

REGULAR ARTICLE

Children adopted from Poland display a high risk of foetal alcohol spectrum disorders and some may go undiagnosed

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ABSTRACT

Aim: Children adopted from Central and Eastern Europe have often had negative early experiences, including prenatal exposure to alcohol. We examined a group of Polish children, adopted by Dutch parents, to see how many were diagnosed with foetal alcohol spectrum disorders (FASD) and to what extent features of FASD were present.

Methods: The 121 children, aged between 6 and 17 years, were adopted from Poland at a mean age of 3 years (standard deviation 1.6 years). Their parents answered a questionnaire regarding FASD diagnosis, growth, educational attainment and the Behaviour Rating Inventory of Executive Function.

Results: Three groups were identified: children with an FASD diagnosis (31%), children whose adoptive parents suspected FASD (21%) and children whose adoptive parents did not suspect FASD (49%). Growth deficiency, enrolment in special education and difficulties with executive functioning were most frequently observed in children diagnosed with FASD. However, features of FASD were also observed in the other two groups.

Conclusion: Children adopted from Poland showed a high risk of FASD and some children may go undiagnosed. Adoptive parents and professionals need to be aware of the potential consequences of prenatal exposure to alcohol.

INTRODUCTION

Research about adopted children has provided us with ample knowledge on the effects of early life risk factors, such as deprivation in institutional care (1,2). A risk factor that has received relatively little attention is prenatal alcohol exposure, which may affect physical development, neurological development and behaviour (3). This study examined prenatal alcohol exposure, and its potential consequences, in Polish children adopted by Dutch parents.

Foetal alcohol spectrum disorders

The range of potential consequences of prenatal exposure to alcohol fall under the nondiagnostic umbrella term foetal alcohol spectrum disorders (FASD), with foetal alcohol syndrome (FAS) at the most severe end of the spectrum. The main features of FAS are prenatal and, or, postnatal growth deficiency (height, weight), a set of facial anomalies (short palpebral fissures, smooth philtrum, thin upper lip) and central nervous system dysfunction (4,5). Prevalence rates in the USA are estimated to be 0.5–2 per 1000 live

Abbreviations

BRIEF, Behaviour Rating Inventory of Executive Function; CEE, Central and Eastern Europe; FAS, Foetal alcohol syndrome; FASD, Foetal alcohol spectrum disorders.

births for FAS and 10 per 1000 live births for FASD (6). Two European studies estimated FAS at 3.7–7.4 per 1000 children and FASD at 20.3–40.8 per 1000 children (7,8).

Individuals with FASD face a range of possible neuropsychological and behavioural problems, including impairments in intellectual functioning, learning, memory, language, visual-spatial ability, attention and adaptive behaviour (3,9). In comparison to a normal control group, individuals with FASD showed more deficits in executive functioning (10): a collection of related abilities that provide intentional, goal-directed, problem-solving action, such as the ability to initiate and stop actions and plan

Key notes

- This study found that the prevalence of foetal alcohol spectrum disorders (FASD) was high in Polish children, adopted by Dutch parents.
- Polish adoptees diagnosed with FASD were more likely to display growth deficiency, be enrolled in special education and have more problems with executive functioning.
- There were strong indications that some children with FASD remain undiagnosed and screening of high-risk groups is needed so that they can be identified early.

future behaviour (11). Deficits in executive functioning in children with FASD occurred to the same degree as for children with or without facial features of FAS (10).

FASD in adopted children

An increased risk of prenatal exposure to alcohol and FASD has been reported in children adopted from Central and Eastern European (CEE) countries. Landgren et al. (12) reported that the prevalence of FASD was 52% among children adopted in Sweden from CEE countries, including Russia, Poland, Romania, Estonia and Latvia. Based on phenotypic characteristics, 58% of the children adopted from Eastern Europe and the former Soviet Union were said to have a high/intermediate risk for prenatal alcohol exposure (13). Information about prenatal exposure to alcohol is unavailable for many adoptees (12,14). Maternal alcoholism has been specifically mentioned in the adoption charts of 33% adoptees from CEE countries (15). In another sample of children adopted from CEE countries, 21% were prenatally exposed to alcohol and exposure in the remaining children was unknown (14).

Current study

Despite the growing number of studies on prenatal alcohol exposure and FASD, few have focussed on adoptees with FASD. Between 2003 and 2012, 3462 Polish children were placed for international adoption (16–18). These children predominantly came from dysfunctional families, and parental alcohol abuse was a common risk factor (19).

Considering the potential risk for FASD in this group of adoptees, we examined what percentage of children adopted from Poland were diagnosed with FASD after adoption and what percentage of the adoptive parents suspected their child had FASD. Secondly, we studied to what extent FASD features were present in children adopted from Poland and whether an FASD diagnosis or suspicion of FASD by the adoptive parents was associated with growth deficiency, special education attendance and executive functioning difficulties in the children.

METHODS

Participants

In a previous study, the Dutch adoptive parents of all children adopted from Poland between January 1999 and December 2006 (n = 181) were invited to participate in a study on the development of Polish adoptees after adoption. The response rate was 73% (19). For this study, conducted 2 years later, we approached the parents who participated in the first study and the adoptive parents of 121 children (91%) agreed to participate again.

The mean age of the 121 children (52% boys) was 10.9 years, with a standard deviation (SD) of 2.7 years and a range of 5.5–17.2. The mean age at adoption was 3.0 years (SD = 1.6, range 0.8–6.9). In most cases, the children were placed for adoption because the parental rights of the biological parents had been terminated by a judge. Some of the children were abandoned or their biological parents

voluntarily gave them up for adoption. The children were adopted by 71 Dutch couples, who had an average of 2.1 children (SD = 0.8, range 1–4). The average age at assessment of the adoptive mothers was 46.0 years (SD = 5.4, range 34.7-57.6), and for the adoptive fathers, it was 47.5 years (SD = 4.6, range 40.3-58.1). The questionnaires were completed by adoptive mothers (73%), adoptive fathers (8%) or both (19%).

Measures

Demographics and pre-adoptive risk factors

A questionnaire was developed for this study to assess the demographic variables of the adoptive parents and their children and the background variables of the children prior to adoption. The adoptive parents were asked whether prenatal alcohol exposure, history of neglect and history of abuse had occurred or not or whether this was unknown. Adoptive parents mainly based their knowledge about the pre-adoptive risk factors on information from the adoption file and information obtained during the adoption process, which involved spending approximately 6 weeks in Poland. The validity of the information on pre-adoptive risk factors could, therefore, not be confirmed.

FASD diagnosis and suspicion

We used a questionnaire to ask adoptive parents whether their child had been diagnosed with FASD. If the parents confirmed this, they were asked to report when and by whom their child had been diagnosed. Adoptive parents, whose child did not have an FASD diagnosis, were asked whether they suspected the presence of FAS or a related condition within the FASD spectrum.

Growth

The child's current height and weight were reported by the adoptive parents. The percentile scores were calculated based on a reference group of Polish children, aged 6–19 years (20). Height and weight scores at or below the 10th percentile were used to indicate mild to severe growth deficiency. An overall category for growth deficiency was also computed. Children who scored at or below the 10th percentile of height and, or, weight, were categorised as having mild to severe growth deficiency (21).

Educational attainment

Adoptive parents indicated the type of education the children were enrolled in, distinguishing between schools for regular and special education. This last group included children with learning difficulties, behavioural problems, physical or sensory disabilities or protracted illness.

Executive functioning

To measure executive functioning, the Dutch version (22) of the Behaviour Rating Inventory of Executive Function (BRIEF) was used (23). This questionnaire consists of 75 items constituting eight clinical scales: inhibit, shift, emotional control, initiate, working memory, plan/organise, organisation of materials, and monitor. Adoptive parents were asked to rate if their child displayed certain behaviour never, sometimes or often. A total score, the global executive composite, was computed by summing the clinical scales. The raw BRIEF scores were transformed into T scores (mean = 50; 1 SD = 10) and T scores above 65 (>1.5 SD) were considered to be clinically significant (22). Huizinga and Smidts (24) reported good internal consistency of the BRIEF, with Cronbach's alphas from 0.78 to 0.96. In this study, the reliability of the eight clinical scales was good (α = 0.86–0.93). Cronbach's alpha for the global executive composite was 0.97.

Data analysis

Three groups were identified: the FASD group, consisting of children diagnosed with a disorder in the FASD spectrum; the suspected FASD group, with children whose adoptive parents suspected FASD; and the no FASD group, with children whose adoptive parents did not suspect FASD. Differences between these three subgroups on growth, educational attainment and executive functioning were analysed with chi-square tests of independence. Post hoc analyses were performed for significant differences. Bonferroni correction was applied to correct for multiple comparisons (adjusted $\alpha=0.017$). For effect size, we used Cramér's V (ϕ c) with the following interpretation: <0.19 very low; 0.20–0.39 low; 0.40–0.69 modest; 0.70–0.89 high; 0.90–1.00 very high (25).

A one-way between-groups multivariate analysis of variance (MANOVA) was used to compare average T scores of the three subgroups on the eight clinical subscales of the BRIEF. Post hoc univariate analyses (ANOVA) were carried out with Bonferroni adjusted alpha level ($\alpha=0.017$). Missing BRIEF data were imputed from the average of the relevant subscale. Two cases were deleted, because of inconsistent scores on the BRIEF. Eta squared η^2 was used as an effect size measure, with 0.01–0.05 indicating a small, 0.06–0.13 a medium and >0.14 a large effect size, respectively (26).

RESULTS

FASD: diagnosis and suspicion

The study included 37 children (31%) who were diagnosed with a disorder in the FASD spectrum (FASD group). There were 25 children (21%), who had not been professionally diagnosed with FASD, but their parents suspected they had FASD (suspected group). Finally, there were 59 children (49%) who had not been diagnosed with FASD and their parents did not suspect they had FASD (no FASD group).

Children in the FASD group were diagnosed with FAS (n=26), partial FAS (n=5), alcohol-related birth defects (n=1) and alcohol-related neurodevelopmental disorder (n=1). The adoptive parents of four children did not specify the diagnosis. Four of the children in the FASD group (11%) had already been diagnosed in Poland and the other children were diagnosed in the Netherlands, on average 4.8 years after adoption $(SD=2.7, range\ 0.0-9.9)$. The average age at diagnosis was 8.2 years $(SD=3.1, range\ 1.0-14.2)$. Most children had received their diagnosis in one of the Dutch FAS outpatient clinics, by a paediatrician and psychologist. Other diagnoses were given by paediatricians (n=5), clinical geneticists (n=4) and a psychiatrist (n=1).

Demographic characteristics and pre-adoptive risk factors of the three subgroups are displayed in Table 1. The three groups differed significantly by age at adoption, the proportion of children prenatally exposed to alcohol and the proportion of children with a history of neglect. Post hoc analyses showed that, in comparison to the children with no suspected FASD, the FASD group and the suspected FASD group were more frequently exposed to alcohol during gestation ($\chi^2 = 21.66$, p < 0.001, $\phi c = 0.56$; $\chi^2 = 11.07$, p = 0.001, $\phi c = 0.44$). They were more frequently neglected as well ($\chi^2 = 8.53$, p = 0.003, $\phi c = 0.32$; $\chi^2 = 7.17$, p = 0.007, $\phi c = 0.32$). The FASD group and the suspected FASD group did not differ significantly with regard to either variable. For age at adoption, post hoc comparisons between the three subgroups were not significant.

	FASD		Suspected FASD		No FASD		Chi-square tests		ANOVA			
	n	%/Mean (SD)	n	%/Mean (SD)	n	%/Mean (SD)	χ²	р	фс	F	р	η^2
Demographic characteristics												
Sex of the child (boys)	37	54%	25	44%	59	54%	0.82	0.663	0.08			
Age at assessment (years)	37	11.0 (2.9)	25	11.4 (2.4)	59	10.5 (2.7)				0.96	0.384	0.02
Age at adoption (years)	37	3.2 (1.7)	25	3.5 (1.6)	59	2.6 (1.4)				3.60	0.030*	0.06
Time with adoptive family (years)	37	7.7 (2.2)	25	7.9 (2.2)	59	7.9 (2.5)				0.07	0.936	0.00
Low birth weight (<2500 g) [†]	35	51%	24	46%	54	35%	2.43	0.296	0.15			
Pre-adoptive risk factors†												
Prenatal exposure to alcohol	30	100%	18	94%	39	49%	28.63	<0.001**	0.57			
History of neglect	33	91%	19	95%	50	62%	13.51	0.001**	0.36			
History of abuse	10	40%	12	33%	32	34%	0.13	0.938	0.05			
Time in institutional care (months)	26	21.7 (10.5)	18	24.3 (9.4)	46	23.5 (10.9)				0.40	0.673	0.01

Features of FASD

Growth

The percentages of Polish adoptees with growth deficiency are displayed in Table 2. The proportion of children scoring below or at the 10th percentile for height and weight were both significantly different for the three groups ($\chi^2 = 10.96$, p = 0.004, ϕ c = 0.33; $\chi^2 = 11.50$, p = 0.003, ϕ c = 0.35). Post hoc analyses showed that, in comparison to children in the no FASD group, significantly more children in the FASD group scored below the 10th percentile on height ($\chi^2 = 10.99$, p = 0.001, ϕ c = 0.38). For weight, children in the FASD group scored more frequently below the 10th percentile than children in both the suspected group and the no FASD group ($\chi^2 = 8.06$, p = 0.005, ϕ c = 0.40; $\chi^2 = 7.69$, p = 0.006, ϕ c = 0.32). The remaining differences between groups were not significant for either height or weight.

Educational attainment

Of the children with an FASD diagnosis, 49% were attending special education. Of the children whose parents suspected FASD, 56% attended special education. For children with no suspicion of FASD, the proportion of children enrolled in special education was 22%. The difference between the three groups was significant ($\chi^2=11.67$, p = 0.003, ϕ c = 0.31). Post hoc analyses indicated that, in comparison to children in the no FASD group, the proportion of children in special education was higher for children in the FASD group ($\chi^2=7.37$, p = 0.007, ϕ c = 0.28), and the suspected FASD group ($\chi^2=9.29$, p = 0.002, ϕ c = 0.33). The difference between the FASD group and the suspected group was not significant.

Executive functioning

The percentage of children scoring in the clinical range of the global executive composite in the FASD group, the suspected group and the no FASD group were 46%, 24%, and 12%, respectively. These proportions were significantly different for the three groups ($\chi^2 = 14.18$, p = 0.001, $\phi c = 0.34$). Post hoc analyses showed a significant difference between the FASD group and the no FASD group ($\chi^2 = 14.09$, p < 0.001, $\phi c = 0.38$). The suspected group did not differ significantly from either of the two other groups.

Results for the clinical scales of the BRIEF are displayed in Table 3. Multivariate analysis of variance indicated a significant difference between the scores of children on the eight combined clinical scales of the BRIEF (F=2.24, p=0.005, Wilks' Lambda = 0.74; $\eta^2=0.26$). The univariate analyses of the individual scales yielded a significant difference between the three groups for the clinical scales inhibit, emotional control, and organisation of materials. Post hoc comparisons showed that the mean scores on these scales were significantly higher for children in the FASD group than for children in the no FASD group. Mean scores of the suspected group were between the scores of the other two groups, but differences were not significant for any of the three scales.

DISCUSSION

Children adopted from Poland were found to have a high risk of FASD. After an average of 8 years with their adoptive parents, 31% of the children had been diagnosed with FASD at some point. Yet a further 21% of the adoptive

Table 2 Growth deficiency for Pol	ish adoptees with FASD, susp	ected FASD and no FASD				
	FASD (n = 30–31), %	Suspected FASD (n $=$ 21), %	No FASD (n = 45–47), $\%$	χ^2	р	фс
Height (≤10th percentile)	58	38	21	10.96	0.004	0.33
Weight (≤10th percentile)	53	14	22	11.50	0.003	0.35
Growth deficiency*	63	38	31	7.87	0.020	0.29

A group of Polish school-aged children and adolescents was used as a reference group (20). N varied from the total sample, because eight children fell below the age range of the reference group and were excluded. In other cases, adoptive parents failed to report height and/or weight.

^{*}Height and/or weight at or below the 10th percentile, indicating mild to severe growth deficiency (21).

	FASD (n = 37)		Suspected FASD $(n = 25)$		No FASD (n = 5	9)	ANOVA		
BRIEF scale	Mean (SD)	% Clinical	Mean (SD)	% Clinical	Mean (SD)	% Clinical	F	р	η^2
Inhibit	65.4 (14.1)	62	62.6 (13.8)	36	55.6 (10.8)	19	7.59	0.001*	0.1
Shift	61.8 (13.0)	41	56.7 (12.9)	20	53.8 (12.7)	20	4.43	0.014	0.0
Emotional control	61.0 (10.2)	35	57.5 (13.1)	32	51.4 (10.8)	9	8.79	<0.001*	0.13
Initiate	56.0 (14.1)	32	53.4 (13.9)	20	49.0 (12.1)	9	3.39	0.037	0.0
Working memory	59.0 (12.4)	43	58.5 (10.5)	28	52.3 (11.9)	15	4.60	0.012	0.0
Plan/organise	54.1 (11.2)	16	53.2 (10.2)	4	49.9 (11.4)	10	1.89	0.156	0.03
Organisation of materials	50.4 (11.0)	5	50.1 (11.7)	12	43.5 (9.5)	2	6.50	0.002*	0.10
Monitor	57.2 (12.6)	24	57.3 (11.3)	20	54.1 (11.4)	14	1.08	0.342	0.02

parents suspected FASD. In addition, some potential features of FASD were observed in the group of children without suspicion of FASD. These results are in line with available research indicating that 52% of the adoptees from CEE countries have FASD (12). Children with an FASD diagnosis were more likely to display growth deficiency, be enrolled in special education and have more problems with executive functioning than children whose parents did not suspect FASD. On executive functioning, parents of children diagnosed with FASD reported more problems with inhibition, emotional control and organisation of materials than parents of children without suspected FASD. Children with suspected FASD typically scored between children diagnosed with FASD and children without a suspicion of FASD, indicating that the suspicion could have been correct in some cases.

Polish children whose adoptive parents did not suspect FASD had fewer features of FASD. However, features of FASD were also present in this group. Some of these children may have had FASD, but had not been diagnosed. Many children and adults with FASD go unrecognised (5,15) and this is possibly the case for some children in the current sample as well. Adoptive parents had no information on prenatal alcohol exposure for 28% of the children in our sample, making a diagnosis of FASD more difficult, especially when the FAS facial characteristics were not present (4).

An alternative explanation might be that many of the Polish adoptees had been exposed to other early life risk factors, such as institutional care and neglect or that prenatal exposure to alcohol occurred alongside these other early life risk factors. The behavioural problems and developmental delays in Polish adoptees cannot, therefore, solely be attributed to prenatal alcohol exposure. Behavioural manifestations of damage to the central nervous system after prenatal exposure to alcohol might resemble behaviour that is often seen in children who have been institutionalised, such as inattention/overactivity, quasi-autism, cognitive impairment and disinhibited attachment (27). As multiple risk factors often occur together, and pre-adoptive history is partly unknown, it is difficult to disentangle behavioural problems resulting from prenatal exposure to alcohol from behavioural problems related to other risk factors.

Strengths, limitations and future directions

This study provides insights into the development of children adopted from Poland. To date, no information has been available on the presence of features of FASD in this group of adoptees. A limitation of the study is that we relied on passive surveillance. The number of children with an FASD diagnosis might, therefore, be an underestimation of the actual number of children with FASD. In addition, the children in our sample diagnosed with FASD were seen by different clinicians, possibly using different diagnostic criteria.

Parental ratings of behaviour were used to measure executive functioning. Previously reported correlations between the BRIEF and experimental tasks on executive functioning were low (24). For future research, we therefore recommend including laboratory tests of executive functioning, as well as parental ratings. Further research is needed to examine whether adoptive parents of children with FASD experience increased levels of stress, as an association between parental stress and behavioural problems in adoptees was found in earlier studies (1,28).

Practical implications

The high prevalence of FASD in children adopted from Poland points to the need for systematic attention that focuses on the potential consequences of prenatal alcohol exposure. Early diagnosis of FASD has been associated with more positive long-term outcomes and is important in the prevention of secondary disabilities, such as mental health problems or later trouble with the law (29). To enable children with FASD to be identified at an early stage, it is recommended that all children adopted from Poland are screened for FASD and subsequently diagnosed if indications for FASD are present. Prior to adoption, prospective adoptive parents of children adopted from Poland and other CEE countries need to be informed about FASD and other possible medical, behavioural problems and developmental delays. Specialised professional assistance with expertise on FASD and adoption related issues is also highly recommended. However, the number of effective interventions for children with FASD is still limited (30) and may be unavailable for families. Therefore, a strong need exists to develop more effective interventions for children with FASD and make these available for adoptive parents and their children in the near future.

CONCLUSION

The prevalence of FASD in children adopted from Poland is high, with strong indications that some children with FASD still go undiagnosed. Adoptive parents and professionals working with adoptees need to be aware of the possible consequences of prenatal exposure to alcohol. Early intervention and a stable family environment may contribute to the prevention of secondary disabilities in these children.

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