierchała:** Writing – review & editing, Writing – original draft, Methodology, Formal analysis, Data curation. **Anna Wołowicz:** Writing – review & editing, Investigation. **Katarzyna Podgórska:** Writing – review & editing, Investigation. **Maria Magdalena Bujnowska-Fedak:** Writing – review & editing, Writing – original draft, Validation, Supervision, Methodology, Investigation, Conceptualization. **Małgorzata Sekułowicz:** Writing – review & editing, Supervision, Methodology, Conceptualization. **Sylwia Wrona:** Writing – review & editing, Methodology, Investigation, Formal analysis. **Mateusz Sobieski:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Resources, Project administration, Methodology, Investigation, Funding acquisition, Data curation, Conceptualization. **Maria Flakus:** Writing – review & editing, Writing – original draft, Methodology, Formal analysis, Data curation.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data Availability

The data presented in this study are openly available in FigShare at https://doi.org/10.6084/m9.figshare.21314982.v1.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.rasd.2024.102454.

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10.3. Screening Accuracy and Cut-offs of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist

PLOS ONE

RESEARCH ARTICLE

Screening accuracy and cut-offs of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist

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Abstract

Background

The first stage of diagnosing autism spectrum disorders usually involves population screening to detect children at risk. This study aims to assess the predictive convergent validity of the Polish version of the Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC) with the Autism Spectrum Rating Scales (ASRS), evaluate its sensitivity and specificity and assess the cut-off points for the possibility of using this questionnaire in population screening among children aged 6 to 24 months.

Method

The study was conducted among 602 children from the general population who had previously participated in the earlier phase of validation of the questionnaire for Polish conditions. The collected data were statistically processed to calculate the accuracy (i.e. sensitivity, specificity) of the questionnaire.

Results

In individual age groups, the sensitivity of the questionnaire varies from 0.667 to 0.750, specificity from 0.854 to 0.939, positive predictive value from 0.261 to 0.4 and negative predictive value—from 0.979 to 0.981. Screening accuracy ranges from 0.847 to 0.923 depending on the age group. The adopted cut-off points are 21 points for children aged 9–12 months, 36 for children aged 13–18 months, 39 for children aged 19–24 months. Cut-off points could not be established for children aged 6–8 months. The convergent validity values with the ASRS ranged from -0.28 to -0.431 and were highest in the group of the oldest children.

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Competing interests: The authors have declared that no competing interests exist.

Conclusions

These results indicate that the Polish version of the CSBS-DP ITC can be used as an effective tool for ASD universal screening.

Introduction

Autism spectrum disorders (ASD) are neurodevelopmental conditions of undetermined and complex etiology, most often manifested in early childhood, which affect the everyday activities of individuals and are characterized primarily by difficulties in the sphere of communication and interpersonal interactions as well as restricted interests or repetitive behaviors [1]. The prevalence of ASD is not clearly defined—the World Health Organization (WHO) estimates that ASD affects 1 in 160 children worldwide [2]. More recent reports from a systematic review by Zeidan et al. estimate the worldwide prevalence of ASD as 1 in 100 children [3] at the same time indicating significant variability in prevalence depending on the country and research assumptions used in individual studies. According to the only official data from the Polish National Health Fund from 2012, the prevalence rate of ASD in individuals under the age of 18 in Poland is 3.4 cases per 10,000 children [4], but more accurate preliminary data from two Polish provinces estimate that ASD occurs in one in 286 children [5].

The first symptoms of ASD usually appear during early child development—prodromal symptoms may be visible as early as 6 months of age [6, 7]. Later in development (from 14 months to 3 years of age) communication and social behavioral symptoms become apparent [8]. It is also possible that development plateaus after a period when the age-appropriate milestones were achieved or that the skills acquired earlier are lost [9, 10]. It is assumed that a reliable diagnosis of ASD in a child can be made as early as the 2nd–3rd year of age [11]. However, due to delays associated with the diagnostic process (or its omission), this diagnosis is made much later—in a world-wide 2019 meta-analysis, the average age of ASD diagnosis is 60.48 months (range 30.90–234.57) [12].

Early diagnosis of ASD enables the initiation of an early, age-adjusted therapy of developmental delays and difficulties. The younger the child, the better the results of therapy can be achieved in the area of communication and social interaction, cognitive abilities, speech development, or behavior appropriate to the situation, which improves the quality of life of people with ASD, reduces the risk of mental disorders, and significantly reduces the burden of ASD [13–16].

Considering this, the American Academy of Pediatrics (AAP) recommends screening at 18 and 24 months of age as part of primary care during well-child care visits [17]. It appears that the increasing availability of screening significantly lowered the age of ASD diagnosis in the US, with diagnosis before the age of 4 made in 71% of children (2018) compared to 58% in 2014 [18, 19]. On the other hand, a 2016 report from the US Preventive Services Task Force shows insufficient evidence to recommend universal ASD screening [20]. However, there is evidence suggesting that including screening tools in routine medical appointments may result in earlier and more accurate identification of children who need further help compared to relying solely on clinical impressions, which is particularly important when care providers are less experienced in diagnosing ASD [21]. Moreover, the use of public ASD screening may reduce social inequities in terms of the age of diagnosis and access to further therapeutic activities [22, 23]. The conclusions of both reports indicate the need for further research on screening tools and their effectiveness, as well as on the effectiveness of further proceedings after screening [24].

Due to the lack of tools for early diagnosis of ASD available in Poland, the authors have prepared a linguistically and culturally adapted version of the original, American version of Communication and Symbolic Behavior Scales-Developmental Profile—Infant-Toddler Checklist (CSBS-DP ITC) [25]. CSBS-DP ITC is one of the available tools created for the early detection of symptoms of autism spectrum disorders. It is a 24-item questionnaire for parents or caregivers. The included questions are arranged into three composites (social, speech, and symbolic composite) and seven development predictors (emotion and eye gaze, communication, gestures, sounds, words, understanding, and object use). Original CSBS-DP ITC version can be used in universal ASD screening of children aged 6 to 24 months in a primary care setting [26]. The research showed that the result obtained in the original version of questionnaire could confidently predict the level of language development two years in advance and is an effective tool for the screening of children with special needs [25–27].

In order to conduct a validation study and disseminate knowledge about the research, the "Spojrzeć w oczy" (Pol. "Look into the eyes") project was established. The project aimed to determine the psychometric properties (e.g. validity and reliability, sensitivity and specificity) of the Polish version of the CSBS-DP ITC. The questionnaire was adapted and translated using the back translate method by three independent translators, and the questions included were adjusted to the phonetics of the Polish language and the speech development of Polish children. A detailed description of the preparation of the tool for Polish cultural conditions is included in the earlier publication on CSBS-DP ITC validation [28]. The Polish version of CSBS-DP ITC is included as S1 File. Questionnaire and its English re-translation as S2 File. Data from the earlier stage of the project indicate a very good fit of the one-factor and three-factor models in confirmatory factor analysis. The total score of the Polish version of CSBS-DP ITC demonstrated satisfactory internal consistency, Cronbach's $\alpha = .92$, and McDonald's $\omega = .92$. The stability of the measurement was confirmed by performing interrater and test-retest reliability analysis, proving perfect and satisfactory level of stability, respectively.

This study aims to assess the predictive convergent validity of the Polish version of the CSBS-DP ITC with the Autism Spectrum Rating Scales (ASRS) questionnaire, used by psychologists for screening children with suspected ASD and those at risk of it, evaluate the sensitivity and specificity of the Polish version of the CSBS-DP ITC and finally assess the cut-off points for the use of the questionnaire in population screening.

Methods

Participants

The participants were children aged 30 months and their parents or caregivers who participated in the earlier phase of the project by correctly filling in the Polish version of CSBS-DP ITC questionnaire when children were between 6 and 24 months of age. The condition for inclusion was living in Poland, speaking Polish as one's primary language, and giving informed, written consent to participate in the study. Project recruitment began on October 25, 2020 using advertisements placed in collaborating health care facilities and on social media, and ended on February 18, 2021. During this period, parents completed the Polish version of CSBS-DP ITC screening questionnaire. In addition, as part of the "Spojrzeć w oczy" project, materials on the symptoms, diagnosis and therapy of autism spectrum disorders were made available to parents and health care specialists—both in physical and online form [29]. The research phase lasted from April 28, 2021, to October 12, 2022 –until the last child included in the project turned thirty months of age. As some of the children (N = 34) were awaiting the final diagnosis, their parents were contacted by phone at a later date. By March 30, 2023, a final diagnosis was obtained in all of them, except one (due to diagnostic

difficulties, it was not possible to unambiguously confirm the presence of autism spectrum disorders in that particular case). During and after the data collection, only MS and SW had the opportunity to identify individual study participants—this was to enable the provision of further psychological and pedagogical assistance and further diagnostics.

Invitations to the follow-up were sent to all 1461 parents of children who were included in the first phase of the project. A total of 678 submissions were received back, 76 reports were excluded from the study– 48 of them because parents had not previously participated in Polish CSBS-DP ITC screening, and in 11 cases siblings were assessed in the follow-up instead of the child who was screened, and another 17 were repeated submissions. When attempts were made to contact the other parents via e-mail or telephone (N = 859), as many as 722 of them admitted that they did not complete the further part of the study due to the lack of symptoms in their children and hence—lack of willingness to remain in the study. Finally, 602 children were enrolled in the study, whose parents had fully completed the ASRS questionnaire and follow-up interview, and who had participated in an earlier CSBS-DP ITC screening, which gives a return rate of 41.2%. The number of participants significantly exceeds the minimum sample size to have a confidence level of 95% and a margin of error of 5% in the Polish population (the calculated minimum sample size was 139) [30]. The mean age of the children was 30.21 months (SD = 1.03). The vast majority of questionnaire forms were filled in by the children's mothers (N = 601, 99.83%)–only one of them was filled in by a father. Sample characteristics of the study participants are presented in Table 1. The individual phases of the project and the number of participants at each stage are collected in Fig 1.

Approval from the Bioethics Committee of the Wroclaw Medical University was obtained to conduct the study (number KB– 641/2020; the full text of the consent is available as S3 File and its translation—as S4 File). All study participants gave written consent to participate in the study. All procedures were performed in accordance with the 1964 Helsinki Declaration and its later amendments.

Evaluation instruments

Autism Spectrum Rating Scales (ASRS). ASRS is a set of questionnaires, used as an auxiliary tool in the diagnostic process or a screening tool, consisting of full and abbreviated versions of tests for younger children (2–5 years) and older children and adolescents (6–18 years), both for parents and teachers [31]. The ASRS was developed in 2009 and its structure corresponds to the diagnostic criteria of Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR) and International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10). The Polish version was prepared in 2016 and is characterized by high reliability in the version for parents and has a confirmed discriminatory, convergent, and differential validity [32]. Additionally, the Polish version corresponds to of Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) diagnostic criteria. In the study, the short version for younger children was used, which contains 15 items that best differentiate between children diagnosed with ASD and the comparative group from the non-clinical population. The analysis of the discriminant function for raw scores revealed that the indicators of correct classification ranged from 88.2% to 91.4%. Cronbach's alpha for the ASRS version used in the study is 0.85. Sensitivity and specificity are 87.8% and 83.5%, respectively.

Further evaluation instruments. As ASRS is a Level 2 ASD diagnostic tool, parents were asked for additional information to determine the need for further diagnosis. In order to further evaluate children's development, a short structured interview (follow-up interview) was created. They were also asked if their child had received an ASD diagnosis from a psychiatrist

Table 1. Sociodemographic characteristics of participants.

Characteristic	Follow-up	
	n	%
Sex		
Female	248	41.20
Male	354	58.80
Preterm born		
>34 weeks	1	0.16
34–37 weeks	17	2.82
Medical conditions		
Serious genetic disorders	4	0.66
Serious health problems*	24	3.99
Sight problems	5	0.83
Hearing problems	4	0.66
Muscle tone or other musculoskeletal system disorders	118	19.60
Physical rehabilitation in the past	240	39.86
ASD in closest family (first degree relatives)	7	1.16
Place of residence		
Village	140	23.25
Town inhabited by less than 20,000 people	54	8.97
City inhabited by 20,000–100,000 people	92	15.28
City inhabited by more than 100.000 people	316	52.49

Note.

*We asked parents to list comorbidities that seemed important to them as "serious health problems". At a later stage, we verified the answers, taking into account only those that, in our opinion, may have any impact on the child's development (examples of disorders reported by parents include neurological diseases, heart and kidney malformations, phenylketonuria, perinatal disorders, i.e. hypoxia, congenital adrenal hyperplasia, neuroblastoma, esophageal atresia, etc.)

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(understood as disorders included in the ICD-10 classification as F84.0, F84.1, or F84.5 or in the DSM-5 as autism spectrum disorder). They were also asked if their child had completed any of the available standardized diagnostic protocols available in Poland: Autism Diagnostic Interview-Revised (ADI-R), Psychoeducational Profile-3rd Edition-PL (PEP-3-PL), or Autism Diagnostic Observation Schedule, 2nd Version (ADOS-2) and what was the result of the conducted examination [33–35]. Information on the diagnostic provider's credentials and experience was collected when possible to avoid the possible impact of extraneous variables on the results. A positive response in terms of receiving a nosological diagnosis of ASD from a psychiatrist or a positive ADOS-2 test result meant qualification to the group of children diagnosed with ASD. If the child received another diagnosis (e.g. language delay—LD), this was also recorded in the database used in the study.

Procedures

Due to the COVID-19 pandemic and difficulties in conducting such a study face-to-face in healthcare clinics, an electronic version of the questionnaire was prepared and made available on the project's website examining the properties of the tool. Initially, the parents completed the Polish Version of CSBS-DP ITC questionnaire along with a short record of the mother's and father's age and the child's comorbidities (genetic, hearing and vision disorders,

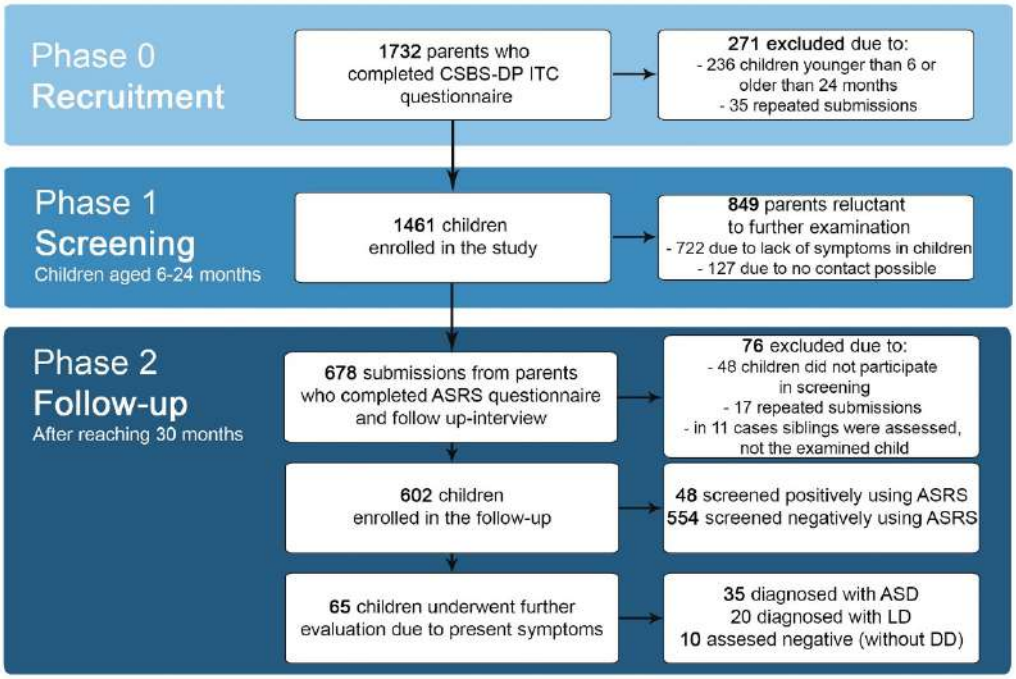

Fig 1. Chart showing the number of study participants at each stage of the Spojrzeć w oczy project. Note. ASRS—Autism Spectrum Rating Scales, CSBS-DP ITC—Communication and Symbolic Behavior Scales-Developmental Profile—Infant-Toddler Checklist, Polish Version, DD—developmental delay, LD—language delay.

https://doi.org/10.1371/journal.pone.0299618.g001

movement disorders, family history), birth weight, and the week of pregnancy in which the delivery took place. Then, upon a child's turning 30 months of age, the respective child's parents were contacted again and asked to fill in the ASRS questionnaire and a short follow-up interview, which included questions about the occurrence of symptoms that were concerning to the parents and further diagnostics.

In the case of suspicion of ASD in a child (understood as a result above ASRS cut-off point or any persistent concerns parents have about their child's development despite negative screening using the ASRS), the ADOS-2 was performed to make it possible to determine with high probability the presence of autism spectrum disorders in children from the risk group. At each stage during the study, parents were offered the possibility of carrying out the ADOS-2 test at the research center in Cieszyn, Silesian Voivodeship. However, due to the distribution of the population throughout the country, the majority of them preferred performing further diagnostics near their place of residence. If children underwent the ADOS test before our intervention or the ADOS test was performed in another center, we tried to verify the test by contacting the examiner to confirm whether the test result indicated the presence of autism spectrum disorders in the child.

Statistical analysis. All the analyses presented in this manuscript were performed using Statistica 13.1 and R 4.3.0 software using packages *ggplot2 3.5.1* and *stats*. Due to the wide variation of results in individual age groups (resulting from rapid development in the age group analyzed) and the insufficient number of participants at a particular age in months, it was necessary to group the participants into age cohorts gathering children at least partially at a similar level of psychomotor development. Participants were divided into four groups based on the substages of Piaget's Sensorimotor Stage theory of cognitive development [36]. The first group included children aged from 6 to 8 months of age, the second group included children between 9 and 12 months, the third group included children between 13 and 18 months, and the fourth group included children above 19 months.

Because more complex tools are also used during further diagnostics of children with suspicion of ASD, first it was checked whether there was any correlation between the results achieved by children in the CSBS-DP ITC questionnaire and the shortened version of the ASRS (used as a Level 2 questionnaire) using the Spearman's rank correlation coefficient method. In addition, since the children were grouped into four age cohorts, it was checked whether the shape of the relationship between CSBS-DP ITC Total Score and Total ASRS is influenced by the variable "Age". The last aspect that was checked was the relationship between the variables of the child's age (in months) and the Total Score obtained in the CSBS-DP ITC questionnaire. To verify the linearity, a normality analysis of the residuals of the linear model was performed using the Kolmogorov—Smirnov and Shapiro—Wilk tests. The tests showed no normality of the residuals in the model. For this reason, non-parametric methods were used for further analysis.

The discriminating ability of CSBS-DP ITC was examined to ensure that it was appropriate for each age cohort. This was achieved by conducting a receiver operating characteristics (ROC) analysis for all four age groups. The discriminating ability of the Polish Version of CSBS-DP ITC was determined by the Area Under the Curve (AUC) statistic. AUC values ranged from 0.5, representing a random chance efficacy, to 1.0, representing excellent performance in discriminating between the two conditions [37]. For each of the groups for which the ROC analysis was successful, potential cut-offs were determined to enable the tool to be used in practice as a clinical guide for further management. Two different methods were used to determine the cut-offs—the first was the cut-off point at which maximum sensitivity and specificity were determined using the Youden index, which represents the overall accuracy of the test [38, 39]. Additionally, the minimizing expected costs method was used by including a "decision threshold" in constructing tangents because potentially classifying a child with ASD as a healthy individual carries a higher cost (in terms of delay in treatment) than the opposite mistake (Zweig & Campbell, 1993). We assumed that the weight of these errors (misclassification costs) increases with the age of the child (and therefore with the delay in diagnosis). It was further assumed that the costs of misclassification (for children falsely classified as healthy—false negative) would be measured as 1, 2, 3, and 5, for the respective groups as age increases; values were adopted arbitrarily.

Due to the unclear prevalence of ASD in the Polish population, three different probabilities were used to determine the cut-off points using the tangent method—random (0.5), declared by WHO for the world population (1:160) and the probability estimated from the study population (depending on the number of cases in a given cohort). Thus, a total of twelve ROC curves were created and analyzed, and the results of these analyses were used to establish the sensitivity and specificity in specific age groups.

Table 2. Sex structure and incidence of developmental disorders in the studied age subgroups.

Variables	Group I *(6–8 months of age)* N = 73	Group II *(9–12 months of age)* N = 131	Group III *(13–18 months of age)* N = 242	Group IV *(19–24 months of age)* N = 156
Sex				
Male	42 (57.53%)	75 (57.25%)	154 (63.64%)	83 (53.21%)
Female	31 (42.47%)	56 (42.75%)	88 (36.36%)	73 (46.79%)
Presence of DD				
ASD	2 (2.74%)	8 (6.11%)	16 (6.61%)	9 (5.77%)
LD	4 (5.48%)	6 (4.58%)	7 (2.89%)	3 (1.92%)

Note. ASD—autism spectrum disorders, DD—developmental disorders, LD—language delay.

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Results

Descriptive statistics

In the study population, among 602 children, 65 of them were further evaluated due to a positive ASRS test result or the presence of disturbing symptoms. Ultimately, 35 children from the entire group were diagnosed with ASD and 20 with language development delay. Data on the sex structure and incidence of developmental disorders in individual age-subgroups are detailed in Table 2.

The largest number of ASD cases (both percentage and total) were confirmed in the subgroup of children aged 13–18 months (n = 16; 6.61%), the smallest—in the subgroup of children aged 6 to 8 months (n = 2; 2.74%). In each age group, boys slightly dominated the study population (from 53.21% to 63.64%).

Correlation between total scores of ASRS and CSBS-DP ITC

To assess predictive accuracy between the earlier CSBS-DP ITC questionnaire result and the later ASRS, Spearman's rank correlation coefficient analysis was used. Since the Total Score achieved in the CSBS-DP ITC questionnaire depends on the age of the examined child, it was additionally checked whether the child's age affects the strength of the relationship between Total CSBS-DP ITC scores. The results are presented in the Table 3 and Figs 2 and 3.

Statistically significant weak or moderate correlations are present between the total ASRS score and the total CSBS-DP ITC score in the three oldest age groups. It should be noted that in each age cohort, there are weak or moderate correlations between the child's age and the total score obtained in CSBS-DP ITC. It should be borne in mind that these two questionnaires have an opposite scale—in the CSBS-DP ITC, the higher the score, the lower the risk of ASD in the child, while for the ASRS the opposite is true, which is clearly visible in Fig 2. The older the children examined, the more pronounced the inverse relationships between the results achieved in the CSBS-DP ITC questionnaire and the ASRS. In order to better present this relationship, an analysis was performed for age groups using ranking (due to the previously mentioned lack of normal distribution of variables).

Sensitivity, specificity of the Polish version of CSBS-DP ITC

In order to determine the sensitivity, specificity and other measures of the classification test for the Polish version of the CSBS-DP ITC, this study used the Youden method of ROC analysis and the tangential method to minimize the potential costs associated with misclassification.

Table 3. Spearman correlation values for the total scores obtained by children in the appropriate age groups in the ASRS, CSBS-DP ITC questionnaire and age when the CSBS-DP ITC was completed (in months).

Variables	Spearman's r	t	p
I (6–8 months of age; N = 73)			
Total ASRS score & Total CSBS-DP ITC score	-0.118	-1.000	0.321
Total CSBS-DP ITC score & Age of child	**0.389**	**3.555**	**0.001**
II (9–12 months of age; N = 131)			
Total ASRS score & Total CSBS-DP ITC score	**-0.280**	**-3.315**	**0.001**
Total CSBS-DP ITC score & Age of child	**0.510**	**6.739**	**<0.001**
III (13–18 months of age; N = 242)			
Total ASRS score & Total CSBS-DP ITC score	**-0.364**	**-6.055**	**<0.001**
Total CSBS-DP ITC score & Age of child	**0.412**	**7.013**	**<0.001**
IV (19–24 months of age; N = 156)			
Total ASRS score & Total CSBS-DP ITC score	**-0.431**	**-5.921**	**<0.001**
Total CSBS-DP ITC score & Age of child	**0.160**	**2.009**	**0.046**

Note. ASRS—Autism Spectrum Rating Scales, CSBS-DP ITC—Communication and Symbolic Behavior Scales Infant-Toddler Checklist

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Results of the analyses and ROC charts for individual age groups using the Youden method are provided in Fig 4a–4c and Table 4. Results using the tangential method with different probabilities of ASD occurrence, due to its lower effectiveness compared to Youden's method, are included in SF5.

Due to the small number of ASD cases in the first group (children aged 6 to 8 months), the ROC analysis in this subgroup could not be performed.

The highest sensitivity and specificity values were achieved using the Youden method and cut-offs did not change depending on the assumed probability of ASD occurrence—when using the prevalence "from the sample", random (0.5) and prevalence based on data from

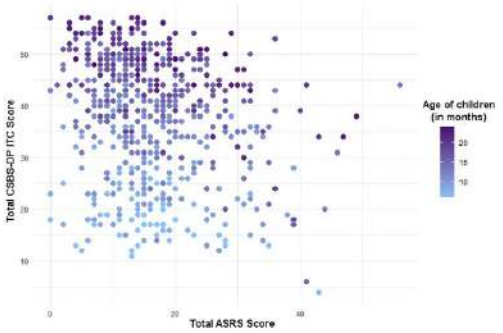

Fig 2. Distribution of children's total scores in the CSBS-DP ITC and ASRS questionnaires by children's age. Each dot indicates one child's score on the CSBS-DP ITC questionnaire (Y-axis) in relation to the score on the ASRS questionnaire (X-axis), the color corresponds to the child's age at the time of completing the CSBS-DP ITC questionnaire. Lower scores on the CSBS-DP ITC are also observed in the group achieving low ASRS scores due to lower age.

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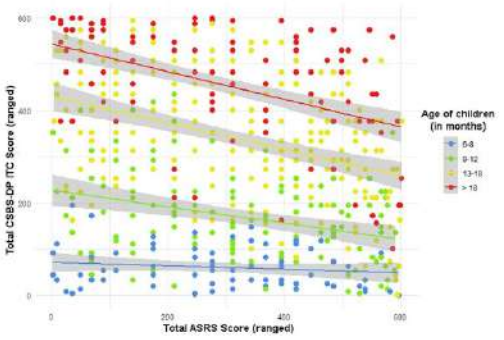

Fig 3. Rank analysis of the distribution of children's total scores achieved in the CSBS-DP ITC and ASRS questionnaires in the accepted age groups. Each dot indicates the rank score one child's score on the CSBS-DP ITC questionnaire (Y-axis) and ASRS (X-axis) with a marked trend (which is the highest for the oldest children and the lowest—for the youngest).

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systematic reviews (1:160 according to WHO), the ROC curves plotted using the Youden method did not differ from each other. In the case of using the tangential method, the adopted cut-offs significantly differed depending on the assumed probability of ASD occurrence in the Polish population and they were usually characterized by lower accuracy or a significant disproportion between sensitivity and specificity in favor of one of these parameters. Moreover, adopting cut-offs determined using this method may be too volatile (e.g., 30 or 54 points in group 19–24 months of age), which is also probably due to the small number of cases of children diagnosed with ASD in the sample.

Among all performed ROC analyses, the area under the curve (AUC) index ranged from 0.782 to 0.856, which is a value allowing for the classification ability for the CSBS-DP ITC to be considered as at least above moderate. Among children in the age group of 9–12 months, a cut-off of 21 points was adopted, allowing for a sensitivity of 0.750 and a specificity of 0.862 with a Youden index of 0.612; in the age group of 13–18 months, a cut-off of 36 points was adopted, giving a sensitivity of 0.750 and a specificity of 0.854 with a Youden index of 0.604, and in the group of 19–24 months—a cut-off of 39 points, giving a sensitivity of 0.667 and specificity of 0.939 with a Youden index of 0.605.

Due to the size of the groups, analyzes of the CSBS-DP ITC subscales (Social, Speech, Symbolic components) using ROC curves did not allow for clear cut-off points for children. For this reason, if it is necessary to use subscales to determine a child's risk, we suggest using the method used in the original version of the CSBS-DP ITC questionnaire (i.e. 1.25 SD below the mean score in the study population). Raw results and cut-off thresholds for this method were described in a previous publication on the Polish version of CSBS-DP ITC [28].

Discussion

The aim of this study was to determine the final psychometric values of the Polish version of the CSBS-DP ITC questionnaire as part of the nationwide project. The questionnaire enables testing children for developmental disorders from the age of 6 months, which is a distinctive feature of this study—previous studies on other diagnostic tools e.g. Modified-Checklist for Autism in Toddlers (Revised) (M-CHAT-R/F), Brief Infant Toddler Social Emotional

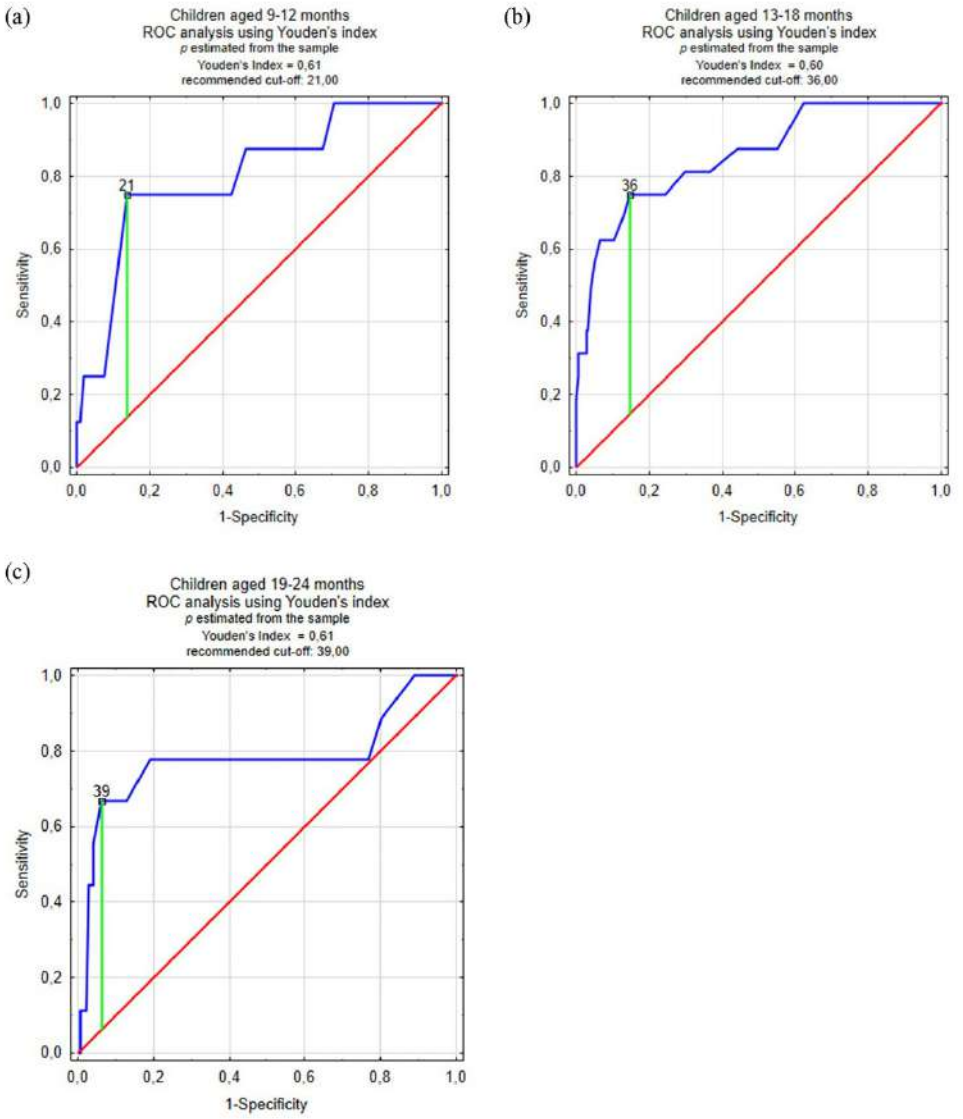

Fig 4. A-C. ROC analysis charts in individual age groups using the Youden method.

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Table 4. Values of sensitivity, specificity and other parameters for adopted, best matching cut-off points in given age groups.

Total result (Cut-off)	Sensitivity	Specificity	Youden index	Accuracy	PPV	NPV	FPR	FNR	LR(+)	LR (-)
Group II (children aged 9–12 months; N = 131) AUC = 0.804; z = 3.664; p < 0.001										
19	0.375	0.911	0.286	0.878	0.214	0.957	0.089	0.625	4.193	0.686
20	0.500	0.894	0.394	0.870	0.235	0.965	0.106	0.500	4.731	0.559
21	**0.750**	**0.862**	**0.612**	**0.855**	**0.261**	**0.981**	**0.138**	**0.250**	**5.426**	**0.290**
22	0.750	0.821	0.571	0.817	0.214	0.981	0.179	0.250	4.193	0.304
23	0.750	0.780	0.530	0.779	0.182	0.980	0.220	0.250	3.417	0.320
Group III (children aged 13–18 months; N = 242) AUC = 0.856; z = 6.941; p < 0.001										
34	0.625	0.898	0.523	0.880	0.303	0.971	0.102	0.375	6.141	0.417
35	0.688	0.872	0.559	0.860	0.275	0.975	0.128	0.313	5.358	0.359
36	**0.750**	**0.854**	**0.604**	**0.847**	**0.267**	**0.980**	**0.146**	**0.250**	**5.136**	**0.293**
37	0.750	0.823	0.573	0.818	0.231	0.979	0.177	0.250	4.238	0.304
38	0.750	0.757	0.507	0.756	0.179	0.977	0.243	0.250	3.082	0.330
Group IV (children aged 19–24 months; N = 156) AUC = 0.782; z = 2.597; p < 0.001										
35	0.444	0.959	0.404	0.929	0.400	0.966	0.041	0.556	10.889	0.579
38	0.556	0.959	0.515	0.936	0.455	0.972	0.041	0.444	13.611	0.463
39	**0.667**	**0.939**	**0.605**	**0.923**	**0.400**	**0.979**	**0.061**	**0.333**	**10.889**	**0.355**
40	0.667	0.925	0.592	0.910	0.353	0.978	0.075	0.333	8.909	0.360
41	0.667	0.918	0.585	0.904	0.333	0.978	0.082	0.333	8.167	0.363

Note. Bold values correspond to the cut-off value that has the best predictive parameters according to the ROC analysis using the Youden method. Sensitivity—true positive rate; specificity—true negative rate; PPV—positive predictive value (presicion); NPV—negative predictive value; FPR—false positive rate (fall-out rate); FNR—false negative rate (miss rate); LR(+)–positive likelihood ratio; LR(-)–negative likelihood ratio.

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Assessment (BITSEA), First Year Inventory (FYI) or Quantitative Checklist for Autism in Toddlers (Q-CHAT) were focused on older children [40–43]. In addition to the CSBS-DP ITC, the only screening questionnaire designed to be completed by parents (rather than trained observers) for children aged 6 months and older is the Canadian-validated Autism Parent Screen for Infants (APSI) [44]. Therefore, this study may provide evidence for the purposefulness of screening for developmental disorders among the youngest children in the primary care setting.

Due to the dynamic development of children's skills in the studied period, it was necessary to take into account the age difference affecting the results achieved by children. In the original version of the CSBS-DP ITC questionnaire, there are separate cut-off points for each individual month of age, however, they have been set as a point below 1.25 SD from the average result in the general population. Due to the lack of sufficient participants in the study with a confirmed diagnosis of ASD, only solution to this problem was to group children into appropriate larger age cohorts.

Data from the analysis of the statistical model indicate that the correlation between the child's age and the total score in the CSBS-DP ITC questionnaire is also present in age subgroups, which means that children close to the age limit of the cohort may be classified differently by the questionnaire. The influence of age on the total score would probably be even stronger if it was examined without grouping children into cohorts, however, due to its significance, this effect cannot be ignored. Moreover, the analyses do not confirm the linearity of this impact—the introduction of a simple correction based on a linear relationship would involve

the risk of error. Therefore, this is another reason why it was necessary to use previously developed theoretical constructs (in this case, Piaget's theory) to divide children into larger age groups. This, in turn, facilitated the conduct of analyses and the determination of threshold values according to the observed acquisition of particular abilities by children at the appropriate age. With the amount of data available, unfortunately, the only remaining solution is to take into account the correlation with age when interpreting the results and leave a margin of distrust for the scale. This is of particular relevance when evaluating children on the verge of age ranges. In case of doubt, as suggested by the authors of the original version, in the current study it is also recommended to check the child's development in the next three months.

Based on a sample of 1,461 simple screening questionnaires, at least 35 children were diagnosed with ASD, which gives a prevalence of ASD in the Polish population of 2.39%–considering that the majority of parents who did not participate in the follow-up with the use of ASRS and follow-up interview dropped out due to the lack of suspicion of any symptoms of developmental disorders in their children. This percentage is even higher if the study considers only children assessed using the ASRS and subsequent tests (e.g. ADOS-2) and it may be estimated as 5.81%. Compared to the preliminary data from two Polish regions (which indicate 32–38 cases of ASD per 10,000 people), the prevalence of ASD in the group of respondents in the current study is markedly higher. The percentage of children diagnosed with ASD in whole project (2.39%) is therefore similar to the most recent data for American children (1:64) [19]. Similarly, the potentially unusual high percentage of children who underwent rehabilitation or with detected muscle tension disorders and constituted the study group does not seem to differ significantly from the general Polish population. This results from the high frequency of their detection in the Polish population in accordance with the guidelines of the Children and Youth Rehabilitation Section of the Polish Rehabilitation Society, as well as the high popularity of rehabilitation in Poland [29]. According to data collected on behalf of the National Chamber of Physiotherapists, as many as 49.9% of Poles use the services of physiotherapists; however, current accurate data on children are lacking [30].

However, it should be borne in mind that the selection of the method (due to the COVID-19 pandemic, the study was conducted online) may have resulted in some bias of the group—the questionnaire was filled in by willing parents, so parents observing the occurrence of any deficits in their children may account for a larger part of respondents than it actually could be the case in stationary primary care setting. Nevertheless, every effort was made to convince also parents who do not suspect any developmental disorders in their children to complete the questionnaire. It seems that these efforts were effective– 383 parents from 1461 included in the study reported concerns about their child's development, which constitutes 25.7% of all participants. This is perfectly in line with the data from a study conducted at the C.S. Mott Children's Hospital, Michigan, US, where it was shown that a quarter of parents also report suspected developmental disorders in their children [45]. Similar conclusions can be drawn from studies in the Netherlands, where even up to 50% of parents reported minor concerns about their children's development [46]. Notwithstanding the foregoing, the data from the current study indicate that despite the potential bias of the group, there is still a large proportion of undiagnosed individuals in the general population who could benefit from appropriate diagnosis and treatment.

In order to reduce the rate of false positive results, it may be effective to include an observational study as a follow-up measure immediately after a positive result. The authors of the original version of the CSBS-DP created additional, further tools that aim to increase the sensitivity and specificity of the ITC questionnaire (i.e. Behavior Sample, where a pre-trained healthcare professional (HCP) assesses the child's development, or Caregiver Questionnaire, where the child's development is assessed by e.g. a caregiver in a nursery or nanny) [47].

Numerous evidence has also been described that the inclusion of follow-up methods increases the sensitivity and specificity also in the case of other ASD screening tools (e.g. M-CHAT R/F) [48]. In addition, the use of a Behavior Sample by HCPs immediately after an ITC assessment could reduce the potential dropout and be used as an immediate validation strategy. The use of a follow-up method right after the questionnaire-based screening method could be all the more important because some of the families, even after obtaining a result of the screening tests suggesting the possibility of ASD in the child and indicating the need for further diagnostics, still do not feel concerned about the development of their child as a study performed in Flanders suggests [49]. Major obstacles in trying to incorporate additional diagnostic methods in primary care settings are the excessive workload, insufficient time, and inadequate knowledge of HCPs about ASD [50–52].

Another issue worth mentioning is the possibility of using the CSBS-DP ITC questionnaire to detect developmental disorders other than ASD, e.g. language development delay (LD), which were observed in 20 respondents in our study (3.32%), or to assess strengths or weaknesses in a child's developmental skills. Early detection of children with LD, especially those from high-risk groups, may benefit them in connection with the initiation of appropriate therapy, however, the effects of such therapy are less pronounced than in the case of ASD, as indicated by the results of the Danish SPELL longitudinal study conducted in the general population [53, 54]. The presence of three main CSBS-DP ITC subscales concerning social skills, symbolic skills, and speech development indicates the child's potential resources and the most important deficits and may serve as a cue for early intervention therapists before further, more specialized diagnostics. However, the unequivocal use of this information in practice requires further research on the CSBS-DP ITC questionnaire specifically in risk groups.

Nevertheless, the evidence from this study may contribute to the discussion regarding the inclusion of ASD screening during well-child care visits. So far, no fully validated questionnaire for the diagnosis of ASD in children under 2 years of age has existed in Poland; only one study on the reliability of the Q-CHAT questionnaire was published, without an attempt to estimate the cut-off in the Polish population or assess the sensitivity and specificity of the tool [55]. Taking into account the positive data from the current study on the CSBS-DP ITC properties, the use of this tool in everyday practice of HCPs would seem justified.

Increasing early diagnostic capabilities through the use of CSBS-DP ITC in screening has the potential to provide large population benefits similar to those observed in the United States, associated with a significant reduction in the average age of diagnosis and an increase in the percentage of children diagnosed with ASD [19]. Taking into account the previously mentioned low official prevalence of ASD in Poland (which is most likely due to incomplete diagnosis, difficulties with access to specialists and lack of guidelines on how to deal with a child with suspected ASD), as well as the high average age at which the diagnosis of ASD in children is made implementing of ASD screening could improve the situation of children and their families [29]. It is estimated that the average age of receiving an ASD diagnosis in Poland is 7 years and 3 months, which is much higher than the world average, which worsens the potential outcomes of ASD therapy in terms of communication or language skills [12, 56].

The Polish version of CSBS-DP ITC is characterized by psychometric parameters slightly lower than the original version, and similar to the Taiwanese version (sensitivity 0.77) or the Italian version (maximum sensitivity 0.67, specificity 0.98, PPV 0.6, NPV 0.98); however, the Italian version achieved significantly lower diagnostic values in younger children [26, 57, 58]. The Polish version of the CSBS-DP ITC questionnaire, probably due to linguistic and cultural adjustment, does not have such a large impact of low age on psychometric values, which means that it can be used in screening children from 9 months of age [59]. This makes it

possible to implement early intervention at a very early stage of the child's development, which will probably enable achieving better final effects of tailored therapy [60].

There are several limitations to this study. The first is the relatively small number of study participants whose development was monitored throughout the study. Although this number fully meets the criteria of scientific research, a larger sample of respondents could provide better evidence of the usefulness of ASD screening in the Polish population and, at the same time, better estimate the prevalence of ASD in this age group in Poland. Another limitation is the method of electronic screening and the related self-selection of participants. It is believed to be one of the main factors for unrepresentativeness in studies conducted via online questionnaires. The attitudes of study participants may have influenced their decision to take part in the survey, which is why parents who suspected any developmental disorders in their children were more willing to participate in the study [61]. However, as mentioned earlier in the discussion, the present authors made every effort to avoid self-selection bias and, given the percentage of parents with any concerns about their child's development compared to other inpatient studies, most likely the attempt to make it negligible have been successful.

Another possible limitation is the tracking of children's development using online and telephone methods. Due to remote contact with parents, the authors relied on answers in questionnaires, surveys, and medical documentation provided. Nevertheless, in order to qualify the examined child to the group of children diagnosed with ASD, it was required to present appropriate documents prepared by a qualified psychologist or psychiatrist. The last flaw of the study is the time of observation of children—with the endpoint set at 30 months of age, and the possibility of extending this time if the child is undergoing further diagnostics, has received a positive ASRS result or parents have further concerns about the child's development. At the same time, according to the DSM-V definition of ASD, the symptoms of autism spectrum disorders may become fully apparent also in later years of development. Despite providing psychological help to children who require it, some of them may develop symptoms later—when social demands exceed the limited capacities of the child [1, 62].

Conclusions

The Polish version of the CSBS-DP ITC questionnaire is characterized by reasonably high values of sensitivity, specificity, and accuracy in children aged from 9 to 24 months and children's performance in the questionnaire may also be a predictor of outcomes in later examinations (i.e. ASRS). The conducted analyses indicate the potential usefulness of the Polish version of the CSBS-DP ITC questionnaire in the everyday practice of healthcare professionals for population screening for ASD. Further research is essential to more accurately define cut-offs among children who are on the verge of the accepted age ranges and among the youngest population (aged 6–8 months).

Supporting information

S1 File. Questionnaire—The Polish version of CSBS DP-ITC.
(PDF)

S2 File. Questionnaire—English translation of the Polish version of CSBS DP-ITC. Retranslated (from Polish to English) Polish version of CSBS-DP ITC.
(PDF)

S3 File. Ethics committee approval. Full text of Ethics Committee Approval for conducting this study obtained by the Wroclaw Medical University Ethics Committee.
(PDF)

S4 File. Translated ethics committee approval. Translated from Polish full text of Ethics Committee Approval for conducting this study obtained by the Wroclaw Medical University Ethics Committee.
(DOCX)

S5 File. Results and discussion on sensitivity, specificity of the Polish version of CSBS-DP ITC calculated using tangential method.
(DOCX)

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Due to obtaining consent from the authors of the original version of the CSBS-DP ITC questionnaire and the general free availability of the questionnaire, the Polish version of the CSBS-DP ITC questionnaire is covered by the CC BY license, with particular emphasis on the original authors and Paul H. Brookes Publishing Co.

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10.4. Implementing an Early Detection Program for Autism Spectrum Disorders in the Polish Primary Healthcare Setting—Possible Obstacles and Experiences from Online ASD Screening

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Implementing an Early Detection Program for Autism Spectrum Disorders in the Polish Primary Healthcare Setting—Possible Obstacles and Experiences from Online ASD Screening

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Abstract: A screening questionnaire for autism symptoms is not yet available in Poland, and there are no recommendations regarding screening for developmental disorders in Polish primary healthcare. The aim of this study was to assess the opinions of parents and physicians on the legitimacy and necessity of screening for autism spectrum disorders, potential barriers to the implementation of the screening program, and the evaluation and presentation of the process of online ASD screening, which was part of the validation program for the Polish version of one of the screening tools. This study involved 418 parents whose children were screened online and 95 primary care physicians who expressed their opinions in prepared surveys. The results indicate that both parents and doctors perceive the need to screen children for ASD in the general population without a clear preference as to the screening method (online or in person). Moreover, online screening is considered by respondents as a satisfactory diagnostic method. Therefore, online screening may prove to be at least a partial method of solving numerous obstacles indicated by participants' systemic difficulties including time constraints, the lack of experienced specialists in the field of developmental disorders and organizational difficulties of healthcare systems.

Keywords: autism spectrum disorders; population screening; autism screening; online screening

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1. Introduction

Autism spectrum disorders (ASD) are neurodevelopmental conditions of undetermined etiology, most often manifested in early childhood, which affect daily activity and are characterized primarily by difficulties in the sphere of communication and interpersonal interactions as well as restricted interests or repetitive behaviors, with prevalence estimated to 1/100 worldwide [1,2]. There are no reliable data available on the prevalence of ASD in Poland—according to official data from the Polish National Health Fund from 2012, the prevalence rate of ASD in individuals under the age of 18 in Poland is 3.4 cases per 10,000 children [3]. More recent preliminary data from two Polish provinces estimate that ASD occurs in one in 286 children [4]. It is not entirely clear why the reported prevalence of ASD in Poland is lower than in neighboring countries (e.g., in Germany—0.38%) and lower than assumed, based on newer data from European Union countries [5,6]—it may be the result of lacking readily available tools for the screening and diagnosis of ASD, insufficient awareness of this problem of healthcare professionals (HCPs) and systemic difficulties and constraints.

Accelerated diagnosis of ASD enables initiation of an early, appropriately adjusted therapy [7]. First symptoms of ASD usually appear during infancy or early childhood—prodromal symptoms may be visible even as early as 6 months of a child's life [7,8] and usually reliable diagnosis of ASD in a child can be made as early as 2–3 years of age [9]. The younger the child, the better the results of therapy that can be achieved in the area of communication and social interaction, cognitive abilities, speech development or behavior appropriate to the situation, which improves

the quality of life of people with ASD, reduces the risk of mental disorders, and significantly reduces the ASD burden [10–13]. The burdens associated with the diagnosis of ASD in a child in the family include lowering the quality of life of the parents of a child with ASD, lowering the quality of interaction with the child and increasing perceived parental stress [14,15].

For this reason, the American Academy of Pediatrics recommends screening at 18 and 24 months of age as part of primary care during well-child care visits [16]. It appears that the increasing availability of screening significantly lowered the age of ASD diagnosis in the US, with diagnosis before the age of 4 made in 71% of children (2018) compared to 58% in 2014 [17,18]. However, a US Preventive Services Task Force from 2016 report shows insufficient evidence to recommend universal ASD screening [19]. On the other hand, there is evidence suggesting that including screening tools in routine medical appointments may result in earlier and more accurate identification of children who need further help than relying solely on clinical impressions, which is particularly important when care providers are less experienced in diagnosing ASD [20]. Moreover, the use of public ASD screening may reduce social inequalities in terms of the age of diagnosis and access to further therapeutic activities [21,22]. The conclusions of both reports indicate the need for further research on screening tools and their effectiveness, as well as on the effectiveness of further proceedings after screening [23].

Throughout infancy and early childhood, primary care professionals are the professionals with whom a young child most often comes into contact [24]. During this time, numerous meetings with doctors, nurses, midwives and other HCPs take place—as a result of visits due to the child's illness, well-child check-ups or qualifying for vaccinations [25]. This enables close observation of development of the child and detection of developmental disorders [26]. Primary care professionals are believed to have the greatest influence on the early diagnosis of ASD (especially due to performing screening for developmental disorders (DD)), and they are responsible for the significant trend in the decline in the average age of children when diagnosed with autism spectrum disorders [26]. In addition, family doctors are coordinators of diagnostics and treatment performed by many of the further specialists, as well as "caregivers" of their patients not only on the health issues, but also on the social issues—which is necessary in the event of ASD in a child.

In Poland, well-child care visits have been conducted in primary healthcare clinics since the 1950s. The system of preventive healthcare for children is based on the principle of continuity of care—according to this assumption, the same doctor looks after a child in health and in sickness. After the newborn is discharged from the hospital, parents are obliged to report the child to a selected primary care clinic. Well-child care visits take place at the ages of 2, 6, 9, and 12 months and later at 2, 4 and 5 years of age. During the latter, an indicative examination of hearing (assessment of hearing behavior and the "show what you hear" test) and vision (especially in the direction of strabismus, later—also other disorders) is performed [27]. At 5–6 years of age, an indicative study of motor and psychosocial development should be carried out on the basis of interviews and observation, but it is rarely a validated, structured study [28]. Until then, the assessment of psychomotor development is primarily an assessment of the child's time to reach milestones—there are no other established guidelines for the use of any tools for DD screening in children. In the case of suspicion of DDs, the child is referred to appropriate psychological and pedagogical clinics, where further examinations and consultations are carried out [29]. The validated diagnostic methods available in Poland include Level 2 tools, Autism Spectrum Rating Scales (ASRS) and Social Communication Questionnaire (SCQ), and Level 3 tools, Autism Diagnostic Observation Schedule (ADOS-2) and Psychoeducational Profile, Third Edition—PEP-3-PL [30–33]. Research on the Polish version of Autism Diagnostic Interview-Revised (ADI-R) is in progress [34]. During the final diagnosis, children with visible developmental disorders may be covered by the Early Development Support program—comprehensive, multidisciplinary activities to support the child's psychomotor development through the intervention of therapists (psychologists,

speech therapists, educators, SI therapists); however, they must be referred there by a doctor or preschool teacher.

In order to increase the effectiveness of early detection of autism spectrum symptoms, many tools have been created to facilitate this process for healthcare workers. Due to the lack of standardized tools for ASD screening in Poland, we started the "Spojrzeć w oczy" (eng. "Look in the eyes") project. The aim of this project was to validate the Polish version of the Communication and Symbolic Behavior Scale-Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC). The CSBS-DP ITC is one of the available tools for the early detection of symptoms of autism spectrum disorders consisting of 24 items, filled out by parents and guardians [35]. The CSBS-DP ITC can be used in ASD screening for the general population of children aged 6 to 24 months in a primary care setting. In previous studies, we showed that the Polish version of the CSBS-DP ITC has good psychometric properties and relatively high specificity and sensitivity and can be used as an effective screening tool [36,37].

During the planning of this study, the COVID-19 pandemic spread globally, which prevented efficiently carrying out the stationary study in primary healthcare clinics. The pandemic has made it necessary to introduce solutions that would allow access to remote medical services, which reduced the risk of coronavirus infection [38,39]. For this reason, it was necessary to create an electronic version of the CSBS-DP ITC questionnaire to be completed by parents at home. Then, the results of examination, instead of being assessed by individual healthcare professionals (HCPs, e.g., GPs, pediatricians, nurses, psychologists), were assessed directly by specialists involved in the project. Epidemiological situation forced the screening to be carried out in a unique way, using the online version, without direct contact with the examined person. The use of telemedicine made it possible to contact the patient, perform the screening, conduct follow-up tests and possibly dispel the parents' doubts and questions.

The aim of this paper is to assess opinions of HCPs and parents performing electronic screening and to present the experiences from one of the first attempts to conduct such a screening program. In addition, we wanted to assess the possible constraints in usage of ASD screening tools in HCPs' everyday practice. Approval from the Bioethics Committee of the Wroclaw Medical University was obtained to conduct this study (number KB—641/2020). All procedures were performed in accordance with the 1964 Helsinki declaration and its later amendments.

2. Materials and Methods

2.1. Assessment of ASD Electronic Screening by Parents

We sent e-mail invitations to 1461 parents of children whose applications in the first phase of the "Spojrzec w oczy" project were included in the assessment of the psychometric values of the CSBS-DP ITC (the criteria for inclusion of children in this study regarding the properties of the questionnaire were being a resident of Poland, the age 6–24 months and usage of Polish as the main language by both parents) [36]. We asked parents to fill out an electronic short questionnaire about their feelings on electronic screening for developmental disorders, recording their age, level of education, place of residence and subjective information technology (IT) proficiency on a scale from 1 to 10 (where 1—is the lowest, 10—the highest) as factors potentially influencing the assessment of the diagnostic process [40,41]. A translated version of the survey is available as Supplementary File S1. We did not link parents' answers to their child's results in the CSBS-DP ITC questionnaire to maintain anonymity; however, we asked them to select if they benefited from additional contact with people involved in the project (e.g., further diagnostics, specialist advice) or if their child was diagnosed with ASD or other developmental disorders, as a factor that could potentially increase satisfaction with online screening. The survey was made available on the project's website—it is available only through the appropriate link, sent only to the parents included in this study. Invitations to evaluate the project were sent on 9 January 2023 and responses were collected until 18 January 2023.

2.2. Assessment of ASD Electronic Screening by Doctors and Potential Difficulties in Implementing the ASD Screening Program

In order to recruit physicians for this study, we sent e-mail invitations to members in the Polish Society of Family Medicine and we propagated the survey on the project's social media profiles. The doctors were verified with their profession practice number—a document possessed by every doctor practicing in Poland. The survey consisted of 18 questions, 7 of which concerned statistical data—age, sex, main place of work, work experience, and the percentage of children with developmental disorders (DD) under their own medical care. The remaining ones touched upon aspects of the use of diagnostic methods in clinical practice, further proceedings when DD is suspected, the willingness to use screening methods during medical visits, the assessment of potential difficulties and limitations in introducing screening tests, and the positives and negatives resulting from online DD screening. A translated version of the survey for doctors is available as Supplementary File S2. Applications were collected from 31 July 2023 to 1 February 2024.

2.3. Statistical Analysis

All the analyses presented in this manuscript were performed using Statistica 13.3 software. The Shapiro–Wilk normality test was applied to check the normal distribution using 0.05 as a significance level. None of the examined variables met the criterion of normality of distribution; therefore, non-parametric tests were used for further analysis. The dependence of the variables on the categories of qualitative variables was tested using the non-parametric Kruskal–Wallis test. This test enabled a direct comparison of the value of a quantitative variable between the two categories of a qualitative variable. In the case of qualitative variables, the uniformity of the category distribution was tested with the chi-square uniformity test (one sample proportions test). In some cases, Spearman's rank correlation was used, allowing variables on ordinal and quantitative scales that did not have a normal distribution to be correlated with each other. In the case of a small number of respondents in subgroups, the Yates correction was used in the chi-square test. A significance level of 0.05 was set for all tests. In the case of some of the examined features, only a descriptive interpretation was possible. This is mainly due to the inability to conduct further statistical analyses—these variables cannot be tested for homogeneity. The respondents could indicate several categories at the same time, so the categories do not meet the separability condition required for homogeneity testing.

3. Results

We received a positive response and a fully completed survey from 418 parents taking part in the "Spojrzeć w oczy" project, giving a response rate of 28.6%. Due to forcing the answers to individual questions and the full anonymity of the survey, it was not necessary to exclude any submissions. The number of participants exceeds the minimum sample size to achieve a confidence level of 95% and a margin of error of 5% in the Polish population (the calculated minimum sample size was 384) [42]. All submissions were completed by female participants except one. The mean age of the parents was 33.86 years (SD = 4.12) and the mean subjective IT proficiency was 8.52 (SD = 1.25). Parents' full sociodemographic characteristics are presented in Table 1.

Table 1. Sociodemographic characteristics of parents taking part in this study.

Characteristic	*n*	%
Sex		
Female	417	99.76%
Male	1	0.24%
Using additional contact with researchers		
Yes	38	9.09%
No	380	90.91%

Table 1. *Cont.*

Characteristic	*n*	%
Diagnosed DD in their child during the project		
Yes	30 (21 ASD, 8 LD, 1 SI)	7.18%
No	388	92.82%
Education level		
Lower education	0	0%
Secondary education	58	13.88%
Higher education	360	86.12%
Place of residence		
Village	99	23.68%
Town inhabited by less than 20,000 people	33	7.89%
City inhabited by 20,000–100,000 people	59	14.11%
City inhabited by more than 100,000 people	227	54.31%

Note. Secondary education is basic vocational, general or technical secondary education with or without a high school diploma and post-secondary studies which are not higher education). DD—developmental disorders.

Among the parents who took part in the ASD electronic screening assessment, 38 additionally contacted us for more information and diagnostic and therapeutic assistance via e-mail and telephone. A total of 30 of their children were diagnosed with a developmental disorder—21 with ASD, 8 with speech development delay, and 1 with sensory integration disorder.

Of all the parents, only 4 (0.95%) had not heard of ASD before and 184 (44.02%) did not suspect any developmental disorders in their child—in this group, one child was diagnosed with ASD, the other 3—with language delay (LD). Each parent was asked to answer questions about ASD awareness and opinions on ASD screening—both online and in person. Chi-square tests and the Kruskal–Wallis test were used for this analysis, depending on the nature of the studied variables. The collected results are presented in Tables 2 and 3.

Table 2. Opinions on ASD screening (also online) and awareness of the problem among parents depending on place of residence, education and final diagnosis of the child.

	Number of Positive Responses	Number of Negative Responses	% of Positive Responses	*df*	X^2	*p*
Before screening, had you heard about ASD?						
Place of residency						
Village	95	4	95.96%	3		
Town inhabited by less than 20,000 people	33	0	100%		13.013	0.005
City inhabited by 20,000–100,000 people	59	0	100%			
City inhabited by more than 100,000 people	227	0	100%			
Education level						
Secondary education	54	4	93.10%	1	13.398	<0.001
Higher education	360	0	100%			
Received diagnosis of DD						
Yes	30	0	100%	1	0.312	0.576
No	384	4	98.97%			
Has it occurred to you that there may be possibility that your children may have ASD?						
Place of residency						
Village	50	49	50.51%	3		
Town inhabited by less than 20,000 people	23	10	69.70%		9.581	0.022
City inhabited by 20,000–100,000 people	25	34	42.37%			
City inhabited by more than 100,000 people	136	91	59.91%			
Education level						
Secondary education	32	26	55.17%	1	0.189	0.663
Higher education	202	158	56.11%			
Received diagnosis of DD						
Yes	26	4	86.67%	1	12.350	<0.001
No	208	180	53.61%			

Table 2. *Cont.*

	Number of Positive Responses	Number of Negative Responses	% of Positive Responses	*df*	X^2	*p*
Should screening for ASD be mandatory?						
Place of residency						
Village	99	0	100%	3		
Town inhabited by less than 20,000 people	31	2	93.94%		6.749	0.081
City inhabited by 20,000–100,000 people	59	0	100%			
City inhabited by more than 100,000 people	221	6	97.36%			
Education level						
Secondary education	56	2	96.55%	1	1.028	0.311
Higher education	354	6	98.33%			
Received diagnosis of DD						
Yes	30	0	100%	1	0.631	0.427
No	380	8	97.94%			
Would you participate in screening for ASD in your other children?						
Place of residency						
Village	99	0	100%	3		
Town inhabited by less than 20,000 people	31	2	93.94%		8.497	0.037
City inhabited by 20,000–100,000 people	57	2	96.61%			
City inhabited by more than 100,000 people	225	2	99.12%			
Education level						
Secondary education	58	0	100%	1	0.913	0.339
Higher education	354	6	98.33%			
Received diagnosis of DD						
Yes	30	0	100%	1	0.471	0.493
No	382	6	98.45%			
Would you participate in online screening for ASD in your other children?						
Place of residency						
Village	96	3	96.97%	3		
Town inhabited by less than 20,000 people	29	4	87.88%		11.429	0.010
City inhabited by 20,000–100,000 people	58	1	98.31%			
City inhabited by more than 100,000 people	223	4	98.24%			
Education level						
Secondary education	56	2	96.55%	1	0.143	0.705
Higher education	350	10	97.22%			
Received diagnosis of DD						
Yes	28	2	93.33%	1	1.670	0.196
No	378	10	97.42%			
Do you prefer screening to be carried out stationary at your clinic?						
Place of residency						
Village	42	57	42.42%	3		
Town inhabited by less than 20,000 people	19	14	57.58%		4.084	0.254
City inhabited by 20,000–100,000 people	33	26	55.93%			
City inhabited by more than 100,000 people	106	121	46.70%			
Education level						
Secondary education	24	34	41.38%	1	0.371	0.542
Higher education	176	184	48.89%			
Received diagnosis of DD						
Yes	186	202	47.94%	1	0.471	0.493
No	14	16	46.67%			

Note. ASD—autism spectrum disorders, DD—developmental disorders, *df*—degrees of freedom, and X^2—chi-square test statistic result

Table 3. Opinions on ASD screening (also online) and awareness of the problem among parents depending on parents' age and IT proficiency.

Variables	H	*df*	*p*
Before screening, had you heard about ASD?			
Age	16.327	19	0.635
IT proficiency	14.801	5	0.011
Has it occurred to you that there may be a possibility that your children may have ASD?			
Age	12.970	19	0.840
IT proficiency	8.374	5	0.137
Should screening for DD be mandatory?			
Age	19.114	19	0.450
IT proficiency	3.277	5	0.657
Would you participate in screening for DD in your other children?			
Age	27.965	19	0.084
IT proficiency	7.550	5	0.183
Would you participate in online screening for DD in your other children?			
Age	16.013	19	0.656
IT proficiency	9.911	5	0.078
Do you prefer screening to be carried out stationary at your clinic?			
Age	9.599	19	0.962
IT proficiency	16.212	5	0.006

Note. ASD—autism spectrum disorders, DD—developmental disorders, *df*—degrees of freedom, and H—Kruskal–Wallis test statistic result.

The significant majority of parents (99.04% of all, N = 414) participating in this study were aware of the issue of ASD at least to a basic extent before the screening test. Nevertheless, there is a significant difference in the level of awareness of ASD among parents with lower education, living in rural areas and with lower IT proficiency compared to other parents ($\chi^2 = 13.013$, $p = 0.005$; $\chi^2 = 13.398$, $p = <0.001$; H = 14.801, $p = 0.011$, respectively). Moreover, as many as 98.09% of parents (N = 410) believe that ASD screening should be mandatory, 98.56% (N = 412) would participate in screening for developmental disorders in their next child again, and 97.13% (N = 406) would participate in online screening. The percentage of parents willing to participate again for another child is lower for parents living in smaller towns. The parents' opinion differs widely in the case of preferences for the form of screening (online or stationary)—a slight majority prefer the online version (52.15%, N = 218), but this does not depend on place of residence, education or the fact of receiving a diagnosis of developmental disorders in a child—the only factor influencing the preference for the online method is higher IT proficiency (H = 16.212; $p = 0.006$).

A rather intriguing issue is the fact that parents whose children were finally diagnosed with developmental disorders report much more often than others that they have had the thought that their child may be at risk of suffering from ASD ($\chi^2 = 12.350$, $p = <0.001$).

We also asked parents four questions regarding their assessment of the electronic screening carried out by us as part of the "Spojrzeć w oczy" project using the Polish online-version of the CSBS-DP ITC questionnaire. Responses to questions were based on a five-point Likert scale. The Kruskal–Wallis test and Spearman's rank correlation were used for this analysis. The results regarding the assessment of the quality of online screening and satisfaction with participation in this study depending on age, IT proficiency, place of residence and education are included in Tables 4 and 5.

Table 4. Assessment of online ASD screening conducted in the "Spojrzeć w oczy" project by parents depending on place of residence and education level.

Variable	M	Me	SD	*df*	H	*p*
The information available during screening was understandable and easily accessible						
Place of residency						
Village	4.773	5	0.516	3		
Town inhabited by less than 20,000 people	4.801	5	0.392		3.821	0.252
City inhabited by 20,000–100,000 people	4.925	5	0.254			
City inhabited by more than 100,000 people	4.751	5	0.532			
Education level						
Secondary education	4.811	5	0.381	1	1.317	0.517
Higher education	4.781	5	0.504			
I felt that I could refer any questions regarding my children's development to the people responsible for screening						
Place of residency						
Village	4.350	5	0.896	3		
Town inhabited by less than 20,000 people	3.877	4	1.088		10.794	0.013
City inhabited by 20,000–100,000 people	4.284	5	0.760			
City inhabited by more than 100,000 people	4.000	5	1.003			
Education level						
Secondary education	3.900	5	1.165	1	1.710	0.452
Higher education	4.144	5	0.927			
I would receive appropriate help or advice in case of suspicion DD from people involved in online screening						
Place of residency						
Village	4.433	5	0.778	3		
Town inhabited by less than 20,000 people	3.913	4	0.918		21.323	<0.001
City inhabited by 20,000–100,000 people	4.240	5	0.863			
City inhabited by more than 100,000 people	3.919	4	0.972			
Education level						
Secondary education	4.264	5	0.935	1	6.043	0.048
Higher education	4.051	4	0.925			
Overall, I am satisfied with my participation in electronic ASD screening						
Place of residency						
Village	4.722	5	0.639	3		
Town inhabited by less than 20,000 people	4.769	5	0.415		3.450	0.278
City inhabited by 20,000–100,000 people	4.705	5	0.544			
City inhabited by more than 100,000 people	4.655	5	0.607			
Education level						
Secondary education	4.734	5	0.614	1	2.067	0.356
Higher education	4.679	5	0.590			

Note. Answers were given on a Likert scale, where 1—I completely disagree with this sentence and 5—I completely agree with this sentence. ASD—autism spectrum disorders, DD—developmental disorders, *df*—degrees of freedom, H—Kruskal–Wallis test statistic result, M—mean, Me—median, and SD—standard deviation.

Table 5. Assessment of online ASD screening conducted in the "Spojrzeć w oczy" project by parents depending on parents' age and IT proficiency.

Variables	*r*	*t*	*p*
Age & The information available during screening was understandable and easily accessible	−0.093	−1.895	0.059
Age & I felt that I could refer any questions regarding my children's development to the people responsible for screening	−0.104	−2.140	0.033
Age & I would receive appropriate help or advice in case of suspicion DD from people involved in online screening	0.033	0.677	0.499
Age & Overall, I am satisfied with my participation in electronic ASD screening	−0.030	−0.618	0.537
IT proficiency & The information available during screening was understandable and easily accessible	0.058	1.176	0.240
IT proficiency & I felt that I could refer any questions regarding my children's development to the people responsible for screening	0.024	0.492	0.623

Table 5. *Cont.*

Variables	r	t	p
IT proficiency & I would receive appropriate help or advice in case of suspicion DD from people involved in online screening	−0.009	−0.185	0.853
IT proficiency & Overall, I am satisfied with my participation in electronic ASD screening	−0.026	−0.528	0.598

Note. ASD—autism spectrum disorders, DD—developmental disorders, *r*—Spearman's rho statistic result, and *t*—the value of the *t* statistic testing the significance of the correlation coefficient.

Collected data indicate a very high overall assessment of the online screening conducted as part of the project by parents. Opinions are characterized by high homogeneity in terms of the examined features, with a few exceptions. Parents living in small towns rated the possibility of contact and the possibility of receiving help the lowest (H = 10.794, $p = 0.013$ and H = 21.323, $p = <0.001$, respectively); similarly, people with higher education rated the possibility of obtaining appropriate help lower than parents with secondary education (H = 6.043, $p = 0.048$). Moreover, there is a very weak negative correlation indicating that older parents evaluate the possibility of obtaining further information and answers from examiners during online screening lower than younger parents (R = −0.104, $p = 0.033$).

In order to obtain doctors' opinions on electronic ASD screening and potential limitations in the implementation of ASD screening in everyday practice, doctors from the Polish Society of Family Medicine were invited to participate in the second part of this study. Finally, 95 doctors took part in this study. The exact number of Society members is not officially available; according to Facebook data, the post reached approximately 1320 users. Mean age of the doctors was 32.58 years (SD = 5.24) and mean subjective IT proficiency was 8.33 (SD = 1.32). The average length of service in primary healthcare facilities of the respondents was 5.23 years (SD = 4.68). Full doctors' sociodemographic characteristics are presented in Table 6.

Table 6. Sociodemographic characteristics of doctors taking part in this study.

Characteristics (Total N = 95)	n	%
Sex		
Female	62	65.26%
Male	33	34.74%
Main place of practicing a profession		
Village	7	7.37%
Town inhabited by less than 20,000 people	9	9.47%
City inhabited by 20,000–100,000 people	22	23.16%
City inhabited by more than 100,000 people	57	60.00%
Percentage of pediatric patients among all doctor's patients		
up to approx. 10% of total	22	23.15%
up to approx. 20% of total	27	28.42%
up to approx. 30% of total	19	20.00%
up to approx. 40% of total	6	6.32%
up to approx. 50% of total	10	10.53%
more than 50% of total	11	11.58%

Physicians were asked about their own methods of management of a pediatric patient in case of suspected developmental disorders—whether they rely on their own clinical assessment or the results of additional tests when choosing further treatment, and whether they use screening methods in their everyday work. In addition, we asked for information on whether they use given methods in all children or only those from the risk group, and what actions are taken when observing DD in a patient; finally, we asked doctors to choose their preferred ASD screening option. Chi-square tests and the Kruskal–Wallis test were used for this analysis, depending on the nature of the studied variables. The collected data are presented in Table S1, Tables 7 and 8. Table S1 is provided as Supplementary File.

Table 7. The usage of screening methods, the choice of management in case of suspected DD and preferred screening method (online vs. stationary) depending on the sex, primary place of work of physicians and percentage of pediatric patients among all patients.

Variable	Total N = 95		Sex				X^2/H*	*p*	Primary Place of Work								X^2/H*	*p*
			Females N = 62		Males N = 33				Village N = 7		Town N = 9		Small City N = 22		Big City N = 57			
	n	%	n	%	n	%			n	%	n	%	n	%	n	%		
Usage of screening methods for DD (e.g., ASD)																		
in all children	39	41.05%	28	45.16%	11	33.33%	1.246	0.265	2	28.57%	3	33.33%	7	31.82%	27	47.37%	2.388	0.496
in at-risk children or suspected of having DD	55	57.89%	33	53.23%	22	66.67%			5	71.43%	6	66.67%	15	68.18%	29	50.88%		
lack of use of screening tools	1	1.05%	1	1.61%	0	0.00%			0	0.00%	0	0.00%	0	0.00%	1	1.75%		
Management of a child under 2 years of age in the event of subtle symptoms of developmental disorders																		
referral for further diagnostics	51	53.68%	32	51.61%	19	57.58%	0.897	0.343	2	28.57%	8	88.89%	13	59.09%	28	49.12%	1.162	0.762
referral for further diagnostics at the insistence of parents	8	8.42%	4	6.45%	4	12.12%			1	14.29%	0	0.00%	2	9.09%	5	8.77%		
further observation of the child's development	36	37.89%	26	41.94%	10	30.30%			4	57.14%	1	11.11%	6	27.27%	24	42.11%		
Management of a child above 2 years of age in the event of subtle symptoms of developmental disorders																		
referral for further diagnostics	90	94.74%	57	91.94%	33	100.00%	0.538	0.463	5	71.43%	9	100.00%	21	95.45%	55	96.49%	12.786	0.005
referral for further diagnostics at the insistence of parents	1	1.05%	1	1.61%	0	0.00%			1	14.29%	0	0.00%	0	0.00%	0	0.00%		
further observation of the child's development	4	4.21%	4	6.45%	0	0.00%			1	14.29%	0	0.00%	1	4.55%	2	3.51%		
Preferred screening method																		
stationary	44	46.32%	30	48.39%	14	42.42%	0.304	0.581	3	42.86%	3	33.33%	6	27.27%	32	56.14%	6.001	0.112
on-line	51	53.68%	32	51.61%	19	57.58%			4	57.14%	6	66.67%	16	72.73%	25	43.86%		

Variable	Percentage of Pediatric Patients Among All Patients												X^2/H*	*p*
	<10% N = 22		<20% N = 27		<30% N = 19		<40% N = 6		<50% N = 10		>50% N = 11			
	n	%	n	%	n	%	n	%	n	%	n	%		
Usage of screening methods for DD (e.g., ASD)														
in all children	4	18.18%	18	66.67%	7	36.84%	0	0.00%	3	30.00%	7	63.64%	19.216	0.002
in at-risk children or suspected of having DD	18	81.82%	8	29.63%	12	63.16%	6	100.00%	7	70.00%	4	36.36%		
lack of use of screening tools	0	0.00%	1	3.70%	0	0.00%	0	0.00%	0	0.00%	0	0.00%		
Management of a child under 2 years of age in the event of subtle symptoms of developmental disorders														
referral for further diagnostics	15	68.18%	12	44.44%	11	57.89%	3	50.00%	6	60.00%	4	36.36%	10.271	0.068
referral for further diagnostics at the insistence of parents	0	0.00%	1	3.70%	2	10.53%	0	0.00%	2	20.00%	3	27.27%		
further observation of the child's development	7	31.82%	14	51.85%	6	31.58%	3	50.00%	2	20.00%	4	36.36%		
Management of a child above 2 years of age in the event of subtle symptoms of developmental disorders														
referral for further diagnostics	20	90.91%	26	96.30%	19	100.00%	6	100.00%	9	90.00%	10	90.91%	8.590	0.127
referral for further diagnostics at the insistence of parents	0	0.00%		0.00%	0	0.00%	0	0.00%	1	10.00%	0	0.00%		
further observation of the child's development	2	9.09%	1	3.70%	0	0.00%	0	0.00%	0	0.00%	1	9.09%		
Prefered screening method														
stationary	13	59.09%	11	40.74%	5	26.32%	4	66.67%	6	60.00%	5	45.45%	6.524	0.259
on-line	9	40.91%	16	59.26%	14	73.68%	2	33.33%	4	40.00%	6	54.55%		

Note. ASD—autism spectrum disorders, DD—developmental disorders, **H**—Kruskal–Wallis test statistic result, and X^2—chi-square test statistic result. * Kruskal–Wallis **H** test used only in preferred screening method analysis.

Table 8. The usage of screening methods, the choice of management in case of suspected DD and preferred screening method (online vs. stationary) depending on the age, estimated number of children with DD under medical care and length of work experience of physicians.

Variables	H	*df*	*p*
Usage of screening methods for DD (in all children/only in children at risk or suspected of DD)			
age	22.435	17	0.169
self-estimated IT proficiency	5.496	6	0.482
estimated number of children with DD under medical care	22.133	18	0.226
length of work experience in primary care facilities	6.089	14	0.964
Management of a child under 2 years of age in the event of subtle symptoms of developmental disorders			
age	15.675	17	0.547
self-estimated IT proficiency	6.881	6	0.332
estimated number of children with DD under medical care	24.169	18	0.150
length of work experience in primary care facilities	24.553	14	0.039
Management of a child above 2 years of age in the event of subtle symptoms of developmental disorders			
age	18.120	17	0.381
self-estimated IT proficiency	19.757	6	0.003
estimated number of children with DD under medical care	13.250	18	0.777
length of work experience in primary care facilities	30.081	14	0.007
Preferred screening method			
age	27.876	17	0.046
self-estimated IT proficiency	5.277	6	0.501
estimated number of children with DD under medical care	24.435	18	0.141
length of work experience in primary care facilities	24.384	14	0.041

Note. DD—developmental disorders, *df*—degrees of freedom, and H—Kruskal–Wallis test statistic result.

The vast majority of surveyed physicians use basic methods to detect symptoms of developmental disorders—the most common ones include observing the child during the physical examination (94.74%), attempting to communicate and establish contact with the child (93.68%), and assessing the pace of achieving developmental milestones (89.47%). Risks related to family history are slightly less frequently taken into account; diagnostic tools completed by caregivers or healthcare workers are used much less frequently (21.05 and 15.79% of respondents, respectively). Moreover, active use of the above-mentioned methods is more common only in children suspected of having DD features; less frequently in the entire pediatric patient population. Quite a significant part of the surveyed doctors has a wait-and-see attitude in the case of subtle features of DD in a child under two years of age (37.89%); in the case of children over two years of age, the vast majority of doctors refer children with DD symptoms for further evaluation (94.74%). These issues are independent of doctors' age, gender, place of work and age; the analyzes showed the significance of the percentage of pediatric patients among all patients on the frequency of screening use in the entire population (the highest in the "up to 20%" and "above 50%" groups, not showing a linear nature; $X^2 = 19.216$, $p = 0.002$), self-estimated IT proficiency on the frequency of referring patients for further evaluation after the age of 2) (the higher it is, the greater the percentage of referring physicians) and the length of work experience in primary care facilities (the longer it is, the more physicians refer patients for further evaluation—both below and over 2 years of age).

As in the case of parents, preferences regarding the type of screening (online vs. stationary) are strongly divided (53.68% of doctors are in favor of online screening). Dependencies were observed only in terms of age and work experience of doctors (younger doctors and those working in primary care for a shorter period more often preferred the online version ($H = 27.876$, $p = 0.046$ and $H = 24.384$, $p = 0.041$, respectively).

If the stationary version was preferred over the electronic version, we asked doctors who would be responsible for calculating the questionnaire results and analyzing the

screening results. The vast majority (n = 36, 81.82%) believe that this should be performed by the child's doctor; then the doctor with the most experience in DD (n = 6, 13.64%). Interestingly, none of the doctors would hand over this function to nurses or the clinic coordinator (in Poland, these are HCPs responsible for helping patients set appointments with specialists outside the clinic and helping with the care of chronically ill patients).

Physicians were also asked how willing they would be to use questionnaires to perform screening of DDs during individual visits (e.g., peri-vaccination visits, well-child visits, when observing DD symptoms during the visit). The Kruskal–Wallis test was used to perform this analysis. The results are summarized in Table 9.

Table 9. Willingness to use DD screening diagnostic questionnaires during specific visits at primary care facilities.

How Willingly Would You Use Tools for Screening Developmental Disorders in the Following Situations?	M	Me	SD	H	*p*
during the vaccination qualifying visit at 18 months of age	3.512	4	1.094	46.069	<0.001
during the well-child visit in 2nd year of life	3.828	4	1.022		
when noticing symptoms of developmental disorders in child during the visit	4.415	5	0.736		
when a parent expresses concerns about their child's development	4.362	5	0.735		
if the child has a sibling with ASD or another developmental disorder	4.207	4	0.787		

Note. Answers were given on a Likert scale, where 1—very reluctantly and 5—very willingly. ASD—autism spectrum disorders, H—Kruskal–Wallis test statistic result, M – mean, Me – median, SD – standard deviation,. Three respondents did not provide answers regarding this part.

The respondents' answers show a clear, statistically significant difference in the willingness to use screening questionnaires during various situations in the physicians' office ($H = 46.069$, $p < 0.001$); doctors are significantly more willing to use screening tools to confirm noticed symptoms (M = 4.362) and to confirm or dispel parents' doubts (M = 4.362). The willingness to use these methods for general screening is significantly lower (in the case of 18 months—M = 3.512; 24 months—M = 3.828).

While preparing the Polish version of the CSBS-DP ITC questionnaire, we encountered many difficulties reported by healthcare professionals (HCPs) regarding the implementation of additional ASD screening. We collected their and our observations in order to obtain a broader opinion of HCPs potentially responsible for screening. The answers are summarized in Figure 1 and Table S2. Table S2 is provided as Supplementary File.

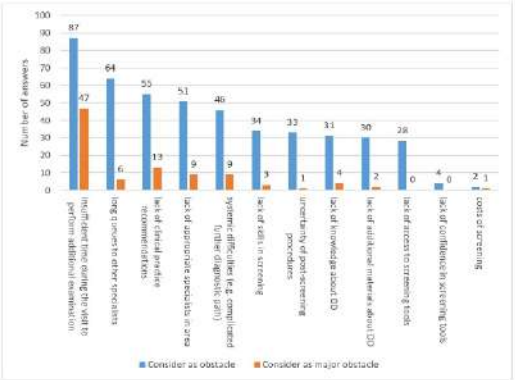

Figure 1. Obstacles to the implementation of screening in primary healthcare facilities according to surveyed physicians.

The surveyed physicians most often point out that there is insufficient time during the patient's visit to be able to carry out screening test for developmental disorders—this answer was given by almost 92% of doctors. It is also clearly the most frequently occurring most serious obstacle according to doctors; almost half of the respondents point out that excessive time burden is the main barrier to conducting population screening tests. Slightly fewer physicians report difficulties in access to specialists (both the lack of specialists and queues to practicing HCPs) and the lack of clear clinical recommendations regarding DD screening.

Parents and doctors were also asked to rate whether they agreed with the opinions about the positives and negatives of online ASD screening. These opinions were collected based on contacts with parents, specialists and the authors' own experiences. Chi-square test analysis was performed to obtain results. The respondents' answers are presented in Table 10.

Table 10. Evaluation of the advantages and positives of online ASD screening by parents and physicians.

Variable	Parents N = 418			Doctors N = 95			X^2 (df = 1)	p
	n	% of Parents	% of Total Answers	n	% of Doctors	% of Total Answers		
Which of the positives of online ASD screening are the most important to you?								
possibility of contact with healthcare professionals qualified in the field of developmental disorders	153	36.60%	13.08%	31	32.63%	13.19%	0.53	0.466
saving time on screening (possibility to perform screening at home)	285	68.18%	24.36%	61	64.21%	25.96%	0,56	0.456
no potential stigmatization of the child in the event of developmental disorders (person examining does not personally know the family/child)	118	28.23%	10.09%	30	31.58%	12.77%	0.42	0.515
possibility of performing the test at any convenient time	327	78.23%	27.95%	64	67.37%	27.23%	5.04	0.025
easier access to the screening (it is not necessary to look for people qualified to perform screening)	287	68.66%	24.53%	49	51.58%	20.85%	9.99	0.002
Total	1170	n/a	100%	235	n/a	100%		
What are the greatest difficulties in conducting online screening in your opinion?								
lack of direct (physical) contact with the examiner (e.g., doctor, nurse, psychologist)	195	46.65%	29.64%	53	55.79%	31.18%	2.59	0.108
inability to confirm the result of the screening test in clinical observation by a doctor/psychologist	242	57.89%	36.78%	50	52.63%	29.41%	0.87	0.350
the need to wait for an explanation of the obtained result	50	11.96%	7.60%	19	20.00%	11.18%	3.63	0.057
inability to quickly clarify doubts regarding the development of the child's behavior	163	39.00%	24.77%	39	41.05%	22.94%	0.14	0.711
lack of trust in people responsible for online screening	8	1.91%	1.22%	9	9.47%	5.29%	11.55	<0.001
Total	658	n/a	100%	170	n/a	100%		

Note. ASD—autism spectrum disorders and X^2—chi-square test statistic result.

Both in the groups of doctors and parents, positive features of online screening were more often marked than negative ones. What parents most often appreciate are the convenience of online screening (saving time and the possibility of conducting the test from home—68.18% and 78.23% of parents, respectively) and easier access to the evaluation. Among the negatives, parents most often point out the inability to verify the result and assess the child's development directly by a specialist (57.89%) and the lack of personal contact with examiners (46.65%). Similar results were obtained among doctors—they also noted the comfort of online screening; however, this effect was significantly lower than in

parents (p = 0.025 and 0.002). Doctors also mainly pointed out the lack of direct contact with the person conducting the examination as main flaw of online screening (55.79%). What is noteworthy is that both groups (especially parents) rarely considered a potential lack of trust in people responsible for screening as a disadvantage.

4. Discussion

Conducted study provides evidence for potential benefits of using online ASD screening. Results indicate high parental satisfaction with participation in online screening, with high willingness to participate again for subsequent children. Moreover, most parents and doctors note the need to screen children for ASD. In both groups, there is no clear preference for the screening method (online vs. stationary). However, in order to implement any of the above-mentioned screening methods, the availability of linguistically and culturally adapted tools must be ensured.

In order to increase the effectiveness of early detection of DD, numerous screening questionnaires have been created, which, as research conducted in the United States shows, may be a useful tool to increase the percentage of early detected cases of developmental disorder [17]. In order to enable the use of this method to accelerate the diagnosis of developmental disorders, a version of the CSBS-DP ITC adapted to Polish conditions was prepared by authors of this paper, which was linguistically and culturally adapted to increase the effectiveness of the tool and reduce the likelihood of obtaining incorrect results. When designing the whole project methodology, it was necessary to adapt to the dynamically changing conditions during the COVID-19 pandemic. The pandemic forced the entire study to be conducted online or via telephone consultation. Nevertheless, this form of conducting this study made it possible—apart from assessing the effectiveness of the Polish version of the CSBS-DP ITC—to examine how parents evaluate on-line screening method.

Preparing a Polish version of a fully validated ASD screening questionnaire could, at least partially, fill the gap related to the low frequency of use of diagnostic methods in the field of developmental disorders in the daily work of doctors, as evidenced by our study results. Polish doctors more often use their own clinical assessment of a child's development, based on assessing the time of reaching appropriate milestones or observing the child's behavior during visit, which, unfortunately, may result in a delay in diagnosis [17]. The use of tools in the practice of Polish primary care physicians is clearly lower than, for example, in the USA (where ASD screening programs are the most popular—63% of pediatricians use screening questionnaires in their practice, and up to 73% of all children are screened in this direction) which allows for lowering the average age of ASD diagnosis [43,44]. Another example of unintentional omissions may be the quite large percentage of Polish physicians observing the child's development in the event of subtle symptoms of developmental disorders in children under two years of age—almost half of the respondents would continue close observation or refer the child for further evaluation based on parental pressure, which may also lead to a potential delay in the detection of developmental disorders. In the case of an older child (over 2 years of age), only 5% of respondents would decide to further observe the development.

The most frequently raised issue by respondents, which may potentially hinder the implementation of a screening program for developmental disorders in Poland, is the insufficient amount of time that can be allocated for screening during a visit of a young patient. Paying attention to the occurrence of ASD symptoms in a child or conducting an appropriate screening test requires spending more time on these activities than is usually allocated to a visit to a primary care facility [45]. Insufficient time during the visit to conduct observations for ASD is also the most frequently mentioned obstacle in studies conducted in other countries, e.g., the USA, Canada or Oman [46–49]. The problem of time constraints is becoming more crucial—data from the UK show increasing workload for doctors and nurses working in primary care [50]. In Poland, this problem is probably even more severe due to the shortage of doctors and the resulting overwork—the number

of practicing doctors and nurses in Poland is among the lowest in the EU and amounts to 2.4 doctors and 5.1 nurses per 1000 inhabitants; this problem is particularly severe in small counties around large cities and in rural areas [51]. However, there is a lack of precise data on the professional burden of Polish HCPs—in one of the studies conducted during the COVID-19 pandemic, Polish healthcare workers reported high levels of burnout and stress—related to, among others, increased workload [52]. Rising costs may also be a problem, including costs related to the preparation of materials for screening (although, as the collected data indicate, according to Polish doctors, this should not be a major obstacle) or, above all, with further care for children with suspected or ultimately diagnosed ASD. Healthcare financing in Poland is lower than the EU average (6.5% to 9.9% of GDP in 2019), most of which is spent on inpatient care; funds for outpatient care are half of the average in the European Union [51]. Low expenditure on outpatient healthcare and a small number of practicing HCPs are also related to the issue of difficulties in access to specialists in the immediate area, which is pointed out by as many as 54% of doctors—psychologists, speech therapists, educators, and child psychiatrists. Over the last decade (2014–2023), the number of child psychiatrists in Poland increased from 346 to 532, while in the years 2019–2023 there was an increase in the number of patients from nearly 150,000 to over 266,000, which means that the difficulties in obtaining appropriate specialist help are constantly increasing [53]. This is an important problem because experience from other countries (e.g., Taiwan) indicates that only increasing access to ASD screening without improving access to further evaluation or therapy may cause increasing frustration and confusion in families due to the lack of a coherent procedural and diagnostic system for people at risk of DD. To address the unmet needs of families with children with ASD, resource imbalances between screening and follow-up interventions in public pediatric care settings must be simultaneously addressed [54].

Another problem potentially troubling the implementation of screening is the lack of appropriate education of HCPs and the lack of systemic activities regarding the early diagnosis of ASD—gaps in knowledge regarding ASD and the ability to use screening tools are indicated by 33 and 36% of respondents, respectively. An even greater percentage of doctors report uncertainty regarding further treatment of a patient suspected of having ASD. Insufficient knowledge about ASD among physicians is a common global problem—a study conducted in 2020 showed that only 23% of primary care physicians had sufficient knowledge about ASD, and the percentage of such doctors was higher in countries with higher income [55]. This is probably due to the lack of experience in working with people with disabilities during medical studies, the small number of classes devoted to developmental disorders, as well as the specific image of people with ASD created by the mass media [56]. The problem of a lack of knowledge is intensified by the lack of clear guidelines (either Polish or European) regarding ASD screening and further diagnostic activities. Existing American guidelines do not fully correspond to the Polish primary care setting and their recommendations are difficult to implement into practice.

In Poland, fully validated screening questionnaires for ASD are virtually unavailable, which is indicated by 30% of respondents as a barrier to the implementation of screening. In addition to the mentioned work on the CSBS-DP ITC, so far, there are only preliminary data on the Polish version of Quantitative Checklist for Autism in Toddlers (Q-CHAT). Research on most frequently used in Poland M-CHAT organized as part of the Badabada project is still in progress [57,58]. To datew, direct translations of the above-mentioned questionnaires available on the Internet were usually used without any cultural and linguistic adaptation and without a validation process. Using incorrectly prepared diagnostic tools may reduce the accuracy of the diagnostic process; the diagnostic tool should be fully adapted to the population in which it will be used so that its psychometric properties are at the highest possible level [59]. Nevertheless, the vast majority of physicians would be willing to use diagnostic materials if they were available, especially when detecting clinically significant signs of developmental disorders or to allay or confirm parental concerns. Unfortunately, there is significantly less willingness in physicians to use these tools during peri-vaccination

or well-child visits at 18 or 24 months of age, which would be the recommended course of action according to American Academy of Pediatrics guidelines [60]. Experience from preliminary screening programs in Spain and the Netherlands indicates that the use of screening methods for ASD during well-baby check-up visits increases the attendance rate [61]. Less willingness to conduct population screening may lead to delays in diagnosis, especially in the case of more subtle symptoms occurring in a child with undiagnosed DD.

Additionally, blurred responsibility for a child with DD makes it difficult to further guide the child in the diagnostic and therapeutic process. Care for a child with DD in Poland is divided into healthcare (within primary care clinics, specialist mental health clinics, community psychological care centers and early intervention centers) and educational care (within special/integrated kindergartens, psychological and pedagogical counseling centers, early childhood supporting child development and leading centers coordinating rehabilitation and care (pol. Wiodący Ośrodek Koordynujący Rehabilitacyjno-Opiekuńczy, WOKRO)). The multitude of facilities whose scope of competences are sometimes unclear and unregulated (e.g., early intervention centers do not have their own legal normative acts) means that obtaining appropriate, full assistance can be troublesome for child's guardians who are placed in a stressful situation. The difficult situation is complemented by the fact that some of the mentioned forms of support are insufficient for all those in need—for example, in Poland, there are only forty early intervention centers [62]. The situation is further complicated by the fact that due to the underfunding of the public system, parents often use private healthcare. This significantly complicates the coordination of ASD diagnosis and therapy in Poland. Similar difficulties are also observed in other European countries—a survey conducted in south-eastern European countries indicates that many parents experienced difficulties or delays in therapy due to queues to specialists, high costs or difficulties in obtaining information; difficulties also concerned problems with obtaining assistance in the field of education [63].

The results of our study prove that respondents more often pay attention to the positive rather than negative effects of electronic screening. Both the groups of doctors and parents indicated primarily the convenience of this solution method, which is consistent with previous research results on the use of teleconsultations and online services in healthcare, which unanimously note greater availability and convenience of use compared to stationary services [64]. However, parents most often pointed out as a flaw that screening is carried out solely on the basis of a questionnaire completed by the parent, without direct contact with the examiner who could observe the child (and potentially confirm or deny the diagnosis resulting from screening). This is probably related to the subjective assessment of the doctor's role as more important during the diagnostic process—not only related to the doctor's experience, but also to interpersonal and communication qualifications, as well as empathy and understanding. Research data indicate that over 50% of patients do not fully believe in medical suggestions using artificial intelligence (AI), even though AI achieves better diagnostic results in some issues [65]. Probably, even in the case of a simple screening questionnaire (such as the CSBS-DP ITC), the issue of trust in the doctor is an important aspect for parents; it was proven that patients were more willing to implement recommendations given by doctors than those given by computers [66,67]. This effect may be even more important due to the fact that the diagnosis of ASD in a child is associated with a huge caregiving burden, even greater than for parents of children with Down syndrome or type 1 diabetes [68]. Preliminary information collected during this study indicates the need to take this fact into account when designing subsequent ASD screening services.

The collected data show that for parents the ability to initially confirm or deny fears is very important—despite this, the vast majority of parents believe that ASD screening should be mandatory for every child and that they would subject their next child to a similar test—regardless of the form of screening. Interestingly, both doctors and parents show an almost equal division in preferences for online or stationary screening. In the case of implementing the former in a larger population, attention should be paid to the

possible effect of digital exclusion, observed more often among people with lower IT proficiency and from rural areas. Nevertheless, conducting diagnostic methods using electronic technologies potentially should speed up the provision of medical assistance and facilitate access to screening [69].

Despite all the positives regarding online screening described above, conducting screening with usage of online technologies is not yet popular—as of 29 November 2023, the phrase "electronic screening" is found in 275 and "online screening" in 258 scientific articles contained in the PubMed database. In the same database, only 2 studies on ASD screening can be found—one conducted in Italy, the other in China—assessing only the effectiveness of ASD online screening [70,71]. Additionally, one large study evaluates the effectiveness of screening using electronic techniques during inpatient visits [72]. We found no study addresses the issue of online screening assessment by parents or doctors. Our work is likely to be the beginning of a future discussion on the role of further online electronic services in the daily work of physicians.

The last issue worth mentioning is the fact that parents whose children were finally diagnosed with DD had previously suspected the occurrence of some developmental disorders in their children. Due to the fact that similar experiences also result from previous studies, it is worth paying attention in everyday practice to the possibilities of dispelling these concerns of caregivers as safely as possible—even in the case of negative tests and screening observations [73].

Limitations

This study has several limitations. Firstly, parents' assessment of satisfaction with the screening was in some cases carried out a considerable time after the end of participation in this study—this may result in an incomplete and slightly inadequate assessment. Moreover, the selection of the study group does not fully reflect the structure of Polish society. The answers in the section regarding parents' assessment were provided almost exclusively by mothers—this is consistent with the sex structure obtained in previous studies on the diagnostic accuracy of the CSBS-DP ITC and is probably related to the culturally preferred family model in Poland, where mothers take most of the care of offspring, especially when children they are younger [74]. Percentage of people with higher education and living in large cities took part more often in the ASD screening during the "Spojrzeć w oczy" project; their participation increased even further in this study [75]. Similarly, the largest group of surveyed doctors are those living in large urban centers, with potential underrepresentation of rural areas of Poland. A large part of the surveyed physicians were younger doctors—residents—who are also overrepresented in the study group. Ultimately, both parents and doctors participated in this study completely voluntarily—there is a risk of biasing the group among people with a keen interest in the subject of ASD, which may result in inflated results regarding, for example, awareness of ASD issues or more frequent use of additional diagnostic materials. The methods used in the statistical analysis also pose certain limitations. Due to the lack of normal distribution of the studied features, it was necessary to use non-parametric tests with lower power than parametric tests. Moreover, due to the lack of homogeneity in the diagnostic methods used by doctors or potential restrictions on the implementation of ASD screening in the Polish primary care setting only a descriptive interpretation was possible to perform.

5. Conclusions

The data collected from our study clearly prove that Polish parents and doctors expect the implementation of screening for developmental disorders as part of routine child healthcare. However, this requires resolving numerous possible obstacles—primarily time constraints, human resources and systemic difficulties. Online screening could partially facilitate access to the test and reduce the workload, but it must be introduced with taking into account limitations—especially the possibility of quick verification of the screening result using more accurate methods during stationary visits. Moreover, it will not fully

replace the doctor–patient relationship. However, it may be one of the ways to speed up the diagnostic and therapeutic process, which may improve the functioning of people affected by ASD.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/brainsci14040388/s1, Table S1: The usage of screening diagnostic methods for developmental disorders in the practice of Polish family doctors and pediatricians; Table S2: Potential obstacles to the implementation of screening in primary healthcare facilities according to surveyed physicians; File S1: Survey for parents; File S2: Survey for family doctors and pediatricians.

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12. Załączniki

12.1. Zgoda komisji bioetycznej na realizację projektu

KOMISJA BIOETYCZNA
przy
Uniwersytecie Medycznym
we Wrocławiu
ul. Pasteura 1; 50-367 WROCŁAW

OPINIA KOMISJI BIOETYCZNEJ Nr KB – 641/2020

Komisja Bioetyczna przy Uniwersytecie Medycznym we Wrocławiu, powołana zarządzeniem Rektora Uniwersytetu Medycznego we Wrocławiu nr 133/XV R/2017 z dnia 21 grudnia 2017 r. oraz działająca w trybie przewidzianym rozporządzeniem Ministra Zdrowia i Opieki Społecznej z dnia 11 maja 1999 r. (Dz.U. nr 47, poz. 480) na podstawie ustawy o zawodzie lekarza z dnia 5 grudnia 1996 r. (Dz.U. nr 28 z 1997 r. poz. 152 z późniejszymi zmianami) w składzie:

prof. dr hab. Jacek Daroszewski (choroby wewnętrzne, endokrynologia, diabetologia)
prof. dr hab. Krzysztof Grabowski (chirurgia)
dr Henryk Kaczkowski (chirurgia szczękowa, chirurgia stomatologiczna)
mgr Irena Knabel-Krzyszowska (farmacja)
prof. dr hab. Jerzy Liebhart (choroby wewnętrzne, alergologia)
ks. dr hab. Piotr Mrzygłód, prof. nadzw. (duchowny)
mgr Luiza Műller (prawo)
dr hab. Sławomir Sidorowicz (psychiatria)
prof. dr hab. Leszek Szenborn, (pediatria, choroby zakaźne)
Danuta Tarkowska (pielęgniarstwo)
prof. dr hab. Anna Wiela-Hojeńska (farmakologia kliniczna)
dr hab. Andrzej Wojnar, prof. nadzw. (histopatologia, dermatologia) przedstawiciel Dolnośląskiej Izby Lekarskiej)
dr hab. Jacek Zieliński (filozofia)

pod przewodnictwem
prof. dr hab. Jana Kornafela (ginekologia i położnictwo, onkologia)

Przestrzegając w działalności zasad Good Clinical Practice oraz zasad Deklaracji Helsińskiej, po zapoznaniu się z projektem badawczym pt.

„Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dieci"

zgłoszonym przez **lek. Mateusza Sobieskiego** uczestnika szkoły doktorskiej w Katedrze i Zakładzie Medycyny Rodzinnej Uniwersytetu Medycznego we Wrocławiu oraz złożonymi wraz z wnioskiem dokumentami, w tajnym głosowaniu postanowiła wyrazić zgodę na przeprowadzenie badania w Centrum Medycznym AD-MED. we Wrocławiu; Gabinecie Psychologicznym Natalia Popławska we Wrocławiu; Modelowej Praktyce Lekarzy Rodzinnych we Wrocławiu pod nadzorem dr hab. Marii Magdaleny Bujnowskiej-Fedak **pod warunkiem zachowania anonimowości uzyskanych danych.**

Uwaga: Badanie to zostało objęte ubezpieczeniem odpowiedzialności cywilnej Uniwersytetu Medycznego we Wrocławiu z tytułu prowadzonej działalności:

Pouczenie: W ciągu 14 dni od otrzymania decyzji wnioskodawcy przysługuje prawo odwołania do Komisji Odwoławczej za pośrednictwem Komisji Bioetycznej UM we Wrocławiu

Opinia powyższa dotyczy: projektu badawczego będącego podstawą rozprawy doktorskiej

Wrocław, dnia 21 października 2020 r.

BW

Uniwersytet Medyczny we Wrocławiu
KOMISJA BIOETYCZNA
przewodniczący
prof. dr hab. Jan Kornafel

12.2. Kwestionariusze wykorzystane w badaniu

12.2.1 – Formularz zgody dla uczestnika badania – rodzica

spojrzeć w oczy

Drodzy rodzice, dziadkowie, opiekunowie,

nazywam się Mateusz Sobieski i jestem doktorantem w Szkole Doktorskiej Uniwersytetu Medycznego we Wrocławiu. Pod opieką dr hab. Marii Bujnowskiej-Fedak oraz prof. Małgorzaty Sekułowicz prowadzę badania dotyczące oceny efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu.

Celem badania jest stworzenie oraz walidacja polskiej adaptacji kwestionariusza Communication and Symbolic Behavior Scales - Developmental Profile (CSBS-DP) autorstwa Wetherby i Prizanta dla populacji polskich niemowląt i dzieci. Kwestionariusz ten jest stosowany w krajach anglojęzycznych jako skuteczna metoda wykrywania wczesnych objawów zaburzeń ze spektrum autyzmu. Głównym celem badania nie jest diagnoza tych zaburzeń u Państwa dziecka, lecz sprawdzenie przydatności badawczej tego kwestionariusza, by w przyszłości ten kwestionariusz mógł pomagać lekarzom we wcześniejszej diagnostyce dzieci, co zwiększa ich szansę na normalne dorosłe życie.

Udział w projekcie składa się z dwóch części – pierwszą jest wypełnienie metryczki, kwestionariusza CSBS-DP oraz wyrażenie zgody na przetwarzanie danych osobistych. Druga obejmuje kontakt telefoniczny/e-mailowy po ukończeniu przez dziecko trzydziestego miesiąca życia, celem kontroli dalszego rozwoju dziecka. W każdym momencie przeprowadzanego badania mają Państwo możliwość odstąpienia od udziału w nim.

Ponieważ oryginalna wersja kwestionariusza została wystandaryzowana na populacji amerykańskich dzieci, możliwe jest niedopasowanie granicznych progów punktowych, które wskazują na potrzebę podjęcia dalszej diagnozy lub jej brak. Może się to wiązać ze stresem i niepotrzebnymi kosztami związanymi z dalszą diagnostyką ASD lub opóźnieniem rozpoznania ASD u dziecka. Korzyścią płynącą z udziału w badaniu jest możliwość bardzo wczesnego wykrycia objawów ze spektrum autyzmu, co umożliwi dziecku rozpoczęcie wczesnej, dopasowanej do jego potrzeb terapii. Jeżeli wynik uzyskany w kwestionariuszu mógłby sugerować potrzebę podjęcia dalszej diagnostyki w kierunku występowania u Państwa dziecka ASD, wraz z osobami zaangażowanymi w projekt postaramy objąć się Państwa dziecko potrzebną opieką specjalistyczną.

Należy pamiętać, że projekt ma charakter badawczy i przesiewowy – kwestionariusz nie daje stuprocentowej skuteczności w potwierdzaniu lub wykluczaniu występowania ASD u Państwa dziecka. W razie pytań lub wątpliwości bardzo proszę o kontakt e-mail (kontakt@spojrzecwoczy.pl) lub telefoniczny (791 331 648). Dziękuję za pomoc w przeprowadzeniu badania oraz wypełnienie kwestionariusza.

Z wyrazami szacunku,
Mateusz Sobieski.

spojrzeć w oczy

Formularz świadomej zgody na udział w badaniu

1. Potwierdzam, że zapoznałem/am się z Informacją o badaniu oraz Informacją o sposobie gromadzenia i przetwarzania danych osobowych i **wyrażam zgodę na udział dziecka w badaniu**.

2. Miałem/am możliwość zadawania pytań i udzielono mi niezbędnych odpowiedzi i wyjaśnień. Zostałam/em poinformowana/y o: planowanej formie i zakresie wykorzystania danych mojego dziecka, zasadach i sposobie prowadzenia badania, warunkach ubezpieczenia uczestników badania, a także zasadach przetwarzania i wykorzystania danych osobowych dotyczących mojego dziecka zgromadzonych w toku prowadzenia badania, jak również, że podstawą ich przetwarzania będzie moja zgoda, a także, że złożenie niniejszego oświadczenia jest dobrowolne, podobnie jak udział w badaniu.

3. Jestem świadomy/a zagrożeń i korzyści związanych z udziałem w badaniu.

4. Rozumiem, że udział mojego dziecka jest dobrowolny oraz że mogę się wycofać udzieloną zgodę na udział dziecka w badaniu w dowolnym momencie bez podania przyczyny, co nie spowoduje jakichkolwiek negatywnych skutków dla mojego dziecka.

5. Wyrażam zgodę, by dla kontroli poprawności wykonania projektu badawczego przedstawiciele krajowych, zagranicznych lub międzynarodowych instytucji nadzorujących badanie, mieli wgląd w moje dane osobowe oraz dokumentację medyczną (dane dotyczące mego stanu zdrowia) pod warunkiem, że są oni związani z badaniem.

6. Wyrażam zgodę na przetwarzanie danych w tym badaniu zgodnie z obowiązującym w Polsce prawem (Rozporządzenie Parlamentu Europejskiego i Rady (UE) 2016/679 z dnia 27 kwietnia 2016 r. w sprawie ochrony osób fizycznych w związku z przetwarzaniem danych osobowych i w sprawie swobodnego przepływu takich danych oraz uchylenia dyrektywy 95/46/WE).

7. Zgadzam się na przekazanie moich anonimowych danych do innych krajów, zarówno w obrębie Europy jak i poza nią.

Wiem, że przyszłości wyniki niniejszego projektu badawczego posłużą do przygotowania publikacji naukowych, a dane w nich dostępne będą użyte jedynie w postaci anonimowej.

Data i podpis osoby składającej oświadczenie .....

Data i podpis osoby przyjmującej zgodę .....

spojrzeć w oczy

Informacje dotyczące przetwarzania danych osobowych

Zgodnie z art. 13 Rozporządzenia Parlamentu Europejskiego i Rady (UE) 2016/679 (Ogólne Rozporządzenie o Ochronie Danych osobowych – RODO) informujemy, że:

1. Administratorem danych osobowych dotyczących Pana/Pani i Pana/Pani dziecka jest Uniwersytet Medyczny we Wrocławiu z siedzibą przy wybrzeżu Ludwika Pasteura 1, 50-367.

2. Administrator wyznaczył Inspektora Ochrony Danych, z którym można się kontaktować w sprawach przetwarzania danych osobowych pod adresem e-mail: IOD@umed.wroc.pl lub telefonicznie 887 420 111.

3. Dane osobowe dotyczące Pana/Pani i Pana/Pani dziecka przetwarzane będą w celu realizacji projektu badawczego „Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci" w Katedrze i Zakładzie Medycyny Rodzinnej Uniwersytetu Medycznego we Wrocławiu, szczegółowo opisanym w dokumencie „Informacja o badaniu".

4. Podstawą prawną przetwarzania Pana/Pani danych oraz danych Pana/Pani dziecka jest art. 6 ust.1 lit. a ogólnego rozporządzenia o ochronie danych osobowych z dnia 27 kwietnia 2016 r.

5. Administrator nie udostępnia Pana/Pani danych osobowych ani danych Pana/Pani dziecka żadnym odbiorcom z wyjątkiem, gdy obowiązek taki wynika z przepisów prawa powszechnie obowiązującego lub umowy zawartej z Centrum Medycznym AD-MED, Centrum Pomocy Psychologicznej lub Modelową Praktyką Lekarza Rodzinnego Maria Bujnowska-Fedak przez Administratora.

6. Administrator może powierzyć innemu podmiotowi, w drodze umowy zawartej na piśmie z Centrum Medycznym AD-MED, Centrum Pomocy Psychologicznej lub Modelową Praktyką Lekarza Rodzinnego Maria Bujnowska-Fedak, przetwarzanie Pani/Pana danych osobowych w imieniu administratora.

7. Dane osobowe będą przechowywane przez czas trwania badania oraz pełnego opracowania i wykorzystania jego wyników. Dane będą przetwarzane w celu udokumentowania wyrażenia zgody na udział w badaniu oraz w celu umożliwienia kontaktu telefonicznego/e-mailowego w sprawie ustalenia dalszego rozwoju dziecka po ukończeniu trzeciego roku życia.

8. Ma Pan/Pani prawo do żądania:
   - dostępu do treści swoich danych osobowych (także dotyczących Pana/Pani dziecka)
   - sprostowania swoich danych osobowych (także dotyczących Pana/Pani dziecka)
   - usunięcia swoich danych osobowych (także dotyczących Pana/Pani dziecka)
   - ograniczenia ich przetwarzania
   - wniesienia sprzeciwu wobec przetwarzania.

9. Posiada Pan/Pani prawo wniesienia skargi do Prezesa Urzędu Ochrony Danych Osobowych, w przypadku podejrzenia, że dane osobowe są przetwarzane z naruszeniem przepisów prawa.

10. Cofniecie zgody na przetwarzanie Pana/Pani danych osobowych lub dotyczących Pana/Pani dziecka pozostanie bez wpływu na zgodność z prawem przetwarzania tych danych, którego dokonano na podstawie zgody przed jej cofnięciem.

11. Podanie danych osobowych jest dobrowolne.

12. Decyzje nie będą podejmowane w sposób zautomatyzowany, nie będzie Pan/Pani ani Pana/Pani dziecko podlegała/ł/ło profilowaniu.

12.2.2 – Metryczka osobowa dla rodzica

Ankieta osobowa:

Dane dziecka:

Imię i nazwisko dziecka:	Data urodzenia:
Płeć dziecka:	**Miejscowość:**
O chłopiec O dziewczynka	

Dane osoby wypełniającej:

Stosunek do dziecka:	Imię i nazwisko osoby wypełniającej ankietę:
O matka O ojciec O babcia/dziadek O inna osoba z rodziny O opiekun prawny	
Adres e-mail:	**Numer telefonu:**

Dane medyczne:

W którym tygodniu ciąży zostało urodzone dziecko?	Aktualny wiek matki:	Aktualny wiek ojca:

Czy dziecko ma:

zdiagnozowane choroby genetyczne?	O tak O nie
poważne problemy zdrowotne?	O tak O nie
poważne problemy ze wzrokiem?	O tak O nie
poważne problemy ze słuchem?	O tak O nie
problemy w rozwoju ruchowym (w tym zaburzone napięcie mięśniowe)?	O tak O nie
jest/było rehabilitowane ruchowo?	O tak O nie
Czy kiedykolwiek dziecko wycofało się ze zdobytych umiejętności (np. mówienia, wskazywania na przedmiot) na okres dłuższy niż 2 tygodnie?	O tak O nie
Czy ktokolwiek w rodzinie ma zdiagnozowane zaburzenie psychiczne (w tym szczególnie zaburzenie ze spektrum autyzmu)?	O tak O nie
Czy u rodzeństwa dziecka występują poważne schorzenia zdrowotne lub psychiczne?	O tak O nie

Jeżeli na którekolwiek powyższe pytanie została udzielona odpowiedź „TAK", proszę o opisanie problemu dziecka/rodzeństwa/członka rodziny.

12.2.3 – Polska wersja kwestionariusza CSBS-DP ITC

Kwestionariusz CSBS-DP ITC

Communication and Symbolic Behavior Scales – Developmental Profile Infant/Toddler Checklist

Imię i nazwisko dziecka: **Data urodzenia dziecka:**

W którym tygodniu ciąży miał miejsce poród: **Data wypełnienia kwestionariusza:**

Osoba wypełniająca: **Pokrewieństwo z dzieckiem:**

Instrukcja dla opiekunów: Kwestionariusz ma na celu identyfikację różnych aspektów rozwoju dzieci i niemowląt. Wiele zachowań pojawiających się przed wykształceniem umiejętności mowy może wskazywać na ewentualne trudności w nauce mowy lub innych trudności rozwojowych u dziecka. Poniższy kwestionariusz rodzic/opiekun dziecka powinien wypełnić gdy dziecko jest **pomiędzy 6 a 24 miesiącem życia** w celu ustalenia czy konieczne są dalsze konsultacje dotyczące rozwoju dziecka. Opiekun może być rodzicem lub inną osobą, która codziennie opiekuje się dzieckiem. Należy zaznaczyć odpowiedzi, które najtrafniej opisują zachowanie dziecka. W razie wątpliwości proszę zaznaczyć najtrafniejszą opinię bazując na własnym doświadczeniu. **U dzieci w danym wieku niekoniecznie muszą pojawiać się wszystkie wymienione zachowania.**

Emocje i kontakt wzrokowy			
1. Czy wiesz, kiedy Twoje dziecko jest szczęśliwe, a kiedy jest zdenerwowane?	◯ Jeszcze nie	◯ Czasami	◯ Często
2. Kiedy Twoje dziecko bawi się zabawkami, czy spogląda ma ciebie, aby zobaczyć, czy je obserwujesz?	◯ Jeszcze nie	◯ Czasami	◯ Często
3. Czy Twoje dziecko uśmiecha lub śmieje się patrząc na ciebie?	◯ Jeszcze nie	◯ Czasami	◯ Często
4. Kiedy patrzysz i wskazujesz na zabawkę po drugiej stronie pokoju, czy Twoje dziecko patrzy na nią?	◯ Jeszcze nie	◯ Czasami	◯ Często
Komunikacja			
5. Czy Twoje dziecko informuje cię, że potrzebuje pomocy lub chce mieć przedmiot poza jego zasięgiem?	◯ Jeszcze nie	◯ Czasami	◯ Często
6. Kiedy nie zwracasz uwagi na swoje dziecko, czy stara się ono zwrócić twoją uwagę?	◯ Jeszcze nie	◯ Czasami	◯ Często
7. Czy Twoje dziecko stara się cię rozśmieszyć?	◯ Jeszcze nie	◯ Czasami	◯ Często
8. Czy Twoje dziecko próbuje skłonić cię do zauważenia interesujących rzeczy - tylko po to, byś spojrzał(a) na daną rzecz, a nie po to, abyś coś z nią zrobiła?	◯ Jeszcze nie	◯ Czasami	◯ Często
Gesty			
9. Czy Twoje dziecko podnosi przedmioty i daje je tobie?	◯ Jeszcze nie	◯ Czasami	◯ Często
10. Czy Twoje dziecko pokazuje ci przedmioty, nie dając ci ich?	◯ Jeszcze nie	◯ Czasami	◯ Często
11. Czy Twoje dziecko macha, by powitać inne osoby?	◯ Jeszcze nie	◯ Czasami	◯ Często
12. Czy Twoje dziecko wskazuje palcem na różne przedmioty?	◯ Jeszcze nie	◯ Czasami	◯ Często
13. Czy Twoje dziecko kiwa głową, aby zasygnalizować *„tak”*?	◯ Jeszcze nie	◯ Czasami	◯ Często
Dźwięki			
14. Czy Twoje dziecko używa dźwięków lub słów, aby zwrócić na siebie uwagę lub poprosić o pomoc?	◯ Jeszcze nie	◯ Czasami	◯ Często
15. Czy Twoje dziecko łączy pojedyncze dźwięki tworząc takie zbitki jak: *mama, gaga, papa, dada*?	◯ Jeszcze nie	◯ Czasami	◯ Często
16. Ile z poniższych dźwięków wymawia Twoje dziecko: *ma, na, ba, da, ga, ka, la, ja, ta, pa?*	◯ Żadnego	◯ 1-2	◯ 3-4 ◯ 5-8 ◯ ponad 8
Słowa			
17. Jak wiele słów Twoje dziecko wymawia w sposób, który rozpoznajesz? (np. jak *am am* na jedzenie, *tote* na kotek itp.)	◯ 0	◯ 1-3 ◯ 4-10	◯ 11-30 ◯ powyżej 30
18. Czy Twoje dziecko składa dwa słowa razem (na przykład: *daj piciu, tata pa pa*)?	◯ Jeszcze nie	◯ Czasami	◯ Często
Rozumienie			
19. Kiedy wołasz swoje dziecko po imieniu, czy reaguje patrząc na Ciebie, lub obracając się w Twoją stronę?	◯ Jeszcze nie	◯ Czasami	◯ Często
20. Ile różnych słów lub zwrotów dziecko rozumie bez gestów? Na przykład, jeśli powiesz "*gdzie jest brzuszek*", *„gdzie jest mama”*, *„daj piłkę”* lub *„chodź do taty”* (bez wskazywania), czy dziecko odpowiednio reaguje?	◯ Na żadne	◯ 1-3 ◯ 4-10	◯ 11-30 ◯ powyżej 30
Użycie przedmiotów			
21. Czy Twoje dziecko wykazuje zainteresowanie zabawą różnymi przedmiotami?	◯ Jeszcze nie	◯ Czasami	◯ Często
22. Ile z następujących przedmiotów Twoje dziecko używa w odpowiedni sposób: kubek, butelka, miska, łyżka, grzebień/szczotka, szczoteczka do zębów, ręcznik, piłka, zabawki: samochód, telefon.	◯ Żadnego	◯ 1-2	◯ 3-4 ◯ 5-8 ◯ ponad 8
23. Ile klocków (lub kółek) Twoje dziecko potrafi ułożyć jeden na drugim?	◯ Żadnego	◯ 2 klocki	◯ 3-4 klocki ◯ 5 lub więcej
24. Czy Twoje dziecko odgrywa scenariusze w zabawie (np. karmi pluszaka, kładzie lalkę spać, wkłada figurkę do samochodu)	◯ Jeszcze nie	◯ Czasami	◯ Często

Czy masz jakieś obawy dotyczące rozwoju dziecka? ◯ tak ◯ nie **Jeśli tak, proszę opisz je na odwrocie.**

12.2.4 – Kwestionariusz ASRS

ASRS®

Wersja skrócona (2–5 lat)

Sam Goldstein, Ph.D. i Jack Naglieri, Ph.D.

Imię i nazwisko dziecka: _____ Płeć: K M (Zakreślić kółkiem)

Osoba wypełniająca kwestionariusz: Rodzic Nauczyciel/Opiekun dziecka (Zakreślić kółkiem

Imię i nazwisko rodzica/nauczyciela: _____ Data badania: _____

Wypełnia tylko nauczyciel/opiekun dziecka:

Jak długo zna Pani (Pan) dziecko: _____ _____ (w latach i miesiącach)

Miejsce sprawowania opieki nad dzieckiem: _____

Data urodzenia: _____

Wiek: _____ (lata/miesiące/dni)

Instrukcja: Proszę przeczytać kolejne pytania, zaczynające się od: **„Jak często w ciągu ostatnich czterech tygodni dziecko..."**, i w każdym przypadku zakreślić kółkiem cyfrę znajdującą się w kolumnie pod słowem, które odpowiada temu, jak często obserwowała Pani (obserwował Pan) dane zachowanie. Proszę przeczytać każde pytanie uważnie i zaznaczyć, jak często zauważała Pani (zauważał Pan) dane zachowanie **w ciągu ostatnich czterech tygodni**. Proszę nie opuszczać żadnego pytania. Jeżeli będzie Pani chciała (Pan chciał) zmienić swoją odpowiedź, proszę przekreślić ją znakiem X i zakreślić kółkiem właściwą cyfrę. Proszę upewnić się, że odpowiedziała Pani (odpowiedział Pan) na wszystkie pytania.

Jak często w ciągu ostatnich czterech tygodni dziecko:	nigdy	rzadko	czasami	często	bardzo często
1. bawiło się z innymi?	0	1	2	3	4
2. patrzyło na innych podczas kontaktu z nimi?	0	1	2	3	4
3. miało trudności podczas rozmowy z innymi dziećmi?	0	1	2	3	4
4. wolało bawić się samo?	0	1	2	3	4
5. podtrzymywało rozmowę?	0	1	2	3	4
6. mówiło w dziwaczny sposób?	0	1	2	3	4
7. unikało patrzenia na ludzi, którzy się do niego odzywali?	0	1	2	3	4
8. miało trudności podczas rozmowy z dorosłymi?	0	1	2	3	4
9. zbyt silnie reagowało na głośne dźwięki?	0	1	2	3	4
10. zbyt długo koncentrowało się na jednej rzeczy?	0	1	2	3	4
11. szukało towarzystwa innych dzieci?	0	1	2	3	4
12. okazywało zainteresowanie pomysłami innych?	0	1	2	3	4
13. miało trudności w kontaktach z rówieśnikami?	0	1	2	3	4
14. rozumiało humor i żarty odpowiednie dla swojego wieku?	0	1	2	3	4
15. miało dobre kontakty z rówieśnikami?	0	1	2	3	4

12.2.5 – Kwestionariusz oceny badania przesiewowego online – zadowolenia z udziału w projektu „Spojrzeć w oczy” dla rodziców

Metryczka:

Wiek:

Płeć: ◯ kobieta ◯ mężczyzna

Miejsce zamieszkania:

- ◯ wieś
- ◯ miasto poniżej 20 tys. mieszkańców
- ◯ miasto od 20 do 100 tys. mieszkańców
- ◯ miasto powyżej 100 tys. mieszkańców

Poziom wykształcenia: ◯ podstawowe ◯ średnie ◯ wyższe

Subiektywna ocena umiejętności korzystania z technologii informatycznych (IT):

(gdzie 1 - bardzo niskie umiejętności; 10 - bardzo wysokie umiejętności)

1 2 3 4 5 6 7 8 9 10

Udział w projekcie:

1. Czy u Państwa dziecka w trakcie projektu Spojrzeć w oczy zdiagnozowano jakiekolwiek zaburzenie rozwojowe (tj. autyzm, zespół Aspergera, opóźnienie rozwoju mowy, ADHD)?

TAK NIE

Uwagi:

2. Czy korzystali Państwo z możliwości kontaktu z nami (w celu zadania dodatkowych pytań, uzyskania dalszej diagnostyki, informacji co do dalszego postępowania)?

TAK NIE

Uwagi:

3. Czy przed udziałem w projekcie słyszała Pan/Pani o zaburzeniach ze spektrum autyzmu?

TAK NIE

4. Czy przeszło Panu/Pani na myśl, że u Pani dzieci mogą występować ASD?

TAK NIE

Pytania dotyczące screeningu ASD:

1. Czy uważa Pani/Pan iż przesiewowe badanie w kierunku zaburzeń rozwojowych wśród dzieci powinno być obowiązkowe dla wszystkich dzieci podczas badań bilansowych (np. podczas bilansu 2-latka)?

TAK NIE

2. Czy ponownie skorzystałaby Pani/Pan z możliwości dobrowolnego badania przesiewowego w kierunku zaburzeń rozwojowych u innych swoich dzieci – niezależnie od rodzaju badania (stacjonarnie/on-line)?

TAK NIE

3. Czy ponownie skorzystałaby Pani/Pan z możliwości badania on-line w kierunku zaburzeń rozwojowych u innych swoich dzieci?

TAK NIE

4. Jaka metoda badania przesiewowego w kierunku zaburzeń rozwojowych jest przez Panią/Pana preferowana?

a) elektroniczna – online

b) stacjonarna – w przychodni podstawowej opieki zdrowotnej

5. Czy informacje udzielane przez badaczy były dla Pani/Pana zrozumiałe i łatwo dostępne?

1 – niejasne 5 – dobrze wyjaśnione i łatwo dostępne

1 2 3 4 5

6. Czy towarzyszyło Pani/Panu uczucie, że mogą się Państwo zwrócić z każdym zapytaniem do badaczy dotyczącym rozwoju Państwa dzieci?

1 – całkowicie się nie zgadzam 5 – całkowicie się zgadzam

1 2 3 4 5

7. Czy uważają Państwo, że w razie trudności rozwojowych u Państwa dziecka, otrzymaliby Państwo odpowiednią pomoc/poradę od osób zaangażowanych w projekt?

1 – całkowicie się nie zgadzam 5 – całkowicie się zgadzam

1 2 3 4 5

8. Czy jest Pani/Pan ogólnie zadowolona z udziału w projekcie badawczym „Spojrzeć w oczy”?

1- nie jestem w ogóle zadowolona 5 – jestem bardzo zadowolona

1 2 3 4 5

9. Jakie trudności przeprowadzenia badania w wersji elektronicznej są dla Państwa największe? (niewymagana odpowiedź)

a) brak bezpośredniego (fizycznego) kontaktu z badającym (np. lekarzem, pielęgniarką, psychologiem)

b) brak możliwości potwierdzenia wyniku badania przesiewowego w obserwacji klinicznej przez lekarza/psychologa

c) konieczność oczekiwania na wyjaśnienie uzyskanego wyniku

d) brak możliwości szybkiego wyjaśnienia wątpliwości dotyczących rozwoju zachowań dziecka

e) brak zaufania do badanych

10. Największe plusy z badania elektronicznego (niewymagana odpowiedź)

a) możliwość kontaktu z wykwalifikowanymi w zakresie zaburzeń rozwojowych pracownikami opieki zdrowotnej

b) oszczędność czasu

c) brak stygmatyzacji dziecka występowaniem zaburzeń rozwojowych (badacz nie zna osobiście rodziny/dziecka)

d) możliwość wykonania badania w dowolnym, odpowiadającym momencie

e) ułatwiony dostęp do badania (nie jest konieczne poszukiwanie osób wykwalifikowanych do przeprowadzenia screeningu)

12.2.6 – Kwestionariusz dla lekarzy – ocena barier we wdrożeniu screeningu zaburzeń rozwojowych

Ankieta dla lekarzy rodzinnych i pediatrów – bariery we wdrożeniu screeningu zaburzeń rozwojowych

Szanowni Państwo, zwracam się z uprzejmą prośbą o udział w badaniu naukowym dotyczącym identyfikacji potencjalnych barier we wdrożeniu przesiewowego badania dzieci w kierunku występowania u nich zaburzeń rozwojowych, szczególnie zaburzeń ze spektrum autyzmu (ASD). Badanie jest w pełni anonimowe.

Niniejsze badanie jest częścią projektu naukowego „Spojrzeć w oczy", którego celem jest stworzenie oraz walidacja polskiej adaptacji kwestionariusza Communication and Symbolic Behavior Scales - Developmental Profile Infant-Toddler Checklist (CSBS-DP ITC) autorstwa Wetherby i Prizanta w populacji polskich niemowląt i dzieci, który jest stosowany w krajach anglojęzycznych jako skuteczna metoda wykrywania wczesnych objawów ASD. Badanie jest przeprowadzane w ramach projektu doktoranckiego "Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci" przeprowadzanego w Katedrze i Zakładzie Medycyny Rodzinnej Uniwersytetu Medycznego we Wrocławiu.

1) Pani/Pana wiek: ..... **2) Pani/Pana płeć:** ◯ kobieta ◯ mężczyzna

3) Staż pracy w przychodni POZ (w latach): .....

4) Jakie jest Pani/Pana główne miejsce praktykowania zawodu?

- ◯ wieś
- ◯ miasto poniżej 20 tys. mieszkańców
- ◯ miasto od 20 do 100 tys. mieszkańców
- ◯ miasto powyżej 100 tys. mieszkańców

5) Jak wysoko ocenia Pani/Pan subiektywnie swoje własne umiejętności korzystania z technologii komputerowych tj. obsługa komputerów, telefonów, oprogramowania elektronicznego?
(gdzie 1 - bardzo niskie umiejętności; 10 - bardzo wysokie umiejętności) *

1 2 3 4 5 6 7 8 9 10

6) Jaki odsetek wśród wszystkich Pani/Pana pacjentów stanowią pacjenci pediatryczni (w wieku do 18 lat)?

- ◯ do ok. 10% wszystkich pacjentów
- ◯ do ok. 20% wszystkich pacjentów
- ◯ do ok. 30% wszystkich pacjentów
- ◯ do ok. 40% wszystkich pacjentów
- ◯ do ok. 50% wszystkich pacjentów
- ◯ więcej niż połowa moich pacjentów to pacjenci pediatryczni

7) Ile szacunkowo dzieci z zaburzeniami psychicznymi bądź rozwojowymi (ADHD, ASD, depresja, zaburzenia rozwoju mowy itp.) ma Pani/Pan pod swoją opieką w przychodni?

Proszę wpisać szacowaną liczbę: .....

8) W przypadku podejrzenia u Pani/Pana pacjenta występowania zaburzeń rozwojowych kierując go do specjalisty opiera się Pani/Pan przede wszystkim na:

- ◯ własnej ocenie klinicznej funkcjonowania dziecka
- ◯ wynikach badań dodatkowych (kwestionariuszy, narzędzi obserwacyjnych)

9) Czy w swojej praktyce stosuje Pani/Pan jakiekolwiek przesiewowe metody diagnostyczne w kierunku występowania u dzieci zaburzeń rozwojowych? (pytanie wielokrotnego wyboru)

- ▫ nie stosuję żadnych metod przesiewowych
- ▫ pytam o osiągnięcie typowych kamieni milowych przez dziecko (np. siadanie, wypowiadanie słów)
- ▫ uzyskuję informację nt. historii chorobowej w rodzinie (np. ASD, ADHD, inne)
- ▫ próbuję nawiązać z dzieckiem kontakt i obserwuję jego zachowanie w zakresie komunikacji i interakcji
- ▫ obserwuję zachowanie dziecka podczas badania fizykalnego
- ▫ wykorzystuję narzędzia diagnostyczne, które są wypełniane przez rodzica/opiekuna
- ▫ wykorzystuję narzędzia diagnostyczne, które wymagają oceny przez lekarza/pielęgniarkę

10) Czy wyżej wymienione metody stosuje Pani/Pan u wszystkich dzieci, czy tylko u wybranych Pacjentów:

- ◯ nie stosuję żadnej metody przesiewowej diagnostyki zaburzeń rozwojowych
- ◯ stosuję je jedynie u wybranych pacjentów (np. z większym ryzykiem, 'podejrzanych' itp.)
- ◯ stosuję je u wszystkich dzieci

11) Jakie będzie Pani/Pana postępowanie w przypadku zaobserwowania subtelnych zaburzeń rozwojowych u dziecka PONIŻEJ 2 roku życia? *

- ◯ uważna dalsza obserwacja rozwoju dziecka
- ◯ skierowanie dziecka do dalszej diagnostyki
- ◯ skierowanie dziecka do dalszej diagnostyki w przypadku nacisków wywieranych przez rodziców/opiekunów

12) Jakie będzie Pani/Pana postępowanie w przypadku zaobserwowania subtelnych zaburzeń rozwojowych u dziecka POWYŻEJ 2 roku życia? *

- ◯ uważna dalsza obserwacja rozwoju dziecka
- ◯ skierowanie dziecka do dalszej diagnostyki
- ◯ skierowanie dziecka do dalszej diagnostyki w przypadku nacisków wywieranych przez rodziców/opiekunów

13) Jak chętnie (w przypadku dostępności kwestionariuszy do wczesnej diagnostyki zaburzeń ze spektrum autyzmu (podobnych do np. Skali Depresji Becka)) użyłaby Pani/użyłby Pan narzędzi przesiewowych w poniższych sytuacjach (proszę postawić X w odpowiedniej kolumnie):

	bardzo niechętnie	niechętnie	nie mam zdania	chętnie	bardzo chętnie
podczas wizyty okołoszczepiennej w 18 miesiącu życia					
podczas bilansu 2-latka					
w momencie, kiedy zauważy Pani/Pan u dziecka objawy zaburzeń rozwojowych podczas wizyty					
w momencie, kiedy rodzic wyrazi objawy dotyczące rozwoju jego dziecka					
w przypadku gdy dziecko ma rodzeństwo z ASD lub innym zaburzeniem rozwojowym					

14) Czy w przypadku dostępności kwestionariuszy do wczesnej diagnostyki zaburzeń ze spektrum autyzmu (podobnych do np. Skali Depresji Becka) preferowałaby/preferowałby Pani/Pan wersję papierową (przeprowadzaną stacjonarnie w przychodni POZ przez pracownika przychodni) czy elektroniczną (wypełnianą przez rodzica w domu, gdzie wynik jest obliczany elektronicznie lub oceniany przez zewnętrznego specjalistę)?

- ◯ wersja papierowa (stacjonarna w POZ)
- ◯ elektroniczna (w domu Pacjenta)

14a) (Tylko w przypadku preferencji wersji papierowej) **Kto z personelu pracowników POZ powinien (Pani/Pana zdaniem) odpowiadać za interpretację wyniku tego badania:**

- ◯ lekarz prowadzący
- ◯ lekarz pediatra (jeśli dostępny)
- ◯ lekarz posiadający największe doświadczenie w zaburzeniach rozwojowych
- ◯ pielęgniarka
- ◯ koordynator
- ◯ inny pracownik przychodni

15) Jakie ograniczenia widzi Pani/Pan, które stoją na drodze do wprowadzenia przesiewowych badań w kierunku zaburzeń rozwojowych? (pytanie wielokrotnego wyboru)

- ▫ niewystarczająca ilość czasu podczas wizyty/bilansu by przeprowadzić dodatkowe badanie
- ▫ koszty związane z przygotowaniem materiałów
- ▫ brak umiejętności w wykorzystaniu dostępnych przesiewowych narzędzi diagnostycznych
- ▫ brak przekonania co do skuteczności dostępnych przesiewowych narzędzi diagnostycznych (np. zbyt niska czułość, zbyt duża ilość przypadków fałszywie dodatnich)
- ▫ brak dostępu do istniejących przesiewowych narzędzi diagnostycznych
- ▫ brak pewności co do dalszego postępowania w zakresie ewentualnego pozytywnego wyniku testu przesiewowego
- ▫ brak wiedzy na temat zaburzeń rozwojowych
- ▫ brak rekomendacji dotyczących praktyki klinicznej w zakresie screeningu zaburzeń rozwojowych
- ▫ brak odpowiednich specjalistów w regionie (psycholodzy, pedagodzy specjalni, psychiatrzy dzieci i młodzieży)
- ▫ brak klarownych, pomocnych materiałów dodatkowych
- ▫ trudności systemowe (np. skomplikowana dalsza ścieżka diagnostyczna, brak jednolitych zasad co do poszczególnych działań w danych jednostkach, np. poradniach psychologiczno-pedagogicznych)
- ▫ długie kolejki do specjalistów

16) Które z poniższych ograniczeń jest wg Pani/Pana naistotniejsze? (pytanie JEDNOKROTNEGO wyboru)

- ▫ niewystarczająca ilość czasu podczas wizyty/bilansu by przeprowadzić dodatkowe badanie
- ▫ koszty związane z przygotowaniem materiałów
- ▫ brak umiejętności w wykorzystaniu dostępnych przesiewowych narzędzi diagnostycznych
- ▫ brak przekonania co do skuteczności dostępnych przesiewowych narzędzi diagnostycznych (np. zbyt niska czułość, zbyt duża ilość przypadków fałszywie dodatnich)
- ▫ brak dostępu do istniejących przesiewowych narzędzi diagnostycznych
- ▫ brak pewności co do dalszego postępowania w zakresie ewentualnego pozytywnego wyniku testu przesiewowego
- ▫ brak wiedzy na temat zaburzeń rozwojowych
- ▫ brak rekomendacji dotyczących praktyki klinicznej w zakresie screeningu zaburzeń rozwojowych
- ▫ brak odpowiednich specjalistów w regionie (psycholodzy, pedagodzy specjalni, psychiatrzy dzieci i młodzieży)
- ▫ brak klarownych, pomocnych materiałów dodatkowych
- ▫ trudności systemowe (np. skomplikowana dalsza ścieżka diagnostyczna, brak jednolitych zasad co do poszczególnych działań w danych jednostkach, np. poradniach psychologiczno-pedagogicznych)
- ▫ długie kolejki do specjalistów

17) Które z potencjalnych pozytywów badania przesiewowego prowadzonego metodą on-line są według Pani/Pana największe? (pytanie wielokrotnego wyboru)

- ▫ możliwość kontaktu z wykwalifikowanymi w zakresie zaburzeń rozwojowych pracownikami opieki zdrowotnej
- ▫ brak stygmatyzacji dziecka spowodowane podejrzeniem występowania zaburzeń rozwojowych (badacz nie zna osobiście rodziny/dziecka)
- ▫ możliwość wykonania badania w dowolnym, odpowiadającym momencie
- ▫ ułatwiony dostęp do badania (nie jest konieczne poszukiwanie osób wykwalifikowanych do przeprowadzenia screeningu)
- ▫ oszczędność czasu

18) Jakie trudności przeprowadzenia badania w wersji elektronicznej są według Pani/Pana największe? (pytanie wielokrotnego wyboru)

- ▫ brak bezpośredniego (fizycznego) kontaktu z badającym (np. lekarzem, pielęgniarką, psychologiem)
- ▫ brak możliwości potwierdzenia wyniku badania przesiewowego w obserwacji klinicznej przez lekarza/psychologa
- ▫ konieczność oczekiwania na wyjaśnienie uzyskanego wyniku
- ▫ brak możliwości szybkiego wyjaśnienia wątpliwości dotyczących rozwoju zachowań dziecka
- ▫ brak zaufania do osób oceniających/interpretujących

12.3 – plik dodatkowy do publikacji "Reliability and validity of the Polish version of Communication and Symbolic Behaviour Scales-Developmental Profile - Infant-Toddler Checklist"

Table 12

Comparison of the results in the CSBS-DP ITC composite "Social" obtained by children from Polish and American standardization groups.

Age (months)	N (Polish)	Score obtained								
		Social								
		Polish				American				
		Mean	1 SD	1.25 SD	Median	Mean	1 SD	Median†	Z	p
6	65	9.08	2.43	3.03						
7	47	11.17	3.56	4.45						
8	57	11.47	3.31	4.13						
9	73	12.51	3.90	4.87						
10	84	14.63	4.41	5.51						
11	81	16.40	4.18	5.22						
12	98	17.82	4.07	5.09	18	19.06	3.96	19	-2,368	0.018
13	127	18.79	3.75	4.68	19	19.43	4.36	19	-0,282	0,778
14	76	18.78	4.25	5.32	19	20.62	3.52	21	-3,866	<0.001
15	94	20.33	3.13	3.91	21	21.13	3.26	21	-1,568	0,117
16	85	19.85	4.37	5.45	21	21.77	3.62	22	-4,093	<0.001
17	74	21.16	3.12	3.90	22	22.35	2.66	22	-1,834	0,067
18	87	20.59	4.00	5.00	21	21.72	3.60	22	-2,739	0,006
19	104	21.03	3.95	4.94	22	22.50	2.93	23	-4,442	<0.001
20	76	20.82	4.22	5.28	22	22.30	3.04	22	-1,611	0,107
21	62	21.37	2.88	3.59	22	22.84	2.79	23	-3,997	<0.001
22	58	21.83	3.02	3.78	23	22.23	3.45	22	0,109	0,914

23	75	21.43	3.67	4.59	22	22.20	3.04	22	-0,772	0,440
24	37	21.54	3.91	4.89	22	22.39	3.02	22	0,056	0,955

Note. Result were compared using one-sample median Wilcoxon test. For an explanation of the methodology see full text.

†Hypotetical median.

Table 13

Comparison of the results in the CSBS-DP ITC composite "Speech" obtained by children from Polish and American standardization groups.

Age (months)	N (Polish)	Score obtained								
		Speech								
		Polish				American			Z	p
		Mean	1 SD	1.25 SD	Median	Mean	1 SD	Median†		
6	65	3.65	1.57	1.96						
7	47	4.47	2.50	3.13						
8	57	5.39	1.76	2.20						
9	73	5.60	2.23	2.79						
10	84	6.46	1.90	2.37						
11	81	7.05	1.91	2.39						
12	98	7.12	2.04	2.55	7	7.81	2.13	8	-3,830	<0.001
13	127	7.97	2.19	2.74	8	8.36	2.09	8	-0,092	0,926
14	76	7.83	2.51	3.13	8	8.77	2.14	9	-3,664	<0.001

15	94	8.90	2.53	3.16	10	9.19	2.11	9	0,044	0,965
16	85	9.08	2.53	3.16	9	9.87	2.30	10	-2,966	0,003
17	74	10.02	2.15	2.68	10	10.35	2.21	10	0,929	0,353
18	87	9.31	2.68	3.35	10	10.59	2.74	11	-5,107	<0.001
19	104	9.86	2.81	3.51	10	10.93	2.74	11	-3,453	<0.001
20	76	10.49	2.84	3.54	11	11.26	2.72	11	-0,935	0,350
21	62	11.18	2.60	3.25	12	11.60	2.49	12	-2,044	0,041
22	58	11.57	2.86	3.57	12	11.84	2.18	12	-0,504	0,614
23	75	12.34	2.30	2.87	13	11.80	2.77	12	2,465	0,014
24	37	11.92	2.66	3.33	13	11.94	2.12	12	0,784	0,433

Note. Result were compared using one-sample median Wilcoxon test. For an explanation of the methodology see full text.

†Hypotetical median.

Table 14

Comparison of the results in the CSBS-DP ITC composite "Symbolic" obtained by children from Polish and American standardization groups.

Age (months)	N (Polish)	Score obtained								
		Symbolic								
		Polish				American			Z	p
		Mean	1 SD	1.25 SD	Median	Mean	1 SD	Median†		
6	65	4.00	1.43	1.78						

7	47	5.36	2.52	3.15						
8	57	5.65	1.83	2.28						
9	73	6.53	2.19	2.73						
10	84	7.75	2.48	3.10						
11	81	9.19	2.53	3.16						
12	98	10.27	2.52	3.15	11	9.58	2.62	10	1,047	0,295
13	127	11.45	2.68	3.35	12	10.44	2.63	10	5,390	<0.001
14	76	11.76	3.00	3.75	12	11.41	2.63	11	2,866	0,004
15	94	13.18	2.23	2.79	13	12.03	2.29	12	4,574	<0.001
16	85	13.58	2.66	3.32	14	13.22	2.60	13	2,870	0,004
17	74	14.61	1.86	2.32	15	13.62	2.13	14	2,856	0,004
18	87	14.08	2.57	3.21	15	13.77	2.37	14	1,118	0,264
19	104	14.75	2.34	2.93	15	13.79	2.74	14	3,929	<0.001
20	76	14.76	2.77	3.46	16	14.12	2.38	14	3,971	<0.001
21	62	15.63	1.53	1.91	16	14.89	1.90	15	3,338	<0.001
22	58	15.60	1.64	2.05	16	14.96	2.47	15	3,078	0,002
23	75	15.69	1.84	2.30	16	14.50	2.56	15	3,780	<0.001
24	37	15.76	1.75	2.19	16	15.03	1.99	15	2,750	0,006

Note. Result were compared using one-sample median Wilcoxon test. For an explanation of the methodology see full text.

†Hypotetical median.

Table 15

Comparison of the total results in the CSBS-DP ITC obtained by children from Polish and American standardization groups.

Age (months)	N (Polish)	Score obtained								
		Total								
		Polish				American			Z	p
		Mean	1 SD	1.25 SD	Median	Mean	1 SD	Median†		
6	65	16.72	3.95	4.93						
7	47	21.00	7.37	9.21						
8	57	22.51	5.51	6.89						
9	73	24.64	7.06	8.82						
10	84	28.85	7.25	9.06						
11	81	32.63	7.45	9.31						
12	98	35.30	7.41	9.26	35	36.46	7.41	36	-0,531	0,595
13	127	38.20	6.81	8.51	39	38.23	7.70	38	0,535	0,593
14	76	38.37	8.35	10.44	40	40.80	6.76	41	-2,133	0,033
15	94	42.41	6.35	7.94	43	42.36	6.28	42	1,286	0,199
16	85	42.51	7.95	9.94	43	44.86	7.40	45	-2,442	0,015
17	74	45.97	5.50	6.88	47	46.32	5.65	46	0,138	0,890
18	87	43.98	8.07	10.08	45	46.08	7.77	46	-1,146	0,252
19	104	45.63	7.96	9.95	48	47.22	7.06	47	-0,120	0,904
20	76	46.07	8.88	11.11	48	47.68	6.72	48	-0,869	0,385
21	62	48.18	5.71	7.14	49	49.32	5.60	49	-0,534	0,593

22	58	49.00	6.39	7.99	50	49.04	7.08	49	1,117	0,264
23	75	49.46	6.77	8.47	51	48.50	7.12	49	1,667	0,096
24	37	49.22	6.58	8.22	51	49.36	5.60	49	0,908	0,364

Note. Result were compared using one-sample median Wilcoxon test. For an explanation of the methodology see full text.

†Hypotetical median.

12.4 – plik dodatkowy do publikacji "Screening Accuracy and Cut-offs of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist"

Sensitivity, specificity of the Polish version of CSBS-DP

ITC using tangential method

Results using the tangential method with different probabilities of ASD occurrence (random, 1:160, and estimated from the sample) are provided in Table 1. For the group aged 9–12 months, the cost of misclassification of a child with ASD as healthy (false negative) was set as equal to 2, in the group of 13–18 months – as equal to 3, in the group of 19–24 months – as 5.

Table 1

Values of sensitivity, specificity, accuracy, positive and negative values for estimated cut-offs using analysis with minimizing expected costs using different presumed prevalences of ASD in Polish population.

Presumed prevalence	Estimated cut-off	Sensitivity	Specificity	Accuracy	PPV	NPV
Group II (children aged 9-12 months; N = 131)						
Assumed misclassification cost = 2						
0.5 (random)	21	0.750	0.862	0.855	0.261	0.981
1:160	12	0.125	1.000	0.947	1.000	0.946
estimated from the sample	14	0.250	0.984	0.939	0.500	0.953
Group II (children aged 13-18 months; N = 242)						
Assumed misclassification cost = 3						
0.5 (random)	44	1.000	0.376	0.417	0.102	1.000
1:160	18	0.188	1.000	0.946	1.000	0.946
estimated from the sample	30	0.563	0.951	0.926	0.450	0.968
Group IV (children aged 19-24 months; N = 156)						
Assumed misclassification cost = 5						
0.5 (random)	54	1.000	0.109	0.160	0.064	1.000
1:160	30	0.111	0.993	0.942	0.500	0.948
estimated from the sample	39	0.667	0.939	0.923	0.400	0.979

It should be noted that the highest values of sensitivity and specificity were achieved using the Youden method, which is consistent with the assumptions of this method. This method assumes the probability of ASD occurrence as 50%, although the probability of ASD could not be unequivocally estimated in the population of children represented by the study sample in the current study. In a situation where the probabilities reported by the WHO were adopted for the analyses, the tangent method gives non-intuitive results and is far too volatile in classifying children into particular risk groups. Usage of cut-off points determined using this method would lead to a situation where the majority of examined children would be referred for further diagnostics. This, in turn, would lead to an increase in unnecessary costs for diagnosis, an increase in the psychological burden for families caused by a potentially unfavorable diagnosis in a child, and a reduction in the availability of specialists in the field of early diagnosis and intervention (because they would have to take care of a larger group of children who would not require this type of assistance). A measurable indicator of the lack of indications for the use of cut-offs determined by the tangent method with minimizing expected costs is a significant decrease in the sensitivity of the questionnaire visible in the ROC analyses.

12.5. Oświadczenia współautorów

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Oświadczam, że w pracy: Sobieski Mateusz, Sobieska Aleksandra, Sekułowicz Małgorzata, Bujnowska-Fedak Maria Magdalena. **Tools for early screening of autism spectrum disorders in primary health care – a scoping review.** BMC Primary Care 23, 46 (2022) mój udział polegał na zbieraniu danych, analizie zebranych danych oraz opracowaniu metodologii, a także opracowaniu finalnej wersji manuskryptu oraz superwizji badania.

Oświadczam, że w pracy: Sobieski Mateusz, Wrona Sylwia, Flakus Maria, Pierchała Kamila, Sobieska Aleksandra, Podgórska Katarzyna, Wołowicz Anna, Sekułowicz Małgorzata, Bujnowska-Fedak Maria Magdalena. **Reliability and Validity of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist**. Research in Autism Spectrum Disorders 117, 102454 (2024) mój udział polegał na współtworzeniu konceptualizacji i metodologii badania, przygotowanie draftu publikacji oraz superwizji badania.

Oświadczam, że w pracy: Sobieski Mateusz, Kopszak Anna, Wrona Sylwia, Bujnowska-Fedak Maria Magdalena. **Screening Accuracy and Cut-offs of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist**. PLOS One 19(8): e0299618 (2024) mój udział polegał na współtworzeniu konceptualizacji i metodologii badania, przygotowaniu draftu publikacji oraz superwizji badania.

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Wyrażam zgodę na wykorzystanie wyżej wymienionych publikacji w rozprawie doktorskiej lek. Mateusza Sobieskiego pt. **„Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci".**

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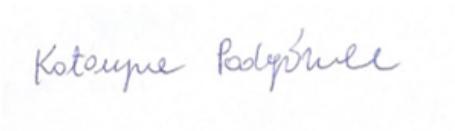

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Oświadczam, że w pracy: Sobieski Mateusz, Wrona Sylwia, Flakus Maria, Pierchała Kamila, Sobieska Aleksandra, Podgórska Katarzyna, Wołowicz Anna, Sekułowicz Małgorzata, Bujnowska-Fedak Maria Magdalena. **Reliability and Validity of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist.** Research in Autism Spectrum Disorders 117, 102454 (2024) mój udział polegał na zbieraniu danych w ramach badania, a także przygotowaniu finalnej wersji manuskryptu.

Wyrażam zgodę na wykorzystanie wyżej wymienionej publikacji w rozprawie doktorskiej lek. Mateusza Sobieskiego pt. **„Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci".**

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Cieszyn, 12.08.2024 r.

Oświadczenie o współautorstwie

Oświadczam, że w pracy: Sobieski Mateusz, Wrona Sylwia, Flakus Maria, Pierchała Kamila, Sobieska Aleksandra, Podgórska Katarzyna, Wołowicz Anna, Sekułowicz Małgorzata, Bujnowska-Fedak Maria Magdalena. **Reliability and Validity of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist**. Research in Autism Spectrum Disorders 117, 102454 (2024) mój udział polegał na współtworzeniu konceptualizacji i metodologii badania, pozyskiwania danych, przygotowania tekstu manuskryptu.

Oświadczam, że w pracy: Sobieski Mateusz, Kopszak Anna, Wrona Sylwia, Bujnowska-Fedak Maria Magdalena. **Screening Accuracy and Cut-offs of the Polish Version of Communication and Symbolic Behavior Scales-Developmental Profile – Infant-Toddler Checklist**. PLOS One 19(8): e0299618 (2024) mój udział polegał na współtworzeniu konceptualizacji i metodologii badania, pozyskiwaniu danych, przygotowaniu tekstu manuskryptu.

Wyrażam zgodę na wykorzystanie wyżej wymienionych publikacji w rozprawie doktorskiej lek. Mateusza Sobieskiego pt. **„Ocena efektywności narzędzi wczesnego wykrywania zaburzeń ze spektrum autyzmu wśród dzieci"**.

Sylwia Wrona

13. Nota biograficzna autora

Mateusz Sobieski urodził się 9 listopada 1995 roku w Opolu. W 2020 roku ukończył kierunek lekarski na Uniwersytecie Medycznym im. Piastów Śląskich we Wrocławiu. Od 2020 roku jest studentem w Szkole Doktorskiej Uniwersytetu Medycznego im. Piastów Śląskich we Wrocławiu. Od 2021 roku jest rezydentem medycyny rodzinnej – szkolenie specjalizacyjne realizuje w Modelowej Praktyce Lekarza Rodzinnego Maria Bujnowska-Fedak. Członek Polskiego Towarzystwa Medycyny Rodzinnej. Certyfikowany diagnosta ASD z użyciem narzędzi z grupy Autism Spectrum Rating Scales. W trakcie studiów doktoranckich autor lub współautor 6 publikacji naukowych, 4 doniesień zjazdowych oraz współautor podręcznika dla lekarzy „*Kiedy dziecko nie rozwija się prawidłowo. Ujęcie interdyscyplinarne*.” poświęconego zaburzeniom rozwojowym.

14. Wykaz dotychczasowych osiągnięć autora

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1. **Sobieski Mateusz**, Kopszak Anna, Wrona Sylwia, Bujnowska-Fedak Maria Magdalena: Screening accuracy and cut-offs of the Polish version of Communication and Symbolic Behavior Scales-Developmental Profile Infant-Toddler Checklist, PLoS ONE, Public Library of Science, vol. 19, nr 8, 2024, art.e0299618 [19 s.], DOI:10.1371/journal.pone.0299618, MNiSW: 100 punktów, IF: 2,9.
2. **Sobieski Mateusz**, Wrona Sylwia, Flakus Maria, Pierchała Kamila, Sobieska Aleksandra, Podgórska Katarzyna, Wołowicz Anna, Sekułowicz Małgorzata, Bujnowska-Fedak Maria Magdalena: Reliability and validity of the Polish version of Communication and Symbolic Behaviour Scales-Developmental Profile - Infant-Toddler Checklist, Research in Autism Spectrum Disorders, vol. 117, 2024, art.102454 [13 s.], DOI:10.1016/j.rasd.2024.102454, MNiSW: 140 punktów, IF: 2,2.
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2. **Sobieski Mateusz**, Bujnowska-Fedak Maria Magdalena: Walidacja polskiej wersji kwestionariusza Communication and Symbolic Behavior Scales – Developmental Profile (CSBS-DP) do wczesnego screeningu zaburzeń ze spektrum autyzmu, W: XII Kongres Polskiego Towarzystwa Medycyny Rodzinnej. Wrocław, 6-8.10.2023. Prezentacja prac naukowych [online], 2023, s. 30

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3. Bujnowska-Fedak Maria Magdalena, Grata-Borkowska Urszula, **Sobieski Mateusz**, Drobnik Jarosław: Healthcare professionals' perceptions and attitudes to teleconsultation during the Covid-19 pandemic, Patient Education and Counseling, vol. 109, nr suppl., 2023, 8 poz.O.3.6, DOI:10.1016/j.pec.2022.10.029
4. Grata-Borkowska Urszula, **Sobieski Mateusz**, Drobnik Jarosław, Fabich Ewa, Bujnowska-Fedak Maria Magdalena: Use of medical teleconsultations during the COVID-19 pandemic in Poland - preliminary results, W: eTELEMED 2021 : the Thirteenth International Conference on eHealth, Telemedicine, and Social Medicine. Nice, France, July 18-22, 2021 / Ardiansyah Prima Oky Dicky, Rajh Arian (red.), 2021, International Academy, Research, and Industry Association, ISBN 978-1-61208-872-3, s. 22-24

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