Early detection of Autism Spectrum Disorders by primary care physicians: a report on the experience of French-speaking Belgium

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Keywords

Autism spectrum disorders; early identification; primary health care

Abstract

Objective
To report on the strategy of early detection of Autism Spectrum Disorder in French-speaking Belgium and to discuss the lessons learned from that experience concerning Autism Spectrum Disorder screening programs, parents' expectations and well-adjusted health care policies.
Methods
The program relies on the existing primary health care network, a "two-visits approach" and the association of the M-CHAT-R/F questionnaire with a brief clinical assessment. We analyzed the evolution of the referral of young children between 2016 and 2019 and we interviewed the parents of 101 infants and toddlers detected during the course of the year 2019 about the support which they receive and their unmet expectations.
Results
Between 2016 and 2019, we noticed a three-fold increase in requests concerning children under 3 years old in our Center. Unfortunately, in the months following early detection of Autism Spectrum Disorder, most French-speaking Belgian families remain without sufficient support and complain of various unmet expectations.
Interpretation
A strategy of early Autism Spectrum Disorder identification aiming at improving the skills of the primary care practitioners and the network already in charge of the developmental follow-up of children may be a valuable and cost-effective approach. However, along with the improvement of early identification, providing accurate support and effective intervention is crucial for children and families.

Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication associated with restrictive and repetitive patterns of behaviors, and interests or activities which have significant consequences in daily life (1). Even if some studies have shown that early signs may be detected before the age of one year most parents notice abnormalities during the course of the first two years of life (2-4). Early identification is the first step for families towards understanding their child, obtaining information, starting a diagnostic process and accessing services and professionals. This is therefore a major concern for both parents and professionals (5). In a recent European survey, families who reported having these disorders detected at an early age expressed greater satisfaction (6). Reducing the delay in detection and diagnosis also reduces the delay before early intervention which can improve the long-term outcome, at least for some children (7,8). For all these reasons, there are worldwide attempts to lower the age of detection and the age of intervention for ASD children. In 2014, Garcia-Primo et al. published an overview of ASD screening studies and ongoing programs in Europe which showed that, even if many countries have studied different screening procedures, in most of them, it is still not part of a current practice (9). In 2019, very few European families (3,1%) reported participation in ASD-specific screening programs (6). Many important questions remain about early and systematic screening, considering uncertainties on the feasibility, reliability, costs and risks for example causing unnecessary anxiety for parents of false-positive children (10). In 2016, while the American Academy of Pediatrics (AAP) approved systematic screening, the U.S. Preventive Services Task Force concluded that there was insufficient evidence to recommend universal toddler screening for ASD (11). In 2019, the Canadian Pediatric Society also considered that there was insufficient evidence for a generalized and systematic screening for ASD (12). After children at risk of ASD have been detected, there are also uncertainties about the right intervention to propose to the right child (13-15). Nevertheless, parents ask for answers to their questions and wait for daily support and specialized interventions (16-18). This is the main challenge that clinicians, researchers and public authorities have to face.

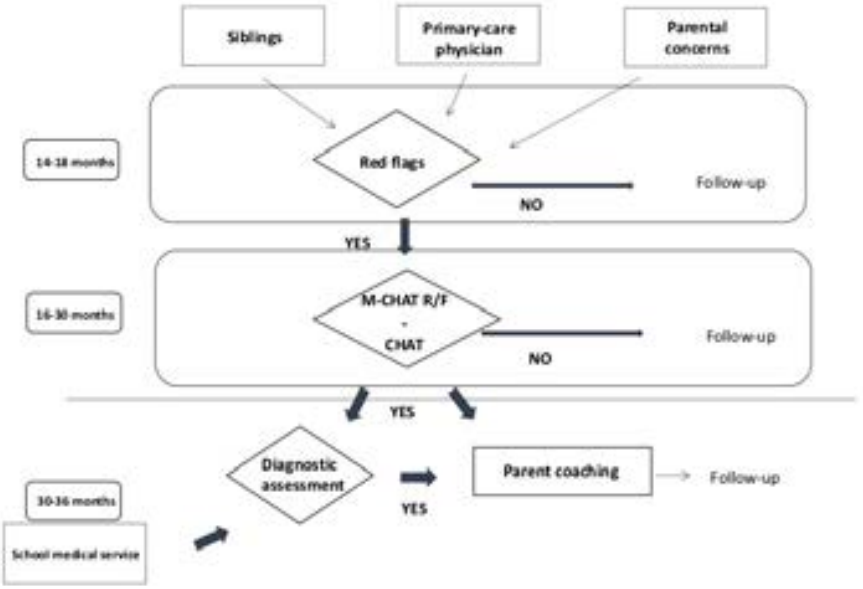
A few years ago, at the request of the governments of Wallonia and Brussels, the "Fondation SUSA" developed a program of "Early Identification of ASD and all the Communication and Social Interaction Disorders" in French-speaking Belgium. In 2016, we published an algorithm to improve detection in the Wallonia and Brussels regions (19). Rather than propose systematic screening, this program called "STARTER" relies on primary-care physicians and the clinical examination in a primary care setting. It proposes a "two-visits approach". The goal of this paper is to report on this strategy, to discuss its effects on early detection practice, to characterize the so-detected children and their families, and to precise their expectations, in order to learn from this experience about programs to develop in the future.

Materials and Methods

Description of the "STARTER" program.

The algorithm is described on figure 1. There are three gateways to enter the program, which correspond to three sub-groups of children. The first group includes the first-degree relatives of an older ASD child, who present a well-known higher likelihood of ASD. The second group is composed of children with an abnormal or atypical development noticed by a primary care provider (pediatrician, general practitioner or any professional involved in the preventive health care network). The third group is the group of children whose parents report developmental or behavioral concerns. The program states three specific moments. Firstly, we encourage primary care physicians to learn and use a short list of "red flags" which draw their attention when visiting every child aged between 14 and 18 months (Table I). These clinical signs are derived from early

Figure 1: *Algorithm of the "STARTER" early identification program*



signs described in familial videotapes, the items of the CHAT, the M-CHAT and the AOSI (20-23). As abnormal clinical signs are noticed, because of the challenge to distinguishing between a variant of development and a significant delay, and because of the poor reliability of a brief assessment especially while the child may potentially be ill or crying, we advise the primary care physician to organize a second appointment a few weeks later (24). This second appointment should be dedicated to a brief developmental assessment and the use of the M-CHAT-R/F, a specific questionnaire for the detection of ASD (22). If the previous concerns have been confirmed at the time of this second assessment, including a M-CHAT-R/F score ≥ 3, we encourage the referral of the child to a specialized team for a more complete assessment and a diagnostic process. Finally, we collaborate with school medical services for the identification of suspect children at the time they enter nursery school. For all of these "so-detected children", because of important waiting lists for most specialized services, we propose to the parents a few sessions of parent-coaching. From 2016 to 2019, we communicated these recommendations at numerous meetings with pediatricians and instructed them by using videos illustrating typical and atypical toddlers' behaviors or developmental milestones. We also contributed to the teaching program of the nurses in charge of the follow-up of infants and toddlers in the preventive health care network, and we created a program of e-learning on a digital platform for primary care physicians.

Procedure

All the children under 3 years referred to the "Fondation SUSA" during the year 2019 because of a suspicion of ASD or because of a sibling with ASD were registered. Socio-demographic data was collected. If not available, the M-CHAT-R/F questionnaire including the follow-up part was completed by a psychologist from our team for all the children between 16 and 36 months. Children with a M-CHAT-R/F score ≥ 3 were sent to services providers in the community. A next appointment was scheduled 2-3 months later to provide educational advises and to ensure that the family obtained effective support. At this time, the psychologist interviewed the parents to find out what support they received and which of their expectations were not met. Parents who did not attend this visit were contacted by phone.

Measures

Sociodemographic questionnaire

To assess common socio-demographic information, we used a questionnaire specifically designed for this study. The following data was registered: sex, age at the time of the first visit, origin of referral, ASD siblings, languages spoken at home.

Table 1. Red flags

- Social or communicative regression
- Parental concerns on social and communication development
- Autism Spectrum Disorder in siblings
- Poor visual contact
- Absence of response to the name
- Lack of joint attention
- Lack of pointing, showing objects
- Lack of imitation
- Lack of pretend to play
- Stereotyped behaviors or gesture

- Absence of babbling, pointing and social gesture at 12 months
- Absence of words at 18 months
- Absence of non-echolalic words association at 24 months

Screening of ASD

To assess the risk of ASD, we used the M-CHAT-R/F questionnaire which encompasses 20 yes/no questions for the parents. Children who score < 3 are in the low-risk range and only need to be rescreened if they are younger than 24 months. If children score ≥ 3, parents are asked structured follow-up questions to obtain additional information and examples of at-risk behaviors. Children whose total score was ≥ 3 initially and ≥ 2 after follow-up have a 47.5% risk of being diagnosed with autism spectrum disorder and a 94.6% risk of any developmental delay or concern. Children who score in the high-risk range may bypass the follow-up (22).

Support expectations questionnaire

Based on the characteristics of our population of toddlers presenting a suspicion of ASD and on our focus on the expectations of support, we created our own simplified questionnaire. We proposed to the parents a pre-determined list of support services and professionals, inspired by the literature and our experience of locally available resources (Table II) (8,15,25). We asked them to answer two questions: (1) "At this time, what kind of support services from that list are you expecting for? Select one or more items". (2) "Rank your selected items from 5 for your highest priority up to 1 for your lowest". We summed up all the scores to establish a global ranking of the parents' expectations.

Table 2 . Parents' expectations

	N = 51 (%)	
		"First priority"
• Professionalized intervention	27 (53)	15 (29,5)
• Daycare setting or school	17 (33,5)	8 (15,5)
• Parent coaching about:		
- communication	44 (86)	21 (41)
- behavior	16 (31,5)	3 (6)
- feeding problems	18 (35)	3 (6)
- sleep disorders	5 (10)	0 (0)
- daily skills	7 (14)	1 (2)
• Administrative and financial support	3 (6)	0 (0)
• Emotional support for family members	1 (2)	0 (0)

Results

Between 2019 January the 1st and December the 31st, 101 families made a contact with our Reference Center concerning a suspicion of autism for a child aged under three years. The sex ratio was 3,2 (77% males - 24 % female) as is usual (1). Fifty-nine children were referred by a medical practitioner, either a family doctor, a pediatrician or a physician working in the preventive health care network. Twenty children were followed because of an older ASD sibling. Twenty-two were referred by another professional (psychologist, teacher or social worker at the nursery, primary care provider).

At the time of the first contact with our Center, the majority of all the children (75%) were aged between 16 and 36 months, the age at which our program was supposed to help identification by using "red flag" signs. The remaining 26 children were younger than 16 months. Sixteen of them were followed on a systematic basis because of an older ASD sibling, and 10 presented developmental difficulties that raised concerns in parents.

Among the 101 families, 35 were multicultural and were talking more than one language at home.

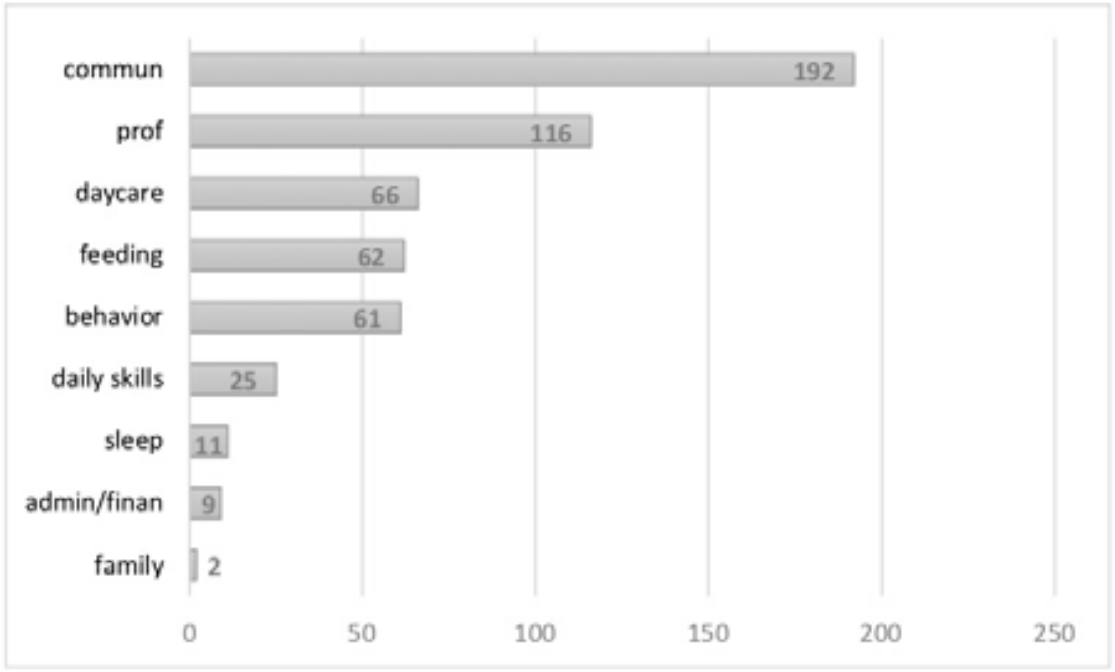
Among the 75 families of children older than 16 months, 70 completed the M-CHAT-R/F, 3 dropped out and 2 questionnaires were not administrated. Sixty M-CHAT-R/F on 70 (86%) scored ≥ 3, which constitutes a moderate to severe risk for ASD or another developmental disorder. When we questioned the parents about the support they obtained between 2 and 3 months after the M-CHAT-R/F had been completed, the majority (68%) declared one hour or less a week, whatever the support (psychologist, speech therapist, physiotherapy, home-based support). None received more than 4 hours support a week. The mean time of support was about one hour a week [mean=1.11; SD=1.21]. Of the 60 families of M-CHAT-R/F positive children, 9 (15%) expressed no need for help because they weren't worried anymore or they wanted to let their child grow up before intervention. 51 families were questioned about their expectations for support and intervention (Table II). For 41%, the first priority is about a support to improve their child's communication and to help them understand him. This is a request from 86% of the parents. A majority (53%) expects to find a professional qualified to provide individual care to the child. This is the priority for 29,5% of parents. One third of parents expect a daycare setting or a nursery school (33,5%), coaching for behavioral issues (31,5%) and feeding problems (35%). The ranking of the higher-priority expectations confirmed a priority given to parent-coaching for communication, qualified professionals for individual care, daycare setting and coaching for feeding and behavioral problems (Figure 2).

Discussion

In terms of public health policy, a program of early identification is recommended on the following conditions: (1) a reliable approach, with good predictive properties, (2) large, easy and cost-effective implementation, (3) a demonstrated benefit of early intervention, with as few side-effects as possible (4) an available and affordable intervention for all the detected children who deserve it. All over the world, each country attempts to respond to that challenge, within its own social, cultural and financial conditions. In French-speaking Belgium, we propose a strategy to improve the early detection of ASD, relying on the existing primary care medical network, a "two-visits approach" and the association of the M-CHAT-R/F questionnaire with a brief clinical assessment. At the age of 14 months, most babies are seen in routine medical visits for preventive or curative care and clinical signs are reliable so that the ASD identification becomes stable (26). However, regular appointments usually do not last more than a few minutes and the conditions for an effective developmental assessment may not be met. Children may not demonstrate characteristic symptoms in a brief observation (24). The algorithm of the STARTER program proposes therefore that a next appointment should take place a few weeks after "red flags" have been waved. In between, parents can draw their attention to some developmental milestones or behavioral targets, and the developmental trajectory of the child can be taken into account. During this second visit especially dedicated to the developmental assessment, parents are invited to fill-in the M-CHAT-R/F questionnaire and the practitioner to develop activities to assess social interaction, according to the "observation items" of the CHAT (21). The M-CHAT-R/F is the most studied and widely used tool for screening ASD. It was revised and completed in 2014 to improve specificity while maintaining a high sensitivity (22). With a score ≥ 3, 47,5 % of children are diagnosed with ASD, and 94,6 % of screen-positive cases present developmental delay or concerns. In the guidelines published in 2020 by the AAP, primary care providers are tasked with identifying all children who would benefit from early intervention, not only children at risk of ASD (27). It is important to identify all developmental delays in children with referral for appropriate diagnostic evaluation and intervention. 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 (27). In our program, the "two-visits approach" and the combination of a validated questionnaire with a brief clinical assessment may improve the efficacy of the identification process (24). The STARTER program involves five steps : (1) identification of "red flag" clinical signs from 14 months ; (2) "development appointment" a few weeks later; (3) parental questionnaire M-CHAT-R/F; (4) simple activities for eliciting and assessing social interactions; (5) if confirmed, specialized service for a more complete assessment and diagnosis. Two years after our first publication, the French Health Authority published guidelines for early detection of ASD children that are quite similar to ours (28).

In the years following the publication of the STARTER program and its guidelines, we recorded a significant increase in requests concerning children under three years to our Reference Centre - from 36 children in 2016 to 101 children in 2019 - confirming that our methodology was effective and contributed to better detection. Unfortunately, these data only concern our Centre, as the current system of data collection doesn't allow us to determine if the same observations were made in other Centers and whether this increase can be partially attributable to the program or to other factors. Nevertheless, the efficiency and the cost-effectiveness of this strategy can be considered to be excellent because the program relies on an existing health care network and requires neither the creation of additional services nor the investment of new

Figure 2: High-priority score
Items correspond to the parents' expectations listed in Table II.



resources. It aims to help primary care physicians fulfill their preventive tasks in identifying early developmental delays or abnormalities, as requested by various international recommendations (11,12,28). Rather than a systematic screening, a strategy relying on the awareness of the primary care providers and the parental concerns would probably miss some children, especially in families underestimating the benefit of a regular follow-up or whose access to the health care system is poorer. On the other hand, a relationship of trust between parents and practitioner enables a favorable context for a stressful announcement. Instead of developing new and expensive programs, we demonstrated that improving the skills of the medical practitioners and the network already in charge of the developmental follow-up of children may be a valuable approach.

During the year 2019, 58% percent of children were referred by a medical practitioner but most M-CHAT-R/F questionnaires were initially either not completed or not transmitted. The questionnaire and the follow-up part were therefore administered by a psychologist from our team. We can hypothesize that an information campaign and red flags awareness are the most useful ways to improve the knowledge of primary-care physicians, but that the use of structured and systematic questionnaires doesn't fit with standards in current practice in French-speaking Belgium. In the group of children aged between 16 and 36 months at the first visit, 70 M-CHAT-R/F questionnaires were completed and 86% were ≥3, which means a significant risk of ASD or developmental delay. Thus, we can conclude that most of the children were referred for relevant reasons and that the awareness of primary care physicians is good, probably partially improved by the "STARTER" program.

Once early signs of ASD have been detected, parents often have to wait and sometimes to struggle to find professionals and services, notably in French-speaking Belgium. In our series, most of parents (68%) do not find more than one hour of support per week. Whilst it is well-recognized that an early intervention could improve the outcome for some children and reduce unfavorable behavioral consequences , there is a lack of available and qualified professionals, affordable and accessible services (29,30). In such conditions, implementing a large-scale screening program may be questionable because, instead of improving the parents' daily lives, it could increase the level of stress and hopelessness. In order to compare the support obtained with the unmet expectations of the families, we questioned the parents, asking them to select one or more types of support services and professionals on a pre-determined list and to rank them according to their priority. From the answers of the parents, we learned the following lessons: (1) parents' expectations largely differ from one to another, (2) most of parents (86%) long to understand their child and help him to communicate. This is the priority for 41% of parents, (3) the majority of parents struggle to find and access qualified professionals: two or three months after suspicion of ASD has been raised, 68% of parents obtained less than one hour support per week, 53% expect to obtain more individualized and specialized support and 29,5% say it is a priority. (4) 35% of parents consider that feeding their child is challenging and ask for advice or support. (5) 33,5% of parents are waiting for a place in a daycare setting or a nursery, because they consider that their child needs more intensive and ongoing intervention, because they do not feel confident in their ability to provide the best care for their child themselves, or because they have to remain in active employment by choice or necessity. (6) 31,5% of parents complain of behavioral difficulties that need to be addressed. Previous studies have already demonstrated that expectations and needs largely differ according to different variables, depending on both the child and the family but also cultural and socio-demographical factors (31). Therefore, well-adjusted health care policies have to take into account not only the characteristics of the children but also the social context. This study allows us to obtain accurate data on this specific population, in order to develop accurate and individualized intervention. It confirms that a "one size fits for all" approach is irrelevant. It also emphasizes that along with the improvement of early identification, investment in support services and intervention programs should be a priority for governments.

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