

University of Groningen

Detection of autism in childhood

van 't Hof, Maarten

DOI:
[10.33612/diss.159007941](https://doi.org/10.33612/diss.159007941)

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version
Publisher's PDF, also known as Version of record

Publication date:
2021

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):
van 't Hof, M. (2021). *Detection of autism in childhood*. [Thesis fully internal (DIV), University of Groningen]. Parnassia Groep. <https://doi.org/10.33612/diss.159007941>

Copyright

Other than for strictly personal use, it is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), unless the work is under an open content license (like Creative Commons).

The publication may also be distributed here under the terms of Article 25fa of the Dutch Copyright Act, indicated by the "Taverne" license. More information can be found on the University of Groningen website: <https://www.rug.nl/library/open-access/self-archiving-pure/taverne-amendment>.

Take-down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Downloaded from the University of Groningen/UMCG research database (Pure): <http://www.rug.nl/research/portal>. For technical reasons the number of authors shown on this cover page is limited to 10 maximum.

Autism spectrum disorder alertness in Dutch youth and family center physicians: Effects of a live online educational program

Maarten van 't Hof^{1,2}, Annemyn D.Y. van Nieuwenhuyzen¹, Ina van Berckelaer-Onnes^{1,3},
Mathijs Deen^{2,4}, Hans W. Hoek^{2,5,6}, Wietske A. Ester^{1,2,7}

1. Sarr Expert Centre for Autism, Lucertis Child and Adolescence Psychiatry, Dynamostraat 18, 3083 AK Rotterdam, The Netherlands,
2. Parnassia Psychiatric Institute, Kiwistraat 30, 2552 DH, The Hague, The Netherlands,
3. Faculty of Social and Behavioural Sciences, Clinical Child and Adolescent Studies, Leiden University, Wassenaarseweg 52, 2333 AK, Leiden, The Netherlands,
4. Faculty of Social and Behavioural Sciences, Institute of Psychology, Methodology and Statistics Unit, Leiden University, Wassenaarseweg 52, 2333 AK Leiden, The Netherlands,
5. Department of Psychiatry, University Medical Center Groningen, University of Groningen, Hanzeplein 1, 9713 GZ Groningen, The Netherlands,
6. Department of Epidemiology, Mailman School of Public Health, Columbia University, 722 West 168th St., New York, New York 10032, USA,
7. Department of Child and Adolescent Psychiatry, Curium-LUMC, Leiden University Medical Center, Endegeesterstraatweg 27, 2342 AK Oegstgeest, The Netherlands.

This chapter is published as:

Van 't Hof M, van Nieuwenhuyzen ADY, van Berckelaer-Onnes I, Deen M, Hoek HW, Ester WA. Autism spectrum disorder alertness in Dutch youth and family center physicians: Effects of a live online educational program. *Journal of Autism and Developmental Disorders*. 2021. doi:10.1007/s10803-020-04842-4

ABSTRACT

We investigated the effect of a live online educational program in 93 Dutch Youth and Family Center (YFC) physicians who were screening for Autism Spectrum Disorder (ASD) in the general child population. The educational program raised the physicians' level of specific ASD knowledge and it remained higher at six months follow-up ($p < .01$). Their self-confidence in detecting ASD was also higher and maintained at follow-up ($p < .01$). The educational program had no effect on the physicians' stigmatizing attitudes toward mental illness nor on the number of potential ASD referrals in children of 4-6 years of age. In conclusion, the online educational program on early detection of ASD has a six month long effect on YFC physicians' level of ASD knowledge and self-confidence.

Keywords

Autism spectrum disorder, screening, educational program, preventive care physicians

INTRODUCTION

Autism spectrum disorder (ASD) is a complex neurodevelopmental disorder with an estimated prevalence of one in 54 (1.85%).¹ ASD often co-occurs with other psychiatric disorders (e.g. anxiety disorders, depressive disorders)² and has a substantial impact on lifespan quality of life.³

The global mean age at ASD diagnosis is currently reported to be 60.48 months (range 30.9–234 months).⁴ The age at ASD diagnosis varies between countries and regions and is affected by multiple clinical (e.g. type of ASD/severity), sociodemographic (e.g. ethnicity), parental (e.g. age), health care system and cohort factors.⁴ The age at ASD diagnosis in The Netherlands is 50-234 months,⁵⁻⁷ thus in the upper part of the reported range. There is a need to improve the early detection of ASD in children in the Netherlands as early treatment improves treatment outcomes.^{8,9}

Preventive care professionals, general practitioners (GPs) and medical specialists play an important role in the early detection of ASD.¹⁰ In the Dutch healthcare system, Youth and Family Centers (YFCs) provide free preventive healthcare to all children living in the Netherlands, with regular consultations with physicians and nurses at set times up to the age of 18. In addition to screening for developmental delays, part of the YFC's program is to screen for (potential) social, psychological and somatic disturbances,^{11,12} which means they have a significant role in the early detection of ASD. Just before the start of this study, a national ASD screening guideline for YFC physicians, which includes a routine developmental screening and an ASD screening questionnaire, was published in 2015.¹¹ The guideline emphasizes attention on children of school age, since behavioral problems can become easier to signal in adaptation to group situations and play between peers. Schooling is obligatory for children from 5 years old in the Netherlands and most children aged 4 years start primary school. Corresponding to this transition, YFC's have a fixed consultation for children aged 4-6 years, with screening for developmental delays as part of the consultation. As all children in the Netherlands attend a YFC, it is of utmost importance that YFC physicians can recognize the early signs of ASD and know how to refer suspected cases.

There has been little research on the role of preventive care physicians (e.g. YFC physicians) in the process of early detection. Research in related primary care providers found inconsistencies in knowledge of ASD and screening procedures around the detection of early signs of ASD.^{13,14} More recent research has indicated that Dutch YFC physicians have insufficient levels of specific ASD knowledge and reasonably positive attitudes toward mental illness.¹⁵ However, little is known about the effect of an early detection program on the ASD alertness of YFC physicians.

O'Brien et al.¹⁶ pointed out that a lack of emphasis on mental health in medical training and a lack of competence in recognizing mental health problems in young people are leading to skill problems in primary care professionals. One study investigating the effect of an elaborate ASD identification- and

ongoing care learning program found it had a positive effect on physicians' self-efficacy in providing care to children with ASD.¹⁷ However, to our knowledge, no research has been done into the effect of an educational program on physicians' self-confidence regarding the detection of ASD. Although previous studies indicate that educational programs for primary care providers (including YFC physicians),^{10,18} pediatricians, GPs,¹⁹ and child and adolescent psychiatrists²⁰ have a positive effect on the age at ASD diagnosis, this effect was not sustained at follow-up. These findings demonstrate the importance of the continuity of training for health professionals and for finding strategies to preserve this positive effect.

GPs, physicians and pediatricians often report low levels of self-confidence in addressing concerns about early development and making appropriate referrals for children with ASD.^{17,21,22}

Implementation of a parent-completed screening tool increased pediatricians' developmental referral rates in children aged 12-24 months²³ and developmental and behavioral detection rate in pediatric practices in children aged 2-3 years.²⁴ A retrospective study found that introducing a routine screening method during well-child visits increased the number and age of developmental and behavioral delays referrals between ages 6-60 months.²⁵

Lack of skills and awareness was suggested to translate into psychiatric stigmatization by primary healthcare professionals, inducing feelings of anxiety and a preference for avoiding patient-provider interactions.²⁶ Psychiatric stigma in healthcare providers has a negative influence on patients' access to mental healthcare as various studies have shown that providers are an important source of stigma towards mental illness.²⁷ Dutch physicians appear to have a positive attitude towards mental illness, although they are less positive than other Western healthcare professionals.¹⁵ Research on the effect of stigma-reducing interventions for primary care physicians and nurses found they had a significant effect.^{28,29} However, we found no studies on what effects education can have on stigmatizing attitudes toward mental illness in physicians screening in the general child population.

Our study therefore evaluated the 6-month effect of a live online educational program on ASD for YFC physicians. Based on previous studies, we hypothesized that the educational program would enhance their level of ASD knowledge, self-confidence, and potential ASD referrals in children aged 4-6 years, and that it would reduce stigma toward mental illness.

METHODS

Study design and population

We present the results of the Live Online Learning (LOL) educational program called *Detection of Autism Spectrum Disorders in Children Aged 4-6 Years by Youth & Family Center Physicians*. The educational program was developed as part of the "Reach-Aut Academic Center for Autism:

Transitions in Education” project. All physicians employed by the YFCs in the Greater Rotterdam area took the educational program between January-December 2016. Written informed consent was obtained from all participants. The Medical Ethics Committee of Leiden University Medical Center approved the study and classified it as not being subject to the Medical Research Involving Human Subjects Act (WMO) (REACH-AUT No. 70-73400-98-002).

Procedure

The online educational program (group size 8-11 participants) was obligatory for all 93 physicians of the Youth and Family Centers in the Greater Rotterdam area between January and November 2016. All physicians were asked to complete online questionnaires at pre-, post and 6 month follow-up measurement. We assessed ASD knowledge and self-confidence regarding the detection of ASD at pre-, post-, and follow-up measurement. Stigmatizing attitudes toward mental illness was assessed at pre-, and follow-up measurement, educational program satisfaction at post measurement and demographic measures at pre measurement.

A subsample of 47 physicians (sample 2) who screened children in the 4-6 year age group also recorded all their referrals made during the two months before the educational program started and the two months before the follow-up questionnaire.

All data extraction, handling and recoding procedures were performed according to a standard operating procedure written by the first and last authors.

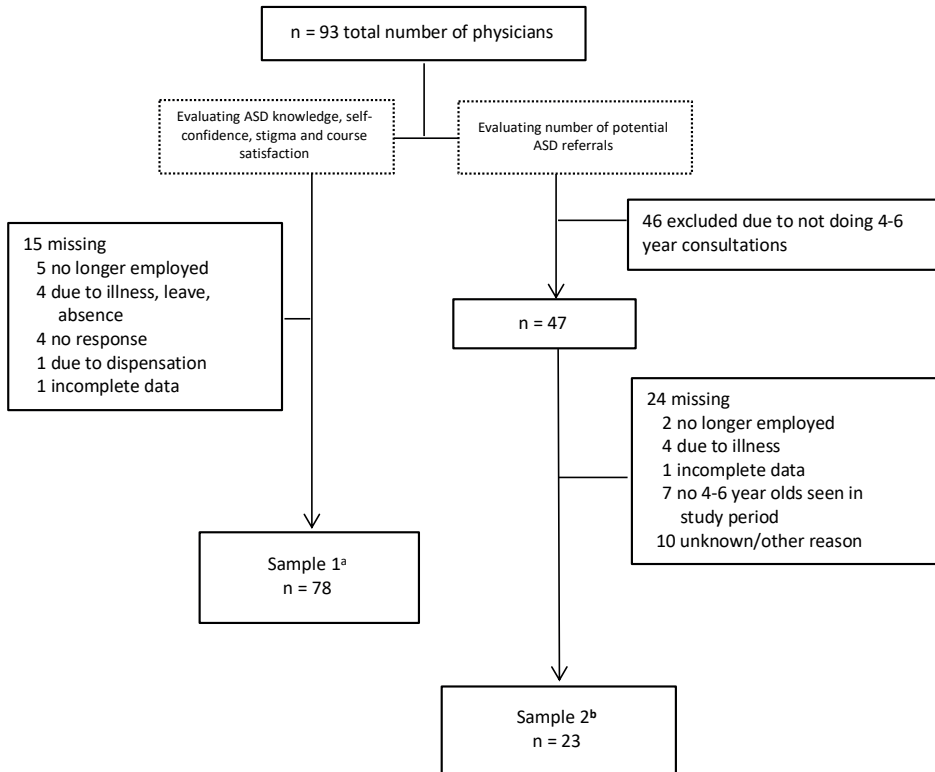
Samples

Of the 93 physicians who started the educational program (sample 1), 78 completed all pre-, post- and follow-up questionnaires (83.9%) (Figure 4.1). Of the subsample (sample 2), 23 of 47 physicians who recorded their referrals completed pre- and follow-up measurements (48.9%).

Live online educational program

The educational program *Detection of Autism Spectrum Disorders in Children Aged 4-6 Years by Youth & Family Center Physicians* entailed three weekly 1.5-hour online sessions using the Live Online Learning methodology. Live Online Learning³⁰ consists of a virtual classroom in which multiple interactive tools (e.g. video, audio, whiteboard) are available during the sessions. The educational program was guided by one of two child and adolescent psychiatrists. The sessions addressed: (1) general information related to ASD and the early detection of ASD, (2) red flags for ASD and early detection during a consultation, and (3) communication with parents and referring potential ASD cases. In addition, each session was preceded by self-preparation (reading material, videos,

preparing case studies and identifying healthcare services), that was discussed during the online sessions.



^a sample questionnaires, ^b sample of referral physicians screening 4-6 year olds.

Figure 4.1 Study populations

Measurements

ASD knowledge

We assessed ASD knowledge with the Autism Spectrum Disorder Knowledge Questionnaire – Physician edition (AKQ-P).¹⁵ The AKQ-P was specifically developed for this study and consists of 32 multiple-choice questions. The first part included 20 questions ($\alpha = 0.21$) on general ASD knowledge, prevalence, sex differences, and risk factors. The second part included 12 physician-specific questions ($\alpha = 0.31$) assessing ASD early signs, detection, diagnostic criteria, and comorbidity. We calculated a general ASD knowledge score and a specific ASD knowledge score on a 1-10 scale (1 = least knowledge, 10 = most knowledge) using weighted item scores to account for the number of possible answers for each question. We calculated the weighted sum score, to correct for guessing, for the general knowledge and physician-specific sections. The AKQ-P was evaluated and revised by an

expert panel consisting of YFC physicians, psychologists and a social worker with experience in working with children with ASD.¹⁵

Self-confidence

As no validated questionnaire was available to evaluate physicians' competence in the early detection of ASD, we assessed their self-confidence associated with the early detection of ASD with a self-reported questionnaire developed for this study. It contained five questions ($\alpha = 0.83$) scored on a 10-point Likert scale: (1) *I have enough knowledge about autism*, (2) *I have enough knowledge about early detection of autism*, (3) *I am confident that I can recognize autism in a child between 4 to 6 years old*, (4) *I am confident that I can communicate a suspicion of autism correctly with a child's parents*, (5) *I am confident that I can correctly refer a child suspected of having autism*. The total sum of the scores (range 5-50) was used for our analysis.

Stigmatizing attitudes toward mental illness

Stigmatizing attitudes toward mental illness were assessed using the Dutch translation of the Community Attitudes to Mental Illness (CAMI) questionnaire.^{31,32} The CAMI is a 40-item, self-reported questionnaire used to measure attitudes toward individuals with mental illness. The internal consistency of the four CAMI scales in our sample were: authoritarianism ($\alpha = 0.60$), benevolence ($\alpha = 0.72$), social restrictiveness ($\alpha = 0.76$) and community mental health ideology ($\alpha = 0.83$). Each of the four CAMI scales contains 10 statements scored on a 5-point Likert scale (1 = strongly agree, to 5 = strongly disagree). A value is assigned to each item and five of the 10 items for each scale are reverse coded. In order to calculate the final CAMI score, we recoded items so that a higher score (sum of all items) would correspond to having a more negative attitude toward people with mental illness. Responses to items were added together to obtain a score between 10 and 50 for each scale and they were then divided by ten (number of items in each scale). Although the CAMI was developed to evaluate public attitudes toward mental illness³¹, it has also been used broadly by health professionals^{33,34} and others.^{35,36}

Potential ASD referrals

We assessed physician referrals using five questions: (1) date of referral, (2) child's gender, (3) age in months, (4) reason for referral, and (5) type of institution the child was referred to. The physicians also reported the total number of children they saw for consultation during the measurement periods. To group the referral data, we calculated pre- and post-referral rates (referrals made/children seen for consultation in the relevant period x 100). Second, referral reasons were classified by ICD-10 category³⁷ by two of our physician co-authors. Discrepancies in classification

between the physicians were jointly re-evaluated. Third, the same two physicians coded the reason for referral into three categories assessing the likelihood of a referral being made based on possible (early) signs of ASD: *potential*, *cannot be excluded*, or *cannot say anything about it*. A referral was coded as *potential* if it matched any of the ASD DSM-5 criteria³⁸, when the reason for referral contained one or more of the following problems: behavior, attention, food intake, development of motor function, anxiety, social and/or emotional functioning. Congenital malformation and psychiatric evaluation were also included in the potential category. Reasons for referral and coded as *cannot be excluded* included: problems in motor functioning, self-esteem, upbringing, speech, dentofacial function, dysphagia, weight, posture, and sleeping, as well as life events, PTSS, encopresis, enuresis and constipation. Other reasons for referral were coded as *cannot say anything about it*. Discrepancies in coding between the physicians were jointly re-evaluated.

Educational program satisfaction

Physicians' satisfaction included a general evaluation of the educational program, evaluation of the LOL method and the usability of the information provided in clinical practice. We assessed participants' satisfaction with the educational program and the LOL method using six questions: (1) *I give the course the following grade*, (2) *The course was useful*, (3) *Live Online Learning is a pleasant way of teaching*, (4) *Contact with other participants was enjoyable*, (5) *Learning in the online school is just as pleasant as learning in a face-to-face group*, (6) *I am going to use the information from the course in my practice*. Question 1 used a 10-point Likert scale and questions 2-6 used a 5-point Likert scale.

Demographic measures

We collected demographic information about the participants in a questionnaire, including age, gender, ethnic background (own, mother and father), and years of work experience as a physician.

Statistical analyses

Non-response analysis was performed using Mann-Whitney U tests (since data were non-normally distributed) in numerical variables and Fisher's exact tests in quantitative variables. We made descriptive analyses to assess general and specific ASD knowledge, self-confidence, stigmatizing attitudes toward mental illness, potential ASD referrals, and educational program satisfaction. Additionally, we compared knowledge and self-confidence levels between the three measurement moments through repeated measures ANOVA. We used Wilcoxon Signed-Ranks tests to compare CAMI subscale scores, referral characteristics, and potential ASD referrals between the pre-measurement period and at follow-up since outcomes were not normally distributed.

RESULTS

Non-response

Non-response analyses were conducted for both samples originating from our study population of 93 YFC physicians (Figure 4.1). Analyses of responses ($n = 78$) in the questionnaire sample showed no significant differences with the non-response group ($n = 15$) on age, years on working experience, ethnicity and educational level. Analyses of the referral sample ($n = 23$) showed a higher percentage ($p < 0.05$) of specialized YFC physicians than junior doctors in the non-response group ($n = 24$).

Sample characteristics

Sample characteristics are presented in Table 4.1. Data were missing on years of work experience in both samples (sample 1 = 4.0%, sample 2 = 4.5%).

Table 4.1 General characteristics of the two study samples of primary healthcare physicians who participated in the Live Online Learning educational program: 78 physicians filled in all questionnaires (Sample 1), and 23 physicians also recorded their referrals for children between 4-6 years of age (Sample 2).

	Sample 1 (n=78)		Sample 2 (n=23)	
	Percentage or median (range) ^a	% missing	Percentage or median (range) ^a	% missing
Gender (%)		0.0		0.0
Female	96.2		91.3	
Age, years	42.5 (24.0 - 64.0)	0.0	43.0 (25.0 - 60.0)	0.0
Years of work experience, years	11.0 (0.3 - 40.0)	4.0	10.5 (1.0 - 33.0)	4.5
Ethnicity (%)		0.0		0.0
Dutch	69.2		65.2	
Western migration background	14.1		13.0	
Non-western migration background	16.7		21.7	
Education (%)		0.0		0.0
Junior doctor	48.7		52.2	
Specialized YFC physician	44.9		47.8	
Other	6.4		0.0	

^aValues are presented as percentage for categorical variables and median including range for continuous non-normal distributed variables.

ASD knowledge, self-confidence and stigma

Table 4.2 shows mean (SD) ASD knowledge grade, stigma towards mental illness and self-confidence scores at pre-, post- and follow-up measurement points. Repeated measures ANOVA showed a significant increase in general and specific ASD knowledge scores as a result of the educational program ($F(2, 154) = 8.93, p < .00$) and ($F(2, 154) = 23.92, p < .01$). The educational program had a lasting effect on physicians' specific ASD knowledge as the mean (SD) was significantly ($p = < .01$) higher at 6 months follow-up (mean = 6.46, SD= 1.65) than before the educational program (mean = 5.87, SD=1.64). We found no lasting effect on general ASD knowledge.

Table 4. 2 ASD knowledge, self-confidence, and stigmatizing attitudes held by YFC physicians at pre- and post-measurement and at 6 months follow-up after the educational program (n=78)

	Pre-measurement Mean (SD)/ Median (range)	Post-measurement Mean (SD)/ Median (range)	6 month follow-up Mean (SD)/ Median (range)
ASD knowledge ^a			
General	7.15 (1.22)	7.79 (1.04) ^e	7.34 (1.18)
Specific	5.87 (1.64)	7.13 (1.39) ^e	6.46 (1.65) ^f
Self-confidence ^b	29.05 (5.56)	37.67 (2.76) ^e	36.95 (4.08) ^f
Stigma towards mental illness ^c			
Authoritarianism	2.15 (1.40-2.80)	-	2.20 (1.40-2.80)
Benevolence	2.20 (1.30-2.90)	-	2.30 (1.40-2.80)
Social restrictiveness	2.20 (1.10-3.20)	-	2.20 (1.20-2.90)
CHMI ^d	2.20 (1.10-3.30)	-	2.20 (1.10-3.20)

^a Measured with the Autism Spectrum Disorder Knowledge Questionnaire – Physician edition (AKQ-P)

^b Measured with a questionnaire developed for this study

^c Measured with the Community Attitudes to Mental Illness (CAMI)

^d Community mental health ideology

^e Significant difference between pre- and post-measurement, $p < .01$

^f Significant difference between pre- and follow-up measurement, $p < .01$

Using repeated measures ANOVA, we found a significant increase in self-confidence regarding the detection of ASD reported by the physicians ($F(1.56, 120.27) = 141.41, p < .01$). However, Mauchley's test indicated that the assumption of sphericity had been violated ($\chi(2) = .72, p < .00$), therefore the degrees of freedom were corrected using Greenhouse-Geisser estimates of sphericity ($\epsilon = .78$). At six months follow-up, the self-confidence was still significantly higher ($p < .01$) (mean 36.95, SD = 4.08) than before the educational program (mean=29.05, SD=5.56).

Wilcoxon Signed-Ranks tests showed no significant differences ($p > 0.05$) on the CAMI scores (authoritarianism, benevolence, social restrictiveness, and community mental health ideology scales) at pre-measurement compared to 6 month follow-up.

Potential ASD referrals

Table 4.3 shows the characteristics of the 178 referrals made by the 23 complete responders. Data were missing on ICD categories (6.2%) and referral institution (1.7%). A total of 10.0% of the 1775 children seen for consultation at the YFC during the study period were referred to other professionals for further consultation (9.8% pre-measurement, 11.1% follow-up, $P = 0.7$). YFC physicians referred children primarily for mental and behavioral problems (35.4%), eye problems (24.2%) and ear problems (13.5%). Children were most often referred to ophthalmologists (23.0%), mental healthcare professionals (20.2%) and speech therapists (12.9%). Our results show no significant differences in the percentage of referrals to the different ICD-10 categories between pre-measurement and follow-up except for a 10-percentage point decrease ($p < 0.05$, from 13.2% to 3.4%) in GP referrals between pre- and follow-up time points.

Table 4.3 Characteristics of 178 referrals made by YFC physicians in study sample 2 (n=23) during the 4-6 year old consultations at pre-measurement and at 6 month follow-up

	Pre-measurement n=91	6 month follow-up n=87	Wilcoxon signed- rank test
Percentage of children referred, based on the number of children seen for consultation, % ^{a,b}	9.8	11.1	NS
Male, %	58.2	62.1	NS
Age child in months, mean (range)	69.2 (56.0;80.0)	68.9 (51.0;133.0)	NS
ICD categories, %			
ICD-IV: Endocrine, nutritional and metabolic diseases	2.2	6.9	-. ^d
ICD-V: Mental and behavioral disorders	37.4	33.3	NS
ICD-VII: Diseases of the eye and adnexa	19.8	28.7	NS
ICD-VIII: Diseases of the ear and mastoid process	15.4	11.5	NS
ICD-XI: Diseases of the digestive system	2.2	0.0	-. ^d
ICD-XII: Diseases of the skin and subcutaneous tissue	1.1	0.0	-. ^d
ICD-XIV: Diseases of the genitourinary system	1.1	0.0	-. ^d
ICD-XVII: Congenital malformations, deformations and chromosomal abnormalities	2.2	0.0	-. ^d
ICD-XVIII: Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	8.8	6.9	NS
ICD-XXI: Factors influencing health status and contact with health services	5.5	4.6	NS
Missing ^c	4.4	8.0	-. ^d
Child referred to type of specialist, %			
Ophthalmologist	19.8	26.4	NS
Mental healthcare professional	20.9	19.5	NS
Speech therapist	12.1	13.8	NS
Physical therapist	11.0	11.5	NS
Ear, nose and throat doctor	7.7	11.5	NS
GP	13.2	3.4	0.02*
Hospital	4.4	5.7	NS
Dietitian	3.3	3.4	NS
Other	6.6	2.3	NS
Missing	1.1	2.3	NS

^a Children seen for consultation by 23 physicians in study period; pre-measurement 915 and at 6 month follow-up 860 (total 1775)

^b (Number of children referred/number of children seen for consultation) * 100 = % children referred

^c Multiple issues in one referral that did not fit in one ICD-10 category

^d Not enough referrals in category to perform Wilcoxon signed-rank test

* p<0.05

Table 4.4 shows that 26.3% of the referrals before the educational program were potential ASD referrals compared to 17.2% at follow-up. Table 4.4. shows that the median potential ASD referral rate ((number of potential ASD referrals/number of total referrals) x 100) at pre-measurement was 0.0 (range = 0.0-18.2) and also at follow-up 0.0 (range = 0.0-22.2). Wilcoxon Signed-rank test

Table 4.4 Percentage of possible ASD referrals made by YFC physicians (n=23) at pre and 6 month follow-up

<i>Possible ASD</i>	Pre-measurement n=91		Follow-up n=87		Wilcoxon Signed-Rank Test	
	%	Percentage ^a Median (range)	%	Percentage ^a Median (range)	Z	p-value
Cannot say anything about it	47.2	3.4 (0.0-18.7)	52.9	2.5 (0.0-23.5)	-0.12 ^b	NS
Cannot be excluded	26.3	0.0 (0.0-9.1)	29.9	1.7 (0.0-25.0)	-1.49 ^c	NS
Potential	26.3	0.0 (0.0-18.2)	17.2	0.0 (0.0-22.2)	-1.22 ^b	NS

^a percentage of the total number of children seen in the consultation period

^b based on positive ranking

^c based on negative ranking

demonstrated no differences ($p = 0.22$) between the potential ASD referral rate at pre- and follow-up time points.

Educational program satisfaction

Participants rated the educational program on a 10-point Likert-scale as positive (mean = 7.8). On a 5-point Likert-scale, participants rated 'Live Online Learning' as a pleasant way of learning (mean = 3.8), the contact with other participants as enjoyable (mean = 3.9), the method just as pleasant as face-to-face (mean = 3.1) and indicated that they would use the information from the educational program in their clinical practice (mean = 4.4).

DISCUSSION

Our study shows the online educational program had a positive effect on the level of physicians' ASD knowledge and self-confidence, which remained high after six months' follow-up. Physician-specific ASD knowledge mean scores were higher after the educational program and remained higher at follow-up compared to pre-measurement means. Self-confidence related to the detection of ASD was higher after the educational program than at the start, and mean scores remained higher at follow-up compared to pre-measurement. However, we found no effect of the educational program on the level of stigmatizing attitudes toward mental illness and the number of potential ASD referrals.

Our results show a lasting effect on the level of overall and physician-specific (e.g. comorbidity, early signals) ASD knowledge levels after the educational program. Other studies previously described improvements in in healthcare professionals' (GPs, primary care workers, pediatricians and other medical specialists) knowledge levels after interventions concerning ASD detection and identification,

but none investigated the effects on ASD knowledge level over a longer period of time.^{18,19} The programs used in these studies were not online-based and outcome variables focused more on mean age at ASD diagnosis than on ASD knowledge in the participating healthcare professionals.^{18,19} One study investigated knowledge acquisition as an effect of a training program for pediatricians and GPs on ASD identification. However, they concentrated on knowledge of diagnostic criteria and ASD treatment instead of knowledge of early ASD detection.¹⁹ It is difficult to compare these results because research on ASD knowledge levels in YFC physicians/preventive care professionals is limited and the instruments used to measure this knowledge vary considerably. Programs that focused on improving early ASD detection, but not specifically on ASD knowledge in healthcare professionals, were effective in reducing the mean age at ASD diagnosis. However, this effect was not sustained 2-8 years after the intervention programs ended.^{10,20} Although our results indicate that a live online educational program has a six-month effect on the level of ASD knowledge, we do not know whether this effect was sustained over a longer period.

Our results show a positive effect of the educational program on YFC physicians' self-confidence regarding the detection of ASD. Earlier research confirmed our findings of improvement in physicians' perceived self-efficacy after an early ASD detection educational program.¹⁷ However, our study is -as far we know- the first one that demonstrates a longer-term effect of education on physicians' self-confidence. Previous research showed that improvements in 14 physicians' self-efficacy had a positive effect on the age at ASD diagnosis.²⁰ Self-perceptions were also named as an essential feature for young doctors taking clinical action based on knowledge.³⁹ Our educational program is successful in increasing YFC physicians' self-confidence regarding the detection of ASD, which we consider to positively affect the early detection of ASD.

Contrary to our expectations we did not find the educational program had any effect on YFC physicians' level of stigma towards mental illness. Our results are comparable to those of a study on the effect of an e-learning course on psychiatric nurses' attitudes towards mental illness.²⁹ Attitudes toward mental illness are complex and have been proven difficult to change through education.⁴⁰ Although educational programs might not have a direct effect on healthcare professionals' attitudes, some indirect effects might be achieved through an increase in knowledge and confidence in treating people with mental illness.⁴¹ However, previous research indicated that especially anti-stigma intervention that included social contact with someone who has had experience of living with mental illness was effective for a broad range of healthcare providers⁴² and medical students.⁴³ Another possible explanation for not finding an effect on the level of stigma is the positive attitudes already held toward mental illness by Dutch YFC physicians in general.¹⁵ Their low level of stigma might be difficult to reduce even further, especially through the indirect effect of an ASD early detection educational program. As healthcare personnel are an important source of stigma that can have a

negative influence on patients' access to mental healthcare,²⁷ further research is needed to find ways to reduce the level of stigmatizing attitudes toward mental illness in preventive care providers. We also found no effect from the educational program on the number of potential ASD referrals. It is possible that an increase in ASD knowledge does not directly translate into an increased number of ASD referrals. This was also reported by Schonwald et al.²⁴, who found that implementing developmental screening in pediatric practice (children 2-3 years old) increased the identification rate for developmental and behavioral problems, but they did not find a significant increase in overall referral rates. Schonwald et al.²⁴ indicated that providers possibly continued to exercise clinical judgment when deciding whether to refer a child or manage it within the office setting. In our educational program, physicians were taught that when they had doubts regarding the early signs of ASD, a follow-up appointment to further explore ASD traits was preferred to a referral. In addition, the limited sensitivity of our ASD coding mechanism could have affected our results as, based on a prevalence of ASD in 1.5% of children under 8 years old,⁴⁴ only 26/1775 of our screened children (pre-measurement = 915, post-measurement = 860) may have ASD. Also, as 1.7-2.6% of the children seen for consultation were coded as a possible ASD referral, this is a higher rate than the ASD prevalence of 1.5% in the general population. This prevalence indicates that non-ASD children might have been included in the potential ASD category due to our coding scheme. Both scenarios would increase the number of false-positives, complicating detecting any change in ASD referrals after the educational program.¹⁰ Furthermore, we did not follow-up on the referrals, so no conclusions can be drawn on how correct the referrals were. Thus, it is possible that the educational program increased the number of ASD referrals, but simultaneously decreased the false-positive referrals, keeping the number of potential ASD referrals equal in pre-measurement and follow-up periods.

Strengths and limitations

Our study has several strengths. First, the follow-up measurement at six months after the educational program created the opportunity to measure the sustainability of the educational program and strengthened the results. The high response rate (97.85% after the educational program, 83.87% at follow-up) of a large number of YFC physicians supports the generalizability of our findings. Finally, the large number of children screened by the physicians strengthened the results.

There were also several limitations. First, we investigated general stigmatizing attitudes toward mental illness instead of stigma related specifically to ASD. The generalizability of the questions used in the CAMI (e.g. questions regarding danger to others, mental healthcare facilities in the community) do not accurately reflect stigma toward young children with ASD. Thus, our observed absence of a relation between the educational program and stigmatizing attitudes toward mental

illness might have been different with an ASD-specific stigma questionnaire. In further research, the Autism Stigma and Knowledge Questionnaire (ASK-Q)^{45,46} would be preferable, as they evaluate both ASD knowledge and stigma and has strong psychometric properties. Secondly, we had a limited amount of information regarding the ASD referrals made by the YFC physicians. More extensive information about: (1) the reason for referral, (2) insight into the number of children who were not referred but recommended to come back for a follow-up appointment, and (3) a follow-up on the potential ASD referrals to confirm their validity would have strengthened our results. Also, the six months follow-up might be a too short timeframe to see an effect of the educational program on the number of potential ASD referrals. However, the increased ASD knowledge and self-confidence of YFC physicians suggests improvement in identification is possible. Also, we had no information regarding the number of children that were detected or diagnosed with ASD before the 4-6 year YFC visit. A high ASD detection rate during the previous YFC visits could have decreased the ASD referrals during 4-6 years consultation. Also, we had no data on the number of children that were previously detected by other healthcare professionals (e.g. GP's and pediatricians). It is however likely that, based on the current mean age at ASD diagnosis in The Netherlands (50-234 months)⁵⁻⁷ the number of children that is detected before the 4-6 year visit is relatively small. In addition, we hypothesize that young children (< age 4 years) with severe ASD will indeed be detected previously. However, the focus of the study is to identify the larger group of children with moderate ASD traits who can enter the regular schools. Consequently, the educational program might have led to a higher ASD referral rate than found in our study. A final limitation was the low response rate (49%) on the pre- and post-referral questionnaires with a higher non-response by specialized YFC physicians. The higher non-response by specialized physicians could have negatively affected our referral results as they may be more skilled in the detection of ASD due to additional education.

Future research should address the full, early ASD detection-referral-diagnostic process in a prospective general population sample. This study should include knowledge regarding the final diagnosis and thereby evaluate: 1) whether physicians are able to identify more children with ASD (reduce false negatives), 2) whether physicians are able to differentiate better between ASD and other disorders (reduce false positives), and/or 3) whether an ASD educational program might lead to over referral (increase false positives). The ASD detection-referral-diagnostic process does not only involve YFC/preventive care physicians, but numerous other professionals and the process often contains multiple referrals from a variety of mental health and non-mental health professionals. So, only a general population cohort followed over a long period of time could fully evaluate the effectiveness of the ASD detection-referral-diagnostic process and indicate further ways to improve it. Such a study design would not only cover the full process, but could also connect with population and clinical data to adjust for confounding factors.

Conclusion

Our study shows that an online educational program had at least a six-month long effect on ASD knowledge and self-confidence regarding ASD referrals by Youth and Family Center physicians. We found no effect from the educational program on stigmatizing attitudes toward mental illness or on the number of potential ASD referrals. Education on the early signs of ASD, communication with parents, and municipal referral options should be given adequate attention in preventive care settings.

ACKNOWLEDGEMENTS

This work was supported by a grant from the Netherlands Organization for Health Research and Development (NWO-ZonMw) [REACH-AUT No. 70-73400-98-002]. We would like to thank the physicians and staff of the Youth and Family Center Rotterdam-Rijnmond for their participation in this study. We further acknowledge help from Ms. Rosalien V. Pasma, MSc, Ms. Anoeska Vellekoop, MSc, Ms. Inge van der Zwaan, MSc, Ms. Monique C. Neuker, MSc, Ms. Romy Hoogveen, MSc in the development, execution and/or coordinating the study and performed measurements. Also special thanks to Dr. Marc Reusens for participating as one of the program teachers and to Ms. Jacqueline T. Bailly, MSc (Manager, Sarr Expert Centre for Autism) for acquiring funding for the study.

DISCLOSURE OF POTENTIAL CONFLICTS OF INTEREST

Funding: This work was supported by a grant from the Netherlands Organization for Health Research and Development (NWO-ZonMw) [REACH-AUT No. 70-73400-98-002].

Conflict of Interest: The authors declare that they have no conflict of interest.

AUTHOR'S CONTRIBUTIONS

MH conceived the study, participated in its design and coordination, performed the statistical analyses, and drafted the manuscript. AN performed statistical analyses, participated in the interpretation of the data, and helped draft the manuscript. IB participated in the interpretation of the data, and helped draft the manuscript. MD performed statistical analyses, participated in the interpretation of the data, and helped draft the manuscript. HH conceived the study, participated in its design and coordination, interpretation of the data, and helped draft the manuscript. WE conceived the study, participated in its design and coordination, and in interpretation of the data, and helped draft the manuscript. All authors critically revised the manuscript and approved the final version.

REFERENCES

1. Maenner MJ, Shaw KA, Baio J, et al. Prevalence of autism spectrum disorder among children aged 8 Years-Autism and developmental disabilities monitoring network, 11 Sites, United States, 2016. *MMWR Surveill Summ.* 2020;69(4):1-12. doi:10.15585/MMWR.SS6904A1
2. Hossain MM, Khan N, Sultana A, et al. Prevalence of comorbid psychiatric disorders among people with autism spectrum disorder: An umbrella review of systematic reviews and meta-analyses. *Psychiatry Res.* 2020;112922. doi:10.1016/j.psychres.2020.112922
3. Graham Holmes L, Zampella CJ, Clements C, et al. A Lifespan Approach to Patient-Reported Outcomes and Quality of Life for People on the Autism Spectrum. *Autism Res.* 2020;13(6):970-987. doi:10.1002/aur.2275
4. van 't Hof M, Tisseur C, van Berckelaer-Onnes I, et al. Age at autism spectrum disorder diagnosis: a systematic review and meta-analysis from 2012 to 2019. *Autism.* doi:10.1177/1362361320971107
5. Begeer S, Mandell D, Wijnker-Holmes B, et al. Sex differences in the timing of identification among children and adults with autism spectrum disorders. *J Autism Dev Disord.* 2013;43(5):1151-1156. doi:10.1007/s10803-012-1656-z
6. Kentrou V, de Veld DMJ, Mataw KJK, Begeer S. Delayed autism spectrum disorder recognition in children and adolescents previously diagnosed with attention-deficit/hyperactivity disorder. *Autism.* 2018. doi:10.1177/1362361318785171
7. Salomone E, Charman T, McConachie H, Warreyn P. Child's verbal ability and gender are associated with age at diagnosis in a sample of young children with ASD in Europe. *Child Care Health Dev.* 2016;42(1):141-145. doi:10.1111/cch.12261
8. Van Tongerloo MAMM, Bor HHJ, Lagro-Janssen ALM. Detecting autism spectrum disorders in the general practitioner's practice. *J Autism Dev Disord.* 2012;42:1531-1538. doi:10.1007/s10803-011-1384-9
9. Zwaigenbaum L, Bauman ML, Fein D, et al. Early screening of autism spectrum disorder: Recommendations for practice and research. *Pediatrics.* 2015;136(Supplement 1):S41-S59. doi:10.1542/peds.2014-3667D
10. Pijl MKJ, Buitelaar JK, Korte MWP de, Rommelse NNJ, Oosterling IJ. Sustainability of an early detection program for autism spectrum disorder over the course of 8 years. *Autism.* 2017;22(8):1018-1024. doi:10.1177/1362361317717977
11. van Berckelaer-Onnes IA, van de Glind G, Anzion P, Werkgroep Richtlijnen ASS-JGZ. *JGZ-Richtlijn Autismespectrumstoornissen: Signalering, Begeleiding En Toeleiding Naar Diagnostiek.* Utrecht: Nederlands Centrum Jeugdgezondheid; 2015. <https://assets.ncj.nl/docs/bc756f24-ff04-45f3-b0fd-3756dec2cabb.pdf>.
12. Centrum voor Jeugd en Gezin Rijnmond. *Alles Voor Een Gezonde Jeugd: Productenboek.* Rotterdam; 2016. <https://cjrjijnmond.nl/wp-content/uploads/Productenboek-CJGRijnmond-2016.pdf>.
13. Dosreis S, Weiner CL, Johnson L, Newschaffer CJ. Autism spectrum disorder screening and management practices among general pediatric providers. *J Dev Behav Pediatr.* 2006;27(Supplement 2):S88-S94. doi:10.1097/00004703-200604002-00006
14. Heidgerken AD, Geffken G, Modi A, Frakey L. A survey of autism knowledge in a health care setting. *J Autism Dev Disord.* 2005;35(3):323-330. doi:10.1007/s10803-005-3298-x
15. van 't Hof M, van Berckelaer-Onnes I, Deen M, et al. Novel insights into autism knowledge and stigmatizing attitudes toward mental illness in dutch youth and family center physicians. *Community Ment Health J.* February 2020. doi:10.1007/s10597-020-00568-w
16. O'Brien D, Harvey K, Howse J, Reardon T, Creswell C. Barriers to managing child and adolescent mental health problems: A systematic review of primary care practitioners' perceptions. *Br J Gen Pract.* 2016;66(651):e693-e707. doi:10.3399/bjgp16X687061
17. Carbone PS, Norlin C, Young PC. Improving early identification and ongoing care of children

- with autism spectrum disorder. *Pediatrics*. 2016;137(6). doi:10.1542/peds.2015-1850
18. Oosterling IJ, Wensing M, Swinkels SH, et al. Advancing early detection of autism spectrum disorder by applying an integrated two-stage screening approach. *J Child Psychol Psychiatry Allied Discip*. 2010;51(3):250-258. doi:10.1111/j.1469-7610.2009.02150.x
 19. Bordini D, Lowenthal R, Gadelha A, de Araujo Filho GM, Mari J de J, Paula CS. Impact of training in autism for primary care providers: a pilot study. *Rev Bras Psiquiatr*. 2015;37(1):63-66. doi:10.1590/1516-4446-2014-1367
 20. Holzer L, Mihailescu R, Rodrigues-Degaeff C, et al. Community introduction of practice parameters for autistic spectrum disorders: Advancing early recognition. *J Autism Dev Disord*. 2006;36(2):249-262. doi:10.1007/s10803-005-0053-2
 21. Golnik A, Ireland M, Borowsky IW. Medical homes for children with autism: A physician survey. *Pediatrics*. 2009;123(3):966-971. doi:10.1542/peds.2008-1321
 22. Unigwe S, Buckley C, Crane L, Kenny L, Remington A, Pellicano E. GPs' confidence in caring for their patients on the autism spectrum: An online self-report study. *Br J Gen Pract*. 2017;67(659):e445-e452. doi:10.3399/bjgp17X690449
 23. Hix-Small H, Marks K, Squires J, Nickel R. Impact of implementing developmental screening at 12 and 24 months in a pediatric practice. *Pediatrics*. 2007;120(2):381-389. doi:10.1542/peds.2006-3583
 24. Schonwald A, Huntington N, Chan E, Risko W, Bridgemohan C. Routine developmental screening implemented in urban primary care settings: More evidence of feasibility and effectiveness. *Pediatrics*. 2009;123(2):660-668. doi:10.1542/peds.2007-2798
 25. Earls MF, Andrews JE, Hay SS. A longitudinal study of developmental and behavioral screening and referral in north carolina's assuring better child health and development participating practices. *Clin Pediatr (Phila)*. 2009;48(8):824-833. doi:10.1177/0009922809335322
 26. Knaak S, Mantler E, Szeto A. Mental illness-related stigma in healthcare: Barriers to access and care and evidence-based solutions. *Healthc Manag Forum*. 2017;30(2):111-116. doi:10.1177/0840470416679413
 27. Thornicroft G, Mehta N, Clement S, et al. Evidence for effective interventions to reduce mental-health-related stigma and discrimination. *Lancet*. 2016;387(10023):1123-1132. doi:10.1016/S0140-6736(15)00298-6
 28. Beaulieu T, Patten S, Knaak S, Weinerman R, Campbell H, Lauria-Horner B. Impact of skill-based approaches in reducing stigma in primary care physicians: Results from a double-blind, parallel-cluster, randomized controlled trial. *Can J Psychiatry*. 2017;62(5):327-335. doi:10.1177/0706743716686919
 29. Pitkänen A, Kontio R, Lahti M, Katajisto J, Välimäki M. The impact of an elearning course on nurses' attitudes towards mental illness. *Open J Nurs*. 2015;5(11):1004-1011. doi:10.4236/ojn.2015.511107
 30. Learning Connected. <https://learningconnected.com/>. Published 2019. Accessed April 30, 2020.
 31. Taylor SM, Dear MJ. Scaling community attitudes toward the mentally ill. *Schizophr Bull*. 1981;7(2):225-240. doi:10.1093/schbul/7.2.225
 32. Van Gampelaere V. *Wat Denkt Politie over Personen Met Een Psychiatrische Stoornis?* Antwerpen: Maklu; 2013.
 33. Chambers M, Guise V, Välimäki M, et al. Nurses' attitudes to mental illness: A comparison of a sample of nurses from five European countries. *Int J Nurs Stud*. 2010;47(3):350-362. doi:10.1016/j.ijnurstu.2009.08.008
 34. Smith AL, Cashwell CS. Stigma and mental illness: Investigating attitudes of mental health and non-mental-health professionals and trainees. *J Humanist Couns Educ Dev*. 2010;49(2):189-202. doi:10.1002/j.2161-1939.2010.tb00097.x
 35. Losinski M, Maag JW, Katsiyannis A. Characteristics and attitudes of pre-service teachers toward individuals with mental illness. *J Educ Pract*. 2015;6(3):11-17.
 36. Bell S, Palmer-Conn S. Suspicious minds: Police attitudes to mental ill health. *Int J Law Public*

- Adm.* 2018;1(2):25-40. doi:10.11114/ijlpa.v1i2.3878
37. World Health Organization. *The ICD-10 Classification of Mental and Behavioural Disorders: Clinical Descriptions and Diagnostic Guidelines*. Geneva: World Health Organization; 1992.
 38. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5)*. Arlington, VA: American Psychiatric Association; 2013. doi:10.1176/appi.books.9780890425596
 39. Kennedy T, Regehr G, Rosenfield J, Roberts SW, Lingard L. Exploring the gap between knowledge and behavior: A qualitative study of clinician action following an educational intervention. *Acad Med.* 2004;79(5):386-393. doi:10.1097/00001888-200405000-00006
 40. Boysen GA, Vogel DL. Education and mental health stigma: The effects of attribution, biased assimilation, and attitude polarization. *J Soc Clin Psychol.* 2008;27(5):447-470. doi:10.1521/jscp.2008.27.5.447
 41. Henderson C, Noblett J, Parke H, et al. Mental health-related stigma in health care and mental health-care settings. *The Lancet Psychiatry.* 2014;1(6):467-482. doi:10.1016/S2215-0366(14)00023-6
 42. Knaak S, Modgill G, Patten SB. Key ingredients of anti-stigma programs for health care providers: a data synthesis of evaluative studies. *Can J Psychiatry.* 2014;59(1_suppl):19-26. doi:10.1177/070674371405901s06
 43. Papish A, Kassam A, Modgill G, Vaz G, Zanussi L, Patten S. Reducing the stigma of mental illness in undergraduate medical education: A randomized controlled trial. *BMC Med Educ.* 2013;13(1):7-9. doi:10.1186/1472-6920-13-141
 44. Christensen DL, Baio J, Braun KVN, et al. Prevalence and characteristics of autism spectrum disorder among children aged 8 years - Autism and developmental disabilities monitoring network, 11 sites, United States, 2012. *MMWR Surveill Summ.* 2016;65(3):1-23. doi:10.15585/mmwr.ss6503a1
 45. Harrison AJ, Bradshaw LP, Naqvi NC, Paff ML, Campbell JM. Development and psychometric evaluation of the autism stigma and knowledge questionnaire (ASK-Q). *J Autism Dev Disord.* 2017;47(10):3281-3295. doi:10.1007/s10803-017-3242-x
 46. Harrison AJ, Paff ML, Kaff MS. Examining the psychometric properties of the autism stigma and knowledge questionnaire (ASK-Q) in multiple contexts. *Res Autism Spectr Disord.* 2019;57:28-34. doi:10.1016/j.rasd.2018.10.002

