

Research Article

Advances in Neurology and Neuroscience

Screening for Autism Spectrum Disorders in Underserved Populations: Global Versus Specific Surveillance Instruments

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Submitted: 11 Sep 2022; Accepted: 20 Sep 2022: Published: 27 Sep 2022

Citation: María E Rouvier, Nora Granana, Fernando Rovira Ruiz, Ana Zeltman, and Sergio Rodriguez Gil (2022). Screening for Autism Spectrum Disorders in Underserved Populations: Global Versus Specific Surveillance Instruments. Adv Neur Neur Sci. 5(3), 155-160.

Abstract

Objective: The identification of developmental delays during well-child visits is insufficient with clinical judgment alone (30-50%).

Method: We analyzed the usefulness of clustering communication and social-personal areas from neurodevelopmental screening ASQ-3 and Denver II PRUNAPE questionnaires, compared with M-CHAT and ADOS as the initial screening for detection of Autism Spectrum Disorders (ASD) at well-child controls at primary-care settings in lowand middle-income countries (LMIC).

Results: One hundred children at the Department of Pediatrics from 12 to 66 months (average: 35 months), were assessed by a developmental pediatrician, trained and supervised by ECHO-model. The assessment was completed with a full assessment for autism with a child neurologist, and a child-juvenile psychiatrist. Thirty-one of 100 children arrived at autism spectrum disorder diagnostic criteria.

Conclusion: Failure at ASQ-3 communication plus social-personal clusters was in good agreement with M-CHAT to predict risk criteria for autism compared with a full assessment protocol. Clustering Communication and Personal-Social developmental skills from global screeners were sufficient at the pediatric visits, let differential diagnoses with global developmental delay, and communication developmental disorders. These scales covered a wide range of ages for early identification of children with an autism spectrum disorder in primary care settings within an on-site or online format.

Keywords: Asq-3, Autism, Screening, Primary-Care, Latin-American, Low-Income

Introduction

Pediatricians working in primary care must accomplish children's development [1]. They need to recognize when the milestones corresponding to each age are deviated, delayed, or eventually lost, to carry out a timely diagnosis and the corresponding intervention with the specialist. Developmental disorders are a growing diagnosis, due to increased alarm among child health professionals. Most

of these developmental disorders can be a cause of disability in adulthood, so early detection allows in many cases to apply therapeutic actions that substantially improve its evolution [2].

Developmental surveillance begins with an exhaustive interrogation of the medical history. It must be collected any noxa affecting the brain at any stage of development. The action of damaging

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agents (malnutrition, toxins, radiation, etc.) or essential substances such as thyroid hormone are critical and can offset the acquisition of higher brain functions [3].

The American Academy of Pediatrics [4] recommends that early detection be performed with a combination of surveillance in the pediatric office at 9, 18, and 30 - 36 months with standardized screening instruments and supplemented with specific surveillance for autism, trained and supervised by ECHO-model [5]. Using pediatric surveillance only 30% of children are diagnosed with developmental disorders, and less than 10% will eventually receive early intervention [3,6]. The use of systematic screening by health and education professionals fostering young children empowers early diagnoses and intervention in more than 75% of cases [7].

Developmental screening tests can be observational, parent questionnaires, or combined. Squires reached detection to more than 90% sensitivity and 91% specificity, with the Ages and Stages Questionnaire in its third version in 2009 (ASQ-3) [8]. A Latin validation for the local population has been provided [9]. It explores five areas: communication, gross motor, fine-adaptive motor, reasoning, and socio-personal developmental skills. Through a questionnaire answered by pediatricians, health professionals, teachers, or self-answered by parents, it does not need specific training and is quick to administer. Not only it saves pediatric consultation time, but also families are keen to complete a structured follow-up of children's development in more than 80% of cases [10].

In addition, ASD-specific screening materials allow parents to quickly identify and report risky symptoms and developmental concerns, with resources as developed by the Center for Disease Control [11]. The Modified Checklist of Autism in Toddlers (M-CHAT) is the most used tool for the detection of autism risk in young children aged 16 to 30 months [12]. There is also a locally validated version [13]. M-CHAT and other versions performed moderately well in accurately identifying cases of autism spectrum disorder in children referred to community child health services [14], or to track outcomes adapted to Latin American children [15].

Other assessment questionnaires that can be used early are the STAT, the Screening Tool for Autism in Toddlers and Young Children [16]. It has the advantage of early detection but requires training similarly to specific diagnostic tools, such as the Autism Diagnostic Observation Scale (ADOS) [17]. STAT has not been validated either for the local population. The Systematic Observation of Red Flags for Autism Spectrum Disorders in Young Children (SORF) as the recorded version [18] was not adopted either. There is a useful version online of the glossary, Autism Navigator [19]. Eisert and Sturner proposed a combination of instruments at different ages to improve detection specificity [20).

Denver II PRUNAPE (National Research Test) is the adaptation of the Denver II Test, fairly used as a general screener in Latin-American countries [2]. The sparse availability of time and resources for primary care providers to add instruments and the discussion as a special practice in the remuneration of care leads to the need to consider simplifying the assessment [4]. Adding multiple evaluations such as M-CHAT to the general screening with ASQ-3 reduces the applicability in daily care.

The main objective was to compare the usefulness of systematic global screening for the detection of autism between ASQ-3 and M-CHAT. The secondary objective was to make correlations between scales: ASQ-3, Denver II - Prunape and M-CHAT to assess social communication development in children at risk for autism.

Materials and Methods

A sample of 2030 Latin American children from pediatric offices by well-child check-ups over two years (20115-2017). There were 1035 boys and 995 girls. One hundred of them were referred to the developmental pediatrics office for suspected deficits in social and/or communication skills.

Inclusion criteria were healthy children over 16 months of age and under 60 months.

Exclusion criteria were preterm infants with antecedents of malnutrition, chronic or congenital systemic pathology, and children with neurological syndromes. Medical records and personal data were evaluated.

Children were screened with ASQ 3 and Prunape- Denver II as developmental screeners and M-Chat for early detection of autism. Socio-economic demographic data were assessed with the Graffar-Mendez Castellano scale.

The ASQ 3 (Ages and Stages questionnaires, 3rd edition) is a questionnaire that can be completed by parents or caregivers, in an accurate way to assess a child with a suspected developmental delay between 1 and 66 months of age [8]. It is the screening questionnaire recommended by the American Neurological Association, the Society for Child Neurology in the United Kingdom, and at the kit from UNICEF [21]. It takes between 10 and 15 minutes to complete for primary care professionals or parents and 3 to 4 minutes to convert them into a score. Scoring gives 10 points if the child can perform the skill ("yes or always"), 5 points ("sometimes"), and 0 points ("not yet") if the child cannot complete the skill. When passing the addition to the corresponding grid, it is visualized that if it coincides with the area: dark if the child is at risk, in a gray area if at risk but indications can be given to the parents and the child can be reassessed in a prudential time, and in the white area if passes or the child has no difficulties in these areas. There are 21 age-appropriate levels that assess five areas of development: communication (C), gross motor, fine motor, problem-solving, and personal-social (PS) skills. For autism deficits, we clustered failure in communication plus personal-social areas (C+PS). It was stated it is better to add global tests together with specific domains [22].

PRUNAPE: is the locally adapted version of the Denver II screening test (DDST II) [23] was used to detect developmental problems from 0 to 6 years. It explores four areas: gross motor, fine-adaptive motor, language (L), and personal-social (PS), which are evaluated through the child's performance and questions to the parents. Performance is classified as Normal, Suspicious, or Delayed Development. It is an American test, which was widely used, with good specificity but with low to moderate sensitivity levels [24]. The Latin version is called Prunape (National Screening Test) from the Denver II screening test. It is a simple tool, carried out with pre-established materials, and takes around 20 to 40 minutes to complete. It was validated in a population of 106 children in Argentina. One of its greatest advantages is that it was adapted based on its own cultural customs and was a pioneer to be used in Latin America. Its disadvantage, in addition to the lack of sensitivity of the Denver II, is that it requires specific professional training, so it can be expensive and take more time for personnel who is placed at socio-environmental risk, or who reside far away from training centers [2]. For autism screening, we included deficits in language and personal social clusters (L+PS) from Prunape Denver II.

M-CHAT: Adapted from CHAT, Checklist of Autism in Toddlers, is a questionnaire that consists of 23 items to detect symptoms of autism. It is self-administered and easy to interpret. The screening is taken to parents or caregivers, it provides a dichotomous answer: yes or no. It is designed for children between 16 to 30 months. Children who fail at three or more items or 2 or more critical items (2-7-9-13-14-15 questions) do not pass the test. It requires approximately 20 minutes, with a personal interview for parents if they have reached low-level reading skills. It showed a sensitivity of 100% and specificity of 90%, a positive predictive value of 80%, and a negative predictive value of 99%. It can be downloaded free of charge for use [12] with a local validation [13].

Graffar-Méndez Castellano method [25], is an international questionnaire used to assess the social-economic status of families. Created by Graffar from Belgium and modified by Méndez Castellano from Venezuela in 1980, considers four variables: parents' professions, maternal education level, income, and type of housing. There are 5 determined strata between 4 and 20 points assigned according to the answers (4 the highest and 20 the most unfavorable) offering the possibility of knowing the basic problems of the family under study.

The full assessment included autism criteria with DSM 5 criteria and ADOS assessment plus Vineland II adaptive quotient for all the samples [26]. All families have been adhered to, and that written informed consent for participation and publication has been obtained.

Parents who agreed to allow their children to participate in the study were asked to sign a consent form. This research has been approved by the Durand Hospital Research Ethics Committee (CODEI) within the committee's recommendations. Neither author had a conflict of interest.

The data were analyzed with the SPSS Program.

Results

From 2030 well-child visits we found a specific group of 415 who did not pass or were at risk on one or more areas. One hundred (4,9%) were derived for communication and/or personal-social clinical concerns screened with ASQ-3. Thirty five children between 16 to 30 months completed also m-CHAT questionnaire.

There were 60 males and 40 females with a 1.5/1 male/female ratio. The age range was between 12 to 58 months with an average of 35 months [Table 1].

Table 1: Sociodemographic Information

N 100	Mean	Range	SD
Age Mean (months)	35	12-58	12.23
Male/female Ratio	1.6/1		
Sociodemographic status	3.3	1-5	0.90

The whole group completed PRUNAPE Denver II developmental screening tests, and a full assessment with ADOS and Vineland II.

About sociodemographic characteristics, it was found that half of the population was between the lowest socioeconomic status (4 to 5 strata on Graffar Mendez scale), about one-third in stratum 3 and the rest in the highest social strata 1 and 2 [Table 2].

Table 2: Socioeconomic Strata

N	100
Low strata 4-5	50
Medium strata 3	23
High strata 1-2	10
Insuficient data	44

Thirty-one of 100 children with social communications deficits were accurately diagnosed with Autism Spectrum Disorder.

Statistical analysis from ASQ-3 is depicted in [Table 3]. From 44 children who failed in (C +PS) areas, 30 reached the diagnostic criteria for autism. There was one false negative. There were 14 false positive cases, that arrived at a diagnosis of language delay with deficits in personal-social skills in nine children and five with global developmental delay. The socio-demographic characteristics were proportional to the whole group.

Table 3: Comparison between all screening tests with ADOS (ages 12-54 months)

(8				
N:100	SENSITIVITY	SPECIFICITY		
ASQ-3 C+PS	0,96	0,94		
PRUNAPE L+PS	0,85	0,83		
M CHAT (N=35 from 16 to 30 months)	1	0,62		

Regarding 41 of children who failed on (L+PS) skills at PRUNAPE, 28 of them fulfilled autism criteria, and 3 were false-negative. From 13 false-positive cases, six of them were diagnosed with global developmental delay and seven with language disorders.

A group of 35 children of the sample, between 16 to 30 months satisfactorily completed M-CHAT. From 22 who screened positive, 8 were false positives and zero false negatives.

Comparing between tools, ASQ-3 and CHAT-M reached high sensitivity but ASQ-3 presented better specificity than M-CHAT for this group of age. PRUNAPE Denver II reached higher specificity but lower sensitivity [Table 3]. There were no significant differences in the sociodemographic strata with the rest of the sample.

Discussion

There is a need for early detection and intervention of developmental disorders in Latin America and few researches, as Marlow et al [27] stated in a revision about screening tools for the identification of autism spectrum disorders and developmental delay in infants and young children in low- and middle-income countries. This is a small sample but with full assessment to compare sensitivity and specificity between screening instruments.

We considered recommendations from Levy et al, on their systematic revision of good standard of practices with screening tools for autism in primary care. We adopted ASQ-3 as a relevant screening clustering failure in communication plus social-personal domains. We compared it with a credible reference standard, like M-CHAT and PRUNAPE-Denver II for local populations. ASQ-3 showed few imprecise results in a reasonable manner [28]. Early detection was feasible with a global developmental scale as ASQ-3, which was faster for the times at pediatrician offices. From 2030 children assessed, around 20% were at risk or did not pass ASQ-3 screening.

Hardy et al analyzed the assessment for autism of 56 children who were screened positively with M-CHAT R/F compared with the only communication domain from ASQ-3 • [29]. They found out that 21 of them were determined to have ASD. Using fail or monitor cutoffs on the Communication domain on ASQ-3, sensitivity was 78%, which improved to 95% with a two-stage screening procedure adding M-CHAT R/F. This study demonstrated the importance of include also the monitor cutoff points on the ASQ-3. We found out a greater sensitivity using fail or monitor cutoffs adding Communication plus Personal Social clusters, with greater specificity and covering a broader range of age than using CHAT.

The AAP recommends screening all children for symptoms of ASD through a combination of developmental surveillance at all visits and standardized autism-specific screening tests at 18 and 24 months of age in their primary care visits, because children with ASD can be identified as toddlers and timely intervention can and does influence outcomes [4,30] (AAP, 2018, Hyman et al, 2020). This autism-specific screening complements the recom-

mended general developmental screening at 9, 18, and 30 months of age [30, 31]. We included a broader range of ages, from 12 to 66 months with ASQ-3 than using M-CHAT alone. Correlation between children who completed ASQ-3 and M-CHAT was notably good between 18 and 30 months.

A systematic review by the US Preventive Services Task Force (USPSTF) concluded that the literature on existing screening tools did not demonstrate sufficient specificity to justify universal screening [32] (Siu et al, 2016). Nevertheless, our "false positive" cases included children with language developmental disorders associated with social deficits and global developmental disorders mainly, who also needed early intervention too. Initial treatment of children younger than 36 months has been shown to result in positive outcomes and participation in early intervention, and in general, is greatest among children who had received structured screening than only clinical surveillance. The whole group was followed for longitudinal outcomes, published elsewhere [33].

Windham et al suggested that ASQ is barely affected by demographic characteristics than the M-CHAT [34]. These patients correspond to a population with modest resources. It is noteworthy that 31 patients with autism followed sociodemographic proportions of the general population of an LMIC. Local adaptation to ASQ-3 let us reduce barriers that limit the reporting of symptoms of ASD.

Limitations

This study did not use the ASQ Social-emotional as Dolata et al proposed [35], because we considered reducing steps at pediatric screening, and with ASQ-3 including deficits at communication plus personal social areas concern cutoff was enough to reach sensitivity.

Similar to Hardy et al, in the current study, screening was not conducted during sick child visits, so the sample may not represent families who did not attend regular well-child exams, potentially reducing the sample's heterogeneity [29]. It must be said that in our Latin population there is a high attendance to well-child visits, around 87% [36].

We used ASQ-3 as a general screener for children referred from pediatricians only for clinical concerns on communication or social skills development. M-CHAT was completed as a second stage, so we could miss ASQ-3 negative cases. On the other hand, we included a group of children who were referred by the pediatricians only for clinical concerns.

It would be important to keep children with communication deficits under obervation, since a later confirmation of the diagnosis could occur [37, 38].

Researchers could not be blind to the screening data because all children derived from being positive at screening measures.

Future Considerations

These results suggest the utility of a general screener to identify risk for ASD using two clusters from ASQ-3. Further research should be conducted with a larger sample size, and across varied settings and demographic characteristics.

It would be of interest to assess child development applied to the general population, with more favorable economic resources.

CONCLUSIONS

Systematic neurodevelopmental screening allowed early diagnoses of children not only for autism but also for language, and global developmental disorders.

We propose ASQ-3 adding failures in communication plus personal social clusters, from general developmental scales is as effective as M-CHAT and Denver II Prunape in determining socio-communicational development disorders. It also let to differential diagnoses with language and global developmental delay in young children. These scales let save time at well-child visits keeping early detection and intervention.

Funding Source: This work was self-funded.

Competing Interests: The authors declare that they have no competing interests, financial or non-financial.

Abbreviations:

ASD: Autism Spectrum Disorder:

ASQ-3: Ages and Stages Questionnaires, 3rd version; M-CHA: Modified Checklist of Autism in Toddlers; ADOS: Autism Diagnostic Observation Scales; LMIC: Low- and Middle-Income Countries.

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