

Orthodontic evaluation of children and adolescents with different types of Foetal Alcohol Syndrome Disorders

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Email: info@czochońska.com**Abstract**

Objective: To assess general and oral health status, including the presence of malocclusion and orthodontic treatment needs, in children and adolescents with FASD in relation to different types of this disorder.

Setting and Sample Population: A total of 67 participants (29 males and 38 females) aged 2.5-17.8 years with confirmed diagnosis of FASD were included.

Materials and Methods: The participants were divided into three subgroups: foetal alcohol syndrome (FAS), partial foetal alcohol syndrome (PFAS) and alcohol-related neurodevelopmental disorders (ARND). General health, oral health status, history of dental trauma, presence of dysfunctions or parafunctions, and occlusion were examined. Additionally, the Index of Orthodontic Treatment Need (IOTN) index was calculated. Selected variables were compared to the results obtained from a national monitoring survey on the oral health conditions in Poland.

Results: In children and adolescents with FASD, a number of systemic anomalies including alimentary, neurological and musculoskeletal disorders were present. Dysfunctions and parafunctions such as mouth breathing and thumb sucking were frequently recorded. Children with FASD had an increased prevalence of distal occlusion and crossbite compared to the general population. Malocclusions were more often identified in the FAS group. No significant differences in the IOTN between different FASD groups were found. Borderline need for orthodontic treatment was more frequent in children with FASD compared to controls.

Conclusion: Early screening for the presence of dysfunctions/parafunctions and malocclusions in children with FASD is recommended, so that orthodontic prophylaxis and state-funded orthodontic care programmes for these children are implemented.

KEYWORDS

fetal alcohol syndrome, fetal alcohol syndrome disorders, index of orthodontic treatment need, prenatal alcohol exposure, solitary median maxillary central incisor syndrome

1 | INTRODUCTION

Foetal alcohol syndrome (FAS) is the most severe form of developmental disorders resulting from prenatal exposure to alcohol and accounts

for approximately 10% of all foetal alcohol syndrome disorders (FASD). Other disorders also include partial foetal alcohol syndrome (PFAS) and alcohol-related neurodevelopmental disorders (ARND).¹ For the first time, FAS was described as a separate medical entity by Lemoine

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et al in 1968.² In 1973, Jones et al presented a full description of features of dysmorphic face and defined it as foetal alcohol syndrome.³

Clinical features of FAS include the following:

1. craniofacial deformities including small cranial circumference, short neck, low, asymmetric position of the ears, deformed auricles, narrow forehead, retrognathia and retrogenia, flat mid-face, narrow palpebral fissures, eyes farther apart than normal, epicanthal fold, short and small nose, long upper lip with narrow vermilion border, flattened or absent midline groove in the philtrum;
2. damage to the central nervous system;
3. low height and weight; and
4. skeletal and circulatory developmental disorders.⁴

National Agency for Solution of Alcohol Problems in Poland performed a multi-specialist study, which revealed that the prevalence of FAS in the population is no less than 4 in 1000, and when all disorders constituting FASD are considered, the figure rises to at least 20 in 1000, which is similar to other countries.^{5,6} However, not all children instantly undergo diagnostic procedures; it is paediatricians, psychologists, adoptive or foster parents who refer the child to specialists for FASD diagnosis. In FAS, the diagnostic process may be problematic due to heterogenic character of the disorder or absence of the standardized diagnostic system. In Poland, the most frequently used system was the 4-Digit Diagnostic Code proposed by the Washington University in Seattle in 2004, which was used to assess the presence and intensity of four features characteristic of children with FAS such as low height and weight, specific facial characteristics, damage to the nervous system and exposure to alcohol.⁴ The Canadian questionnaire of 2005 was also applied.⁷ In 2017, Astley et al demonstrated that the two systems show similarities with respect to application of precise criteria and diagnosing the whole spectrum of consequences of prenatal exposure to alcohol.⁸ Yet, Canadian guidelines are more accurate in defining neuropsychological diagnosis. Due to these diagnostic challenges, and difficulties to collect a representative sample, the knowledge of dental and orthodontic problems of children with FAS is limited. So far, no study has evaluated different types of FASD with regard to orthodontic diagnostics. The present study aims at assessing general and oral health status, including the presence of malocclusion and orthodontic treatment need index in children and adolescents with FASD in relation to different types of this disorder.

2 | MATERIALS AND METHODS

All eligible individuals, who were diagnosed with the foetal alcohol syndrome disorders (FASD), were enrolled. The sample was collected with the help offered by foundations for disabled children, FASD diagnostic centres, associations between adoptive and foster families, and orphanages. Participation in training courses and support group

meetings for families with FASD children, as well as announcements about this particular study in social media, was also applied to approach willing participants.

In all the children, medical and dental histories, and the clinical examination, were taken and performed by the same researcher (KL). The same researcher took extraoral and intraoral photographs as well as impressions for diagnostic models. The evaluation of the records was performed together with a senior researcher (EC) who was blinded to the group's status.

2.1 | FASD diagnostic procedures

All the participants included in the study enclosed a written diagnosis confirming the presence of FASD, that is 4-digit diagnostic questionnaire from Seattle and the Canadian questionnaire, which were used by two and three diagnostic centres respectively.

2.2 | Medical and dental histories

The medical history concerned the occurrence of pain sensations, systemic diseases and allergies, history of trauma and treatments within the head and the neck. The dental history focused on the presence of dentofacial deformities, and cleft lip and/or palate in particular, history of dental trauma, presence of oral dysfunctions or parafunctions, and previous orthodontic treatment (Table 1). In general, oral dysfunctions or parafunctions include malfunction of the lips, the tongue or the oropharynx (mouth breathing), or abnormal functions or habits such as thumb/finger and dummy sucking or nail biting.

Selected variables recorded in the study sample were compared to the results obtained from a national monitoring survey on the oral health conditions in the Polish population. The subjects were selected through a 3-stage cluster sampling procedure representing 46 lower secondary schools in urban and rural areas in 10 out of 16 provinces of the country.⁹

2.2.1 | Clinical dental examination

All the participants underwent the clinical examination including the assessment of active caries (Yes/No), enamel hypoplasia (Yes/No) and oral hygiene status. Oral hygiene was assessed as "poor" if dental plaque covered >70% of tooth surfaces, and as "good" if it was <30%. For values between 30% and 70%, the overall oral hygiene was assessed as "moderate".

In all children younger than 6 years, it was not possible to obtain impressions for diagnostic models. The Baume's classification of primary molar relations (flush terminal plane, distal step, mesial step), the presence of open bite and tooth crowding was recorded during the clinical examination.¹⁰

Intraoral and extraoral photographs were taken in all the participating children.



TABLE 1 Description of the FASD groups and the outcomes of the medical anamnesis

Diagnosis	FASD	FAS	PFAS	ARND
Number of children (%)	67 (100%)	34 (51%)	11 (16%)	22 (33%)
Age (range, mean)	2.5-17.8 (10.4)	2.5-17.8 (10.3)	5.5-15.7 (10.3)	16.11 (10.5)
Medical history				
Neurological disorders	14 (21%)	6 (18%)	0 (0%)	8 (36%)
Thyroid diseases	4 (6%)	1 (3%)	2 (18%)	1 (5%)
Postural defects	13 (19%)	7 (21%)	0 (0%)	8 (36%)
Muscle tone abnormalities	17 (27%)	7 (21%)	0 (0%)	10 (45%)
Heart defects	2 (3%)	1 (3%)	1 (9%)	0 (0%)
Kidney insufficiency	1 (2%)	0 (0%)	0 (0%)	1 (5%)
Disturbed sensation of pain	4 (6%)	0 (0%)	1 (9%)	3 (14%)
Disturbed sleep	2 (3%)	1 (3%)	0 (0%)	1 (5%)
Tactile hypersensitivity	3 (5%)	2 (6%)	1 (9%)	0 (0%)
Headaches	4 (6%)	1 (3%)	2 (18%)	1 (5%)
Arthralgia	1 (2%)	0 (0%)	0 (0%)	1 (5%)
Allergy to nickel, latex, acrylic	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Food allergies	19 (28%)	8 (24%)	4 (36%)	7 (32%)
Dust allergies	1 (2%)	1 (3%)	0 (0%)	0 (0%)
Adenoidectomy	12 (18%)	5 (15%)	1 (9%)	6 (27%)
Corrected nasal septum	2 (3%)	0 (0%)	2 (18%)	0 (0%)

2.2.2 | Model analysis

Dental relations were assessed in all the participants >6 years, in whom it was possible to take alginate impressions for diagnostic models. The occlusal relations were assessed in three planes:

Sagittal

- Molar relation (Angle Class I, II, III),
- Canine relation (Class I, II, III),
- Overjet: normal (1-<3.5 mm), increased (>3.5 mm) and decreased/reverse (<1 mm).

Vertical

- Overbite: normal (1-3 mm), deep (\geq 4 mm) and reduced (\leq 1 mm).

Transversal

- Presence of crossbite or scissor bite (unilateral/bilateral, anterior/posterior),
- Discrepancy between the lower and the upper midlines (Yes/No, if discrepancy exceeded 3 mm).

Space conditions (crowding/spacing) were additionally assessed.

Molar and canine relations were recorded as Class II or Class III, if present only on one side.

2.2.3 | Orthodontic treatment need

The need for orthodontic treatment was assessed using the Index of Orthodontic Treatment Need (IOTN) in all the participants who

had diagnostic models made.¹¹ The chi-square test was used to determine relationships between the scores of the IOTN and different FASD groups.

The study was approved by the Bioethics Committee of the Medical University of Warsaw, Poland (No KB/204/2016).

3 | RESULTS

A total of 67 participants (29 boys and 38 girls) were enrolled. They were aged between 2.5 and 17.8 years (mean age: 10.4 years). The study group was divided into three subgroups based on the diagnosis of FASD: foetal alcohol syndrome (FAS): 34 subjects, partial foetal alcohol syndrome (PFAS): 11 subjects and alcohol-related neurodevelopmental disorders (ARND): 22 subjects. Most of the children were living in foster families (51 children), two children were in orphanages, and fourteen children had been adopted. They were mainly living in Masovia, Pomerania, Kuyavia-Pomerania, Holy Cross and Silesia provinces, whose population accounts for approximately 41% of inhabitants of Poland. The mean age of study subjects in all three subgroups was similar and mostly included participants in the late mixed or the permanent dentition periods. The controls included 628 children with the mean age similar to the study sample (10 years).

3.1 | Medical and dental histories

Alimentary tract disorders were the most commonly reported ones with approximately 1/3 of the participants having food allergies in



each of the three FASD subgroups (Table 1). These were followed by muscle tone abnormalities and incorrect posture reported by the participants with ARND and FAS. Neurological disorders were reported in the FAS and ARND groups.

In Table 2, the outcomes of dental anamnesis including the controls are presented. Very few participants with FASD had a previous orthodontic treatment as opposed to 27% of the control children. Dysfunctions and parafunctions were common among children with FASD. More than half of the FASD participants were mouth breathers compared to 24% reported in the control sample. In subjects with PFAS, the tongue thrusting was common, but in other subgroups this dysfunction was infrequent and not higher than in the controls. Thumb sucking was common in children with FASD (41%). The frequency of nail biting in the FASD group was not much different from the controls. Caregivers of the children with PFAS and ARND reported that some of them were sleeping with their heads tilted backward. None of the participants or the controls manifested cleft lip and/or palate.

3.1.1 | Clinical dental examination

Table 3 presents the results of the clinical dental examination in relation to FASD diagnostics. The national monitoring survey performed in 10-year-olds did not include the assessment of caries, enamel hypoplasia or oral hygiene. Active caries was found in 45% of the participants with FASD, but good oral hygiene was seen in 53 per cent. Enamel hypoplasia was present in two subjects (3%). There were no participants with clinical signs of dentofacial deformities, except for one child with the solitary median maxillary central incisor syndrome (SMMCI) in the FAS group.

In the fourteen youngest children with deciduous or early mixed dentition, it was not possible to make diagnostic models due to lack of cooperation and severe retardation. Table 4 depicts occlusal relationships in these patients. In the majority of cases, the occlusal

norm was present (78%). The presence of anterior open bite was diagnosed in two children with FAS.

3.1.2 | Analysis of diagnostic models

In fifty-three of the participants (79%) aged between 6.5 and 17.8 years (mean: 11.7 years), plaster diagnostic models were obtained. Table 5 presents the results of the model analysis.

In the study group, normal occlusion was present in 10 participants (19%), and 2 had had previous orthodontic treatment. Only one child with FAS presented a normal occlusion. The remaining 43 children (81%) had different occlusal disturbances. Distal occlusion was present in 55% of the children with FASD in comparison with 30% of the controls. Children with FAS presented the highest number of occlusal disturbances in comparison with the other FASD groups.

3.2 | Sagittal relations

Due to the absence of mandibular and/or maxillary molars, it was not possible to evaluate Angle classification in four children with FASD, while it was determined in all control subjects. Angle Class I was the most frequently recorded in the control sample and Angle Class II in the FASD sample. More than 80% of subjects with FAS had Angle Class II or Class III. In seventeen participants, the Angle Class I co-existed with other dental anomalies such as tooth crowding (6), crossbite (5), anterior open bite (2), tooth spacing (1) or deep bite (1).

It was not possible to evaluate canine classification in sixteen children with FASD due to the absence of canines, but it was recorded in all the control children. Here, canine Class I was more frequently recorded. Increased overjet was present in 53% of the subjects with FASD in comparison with 30% of the controls.

TABLE 2 Description of the FASD groups and the outcomes of the dental anamnesis including controls

Diagnosis	FASD	FAS	PFAS	ARND	Controls
Number of children (%)	67 (100%)	34 (100%)	11 (100%)	22 (100%)	628 (100%)
Age (range, mean)	2.5-17.8 (10.4)	2.5-17.8 (10.3)	5.5-15.7 (10.3)	16.11 (10.5)	10 (10)
Previous orthodontic treatment	5 (7%)	2 (6%)	0 (0%)	3 (14%)	169 (27%)
Upper incisor trauma	3 (4%)	2 (6%)	0 (0%)	1 (5%)	Nr
Mouth breathing	37 (56%)	16 (47%)	6 (55%)	15 (68%)	149 (24%)
Tongue thrusting	10 (15%)	3 (9%)	4 (36%)	3 (14%)	143 (23%)
Thumb sucking	28 (41%)	16 (47%)	3 (27%)	9 (41%)	Nr
Nail biting	25 (37%)	12 (36%)	4 (36%)	9 (41%)	187 (30%)
Head tilted backward during sleep	8 (12%)	2 (6%)	3 (27%)	3 (14%)	Nr
Bruxism	0 (0%)	0 (0%)	0 (0%)	0 (0%)	Nr
Cleft lip and/or palate	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)

Abbreviation: Nr, Not recorded.

**TABLE 3** Results of the clinical dental examination in relation to different FASD groups

Diagnosis	FASD	FAS	PFAS	ARND
Number of children (%)	67 (100%)	34 (51%)	11 (16%)	22 (33%)
Age (range, mean)	2.5-17.8 (10.4)	2.5-17.8 (10.3)	5.5-15.7 (10.3)	16.11 (10.5)
Active caries	30 (45%)	16 (47%)	3 (27%)	11 (50%)
Enamel hypoplasia	2 (3%)	1 (3%)	1 (9%)	0 (0%)
Oral hygiene				
Good	35 (52%)	16 (47%)	7 (64%)	12 (55%)
Moderate	23 (34%)	13 (38%)	3 (27%)	7 (32%)
Poor	9 (13%)	5 (15%)	1 (9%)	3 (14%)

TABLE 4 The results of occlusal analysis based only on clinical examination in fourteen youngest children in relation to different FASD groups

Diagnosis	FASD	FAS	PFAS	ARND
Number of children (%)	14 (100%)	8 (57%)	3 (21%)	3 (21%)
Age (range, mean)	2.5-7.11 (5)	2.5-7.11 (4.3)	5.5-7.6 (6.4)	5-6.1 (5.7)
Flush terminal plane	11 (78%)	5 (63%)	3 (100%)	3(100%)
Mesial step	0 (0%)	0 (0%)	0 (%)	0 (0%)
Distal step	1 (7%)	1 (13%)	0 (%)	0 (0%)
Anterior open bite	2 (14%)	2 (26%)	0 (0%)	0 (0%)
Crossbite	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Tooth crowding	1 (7%)	0 (0%)	0 (0%)	1 (33%)

3.3 | Vertical relations

Most participants with FASD, regardless of subgroup divisions, had normal or deep overbite. Reduced overbite was more common in subjects with ARND (21%).

3.4 | Transversal relations

Crossbites were present in 28% of children with FASD in comparison with 13.5% in the controls. Crossbites and lower midline deviation predominated in subjects with FAS. Scissor bite was most frequently diagnosed in the FAS and PFAS groups.

3.5 | Space problems

Lack of space for the permanent teeth was observed in more than 40% of participants with FASD. In subjects with FAS, tooth crowding or spacing were more frequently recorded than in other groups.

3.5.1 | Orthodontic treatment needs

The IOTN relative to different subgroups of FASD and the controls is shown in Table 6. Significant need for orthodontic treatment was more often recorded in subjects with FAS. No significant differences between the FASD group scores and the scores of the IOTN were present ($P = .99$). Little or no need for orthodontic treatment was the most common finding in the control sample (41%), while the participants

with FASD were frequently categorized as borderline need of treatment (43%). No difference in great need for treatment was seen between children and adolescents with FASD and the controls.

4 | DISCUSSION

The present study is the first to describe orthodontic evaluation in relation to FASD subgroups. In recent years, women have been observed to consume more alcohol, particularly in the 18- to 29-age range.⁵ This, in turn, may lead to the increase in the number of children born with FASD who require dental and orthodontic care. Adoptive or foster parents may benefit from the results of the present study so that orthodontic prophylaxis and therapy is better suited to the needs of children with FASD.

The main problem when a study on children and adolescents with FASD is conducted is gathering a reliable sample. In most cases, the children are adopted, live in foster families or orphanages and are, therefore, scattered all over the country. All available means to access children and adolescents with FASD had been exhausted in the present study, which led to a relatively high number included.

4.1 | General medical conditions

Medical history findings are in line with existing reports of how alcohol consumption during the gestation period affects several body systems including the nervous, cardio-vascular, alimentary, endocrine and musculo-skeletal system.^{12,13} In the present study, the increased presence of food allergies, muscle tone dysfunction,



TABLE 5 Results from the model analysis in different FASD groups and controls

Diagnosis	FASD	FAS	PFAS	ARND	Controls
Number of children (%)	53 (100%)	26 (100%)	8 (100%)	19 (100%)	628 (100%)
Age (range, mean)	6.5-17.8 (11.7)	7-17.8 (12.6)	7-15.7 (11.7)	6.6-16.11 (11.2)	10 (10)
Normal occlusion	10 (19%)	1 (4%)	3 (37%)	6 (31%)	34 (5.4%)
Distal occlusion	29 (55%)	16 (62%)	4 (50%)	10 (53%)	186 (30%)
Mesial occlusion	2 (4%)	2 (8%)	0 (0%)	0 (0%)	24 (4%)
Angle Class I	19 (36%)	4 (19%)	4 (50%)	10 (53%)	371 (59%)
Angle Class II	28 (53%)	16 (61%)	4 (50%)	8 (42%)	238 (38%)
Angle Class III	2 (4%)	2 (8%)	0 (0%)	0 (0%)	19 (3%)
Canine Class I	13 (25%)	6 (23%)	1 (13%)	6 (32%)	347 (55%)
Canine Class II	22 (41%)	11 (42%)	4 (50%)	7 (37%)	258 (41%)
Canine Class III	2 (4%)	2 (8%)	0 (0%)	0 (0%)	23 (4%)
Overjet					
Normal	22 (41.5%)	7 (27%)	4 (50%)	11 (58%)	410 (65%)
Increased	28 (53%)	16 (61.5%)	4 (50%)	8 (42%)	197 (31.5%)
Decreased/reverse	3 (5.5%)	3 (11.5%)	0 (0%)	0 (0%)	21 (3.5%)
Overbite					
Normal	25 (47%)	13 (50%)	4 (50%)	8 (42%)	362 (57.5%)
Deep	22 (42%)	11 (42%)	4 (50%)	7 (37%)	162 (26%)
Reduced	6 (11%)	2 (8%)	0 (0%)	4 (21%)	104 (16.5%)
Crossbite	15 (28%)	10 (38%)	0 (0%)	5 (26%)	85 (13.5%)
Scissor bite	4 (7.5%)	3 (11.5%)	1 (12.5%)	0 (0%)	4 (0.6%)
Lower midline deviation	18 (34%)	11 (42%)	3 (37.5%)	4 (21%)	Nr
No deviation	16 (30%)	2 (8%)	4 (50%)	10 (53%)	Nr
No space deficiency	23 (43%)	6 (23%)	4 (50%)	13 (68%)	Nr
Crowding	23 (43%)	14 (54%)	4 (50%)	5 (26%)	Nr
Spacing	4 (8%)	3 (11.5%)	0 (0%)	1 (5%)	Nr
Diastema	3 (6%)	3 (11.5%)	0 (0%)	0 (0%)	Nr

Abbreviation: Nr, Not recorded.

TABLE 6 The Index of Orthodontic Treatment Needs (IOTN) in different FASD groups and controls

IOTN	FASD	FAS	PFAS	ARND	Controls
Number of children (%)	53 (100%)	26 (100%)	8 (100%)	19 (100%)	628 (100%)
Age (range, mean)	6.5-17.8 (11.7)	7-17.8 (12.6)	7-15.7 (11.7)	6.6-16.11 (11.2)	10 (10)
No or little need (1 & 2)	15 (28.5%)	6 (23%)	2 (25%)	7 (37%)	256 (41%)
Borderline need (3)	23 (43%)	12 (46%)	4 (50%)	7 (37%)	195 (31%)
Great need (4 & 5)	15 (28.5%)	8 (31%)	2 (25%)	5 (26%)	117 (28%)

postural defects and neurological disorders were seen more often in subjects with FAS.

4.2 | General dental conditions

4.2.1 | Dental caries and oral hygiene status

Active caries was present in approximately 50% of the participants, but in more than 50% oral hygiene was assessed as "good". This may

be due to the fact that some of the participants had not been long in adaptive or foster families, in which proper oral hygiene and the need for dental treatment are usually emphasized. Naidoo et al reported a higher percentage of tooth surfaces covered with bacterial plaque (>75%); bleeding on probing was observed in almost two-thirds of children with FAS.¹⁴ The DMFT score for the FAS sample, on the other hand, was slightly higher, though not significantly different from that of the controls. Blanck-Lubarsch et al demonstrated significant differences within the DMFT index in children with FAS in comparison with the controls, in whom diet may have impacted the



results.¹⁵ A study by Amos-Kroohs et al revealed dietary irregularities in patients with FAS including decreased satiety, constant snacking and higher number of meals daily in comparison with healthy controls.¹⁶ Snacking between meals contributes to caries, and these improper dietary behaviours of patients with FAS may be reflected by a higher DMFT index or caries despite good oral hygiene, as found in the present study. Dietary counselling and further studies on dietary habits of patients with FAS would be recommended.

4.2.2 | Enamel hypoplasia

Low prevalence of enamel opacities was confirmed in children with FAS.^{14,17} In a study by Blanck-Lubarsch et al, developmental defects of enamel were more prevalent in children with FAS in comparison with the controls, but no other groups of the FASD spectrum were assessed.¹⁵

4.2.3 | Cleft lip and/or palate

Yin et al conducted a meta-analysis concerning the co-existence of FAS with cleft lip and/or palate.¹⁸ They observed no significant differences between drinking and non-drinking mothers in the risk of having a child with a non-syndromic oral cleft. Also, they found no confirmatory evidence for the presence of a dose-response relation between alcohol consumption and the increase in the risk of oral cleft, which was confirmed in the present study. During the enrolment process for the present study, two children aged 9 and 12 years with suspected FAS who manifested cleft lip and palate were contacted. They were, subsequently, excluded because of the absence of a written confirmation of the FAS diagnosis.

4.2.4 | Dysfunctions and parafunctions

Dysfunctions and parafunctions were common in the FASD group, and they could impact the occurrence of malocclusions. While mouth breathing is usually associated with the presence of occlusal disturbances, oral habits play a significant role only in some of them, probably because their relevance is lower than other factors implicated in the aetiopathogenesis of malocclusions.¹⁹ Results of the study confirmed higher incidence of distal occlusion, increased overjet and presence of crossbites in the FASD sample in comparison with the controls, which may indicate an aetiological relationship of their occurrence with mouth breathing. Mouth breathing was found in 56% of the FASD participants, which is similar to the study by Blanck-Lubarsch et al where this dysfunction had also a higher prevalence than in the control group.²⁰ Contrary to the results obtained by Blanck-Lubarsch et al, prolonged dummy or thumb sucking was not common in the study sample. A higher number of mouth breathers was present in subjects with ARND, which may be related to the more frequent occurrence of neurological disorders in this group.

An interesting finding reported by the caregivers is a frequent occurrence of characteristic position with the head tilting backwards during sleep. This atypical head position, and sleeping on one's back, which was reported in all FASD subgroups, may indicate nasal obstructions or mouth breathing. The relevant literature confirms the occurrence of significantly shorter total sleep duration, lower sleep efficiency and more nocturnal waking episodes in children with FASD.^{21,22} Thus far, a detailed description of a child's position with this syndrome during sleep has not been described.

4.2.5 | Orthodontic treatment

Although different malocclusions were often seen in the study group, few subjects with FASD, and none with PFAS, were orthodontically treated. This is apparently due to the absence of state-funded orthodontic care for children with FAS in Poland and lack of awareness of orthodontic treatment needs on the part of legal caregivers. Previous orthodontic treatment was reported in 27% of the control children. In Poland, only orthodontic treatment with removable appliances up to the age of 12 years is refunded by the state except for patients with severe dentofacial deformities. It significantly restricts access to treatment by low-income families. The results of this study may possibly help to increase awareness of orthodontic needs in children with FASD and to include them in the state-funded orthodontic care system.

4.2.6 | Prevalence of malocclusions

In fourteen youngest children, no significant deviations from the occlusal norm were seen during clinical examination. This may indicate that in younger children with FASD, the incidence of malocclusions does not significantly differ from the general population, yet they have to be interpreted with caution due to the small size of the sample.

Different types of malocclusion were diagnosed in over eighty per cent of participants in the present study. Church et al examined twenty-two subjects with FASD and also reported frequent occurrence of malocclusions in this group. However, in this study orthodontic diagnostics was performed only in fifteen participants of whom four had Class I and six had Class II and III malocclusions; the age range from 3 to 26 years was also wide.¹⁷ Naidoo et al assessed the prevalence of occlusal defects in a group of ninety children with FAS with mean age of 8.9 years. Significantly more dentofacial anomalies were found in the study group than in the controls. The most frequently occurring anomalies were crowding, increased overjet and open bite, but no classification of malocclusion was undertaken.¹⁴ In the present study, more than half of participants with FASD were diagnosed with distal occlusion, and in subjects with FAS, this percentage was sixty two. Increased overjet and deep bite, which are often seen in disocclusions, were more frequent in the FASD group in comparison with the controls. These figures are also



higher than those reported for the children with mixed dentition in Poland, where disocclusion was observed in 12% of subjects.²³

Crossbites, which often accompany respiratory dysfunctions, were more frequent in the FASD sample than in the controls. Subjects with FAS were more often diagnosed with crossbite and lower midline deviation than children and adolescents in other FASD subgroups, which is similar to other report.^{19,20}

Half of the children with FAS manifested tooth crowding, which was also seen in the PFAS and ARND groups. Distinct differences in the presence of malocclusions were not demonstrated between different FASD groups, but children and adolescents with FAS had more deviations from the occlusal norm than subjects in other groups. This may indicate that the severity of the syndrome has an impact on the development of occlusal disturbances.

4.2.7 | SMMCI Syndrome

In one participant, the solitary median maxillary central incisor syndrome (SMMCI) was present. The term refers to the presence of only one central incisor in the centre of the anterior maxilla with a concomitant absence of the upper lip fraenum. SMMCI occurs at a rate of 1:50 000 births and is a complex disorder involving midline structures of the head and that includes the central incisor tooth germ.²⁴ SMMCI may appear as an isolated trait or in association with other midline developmental anomalies, although its association with FASD would be hard to establish on the basis of the present study.²⁵

4.3 | IOTN

So far, orthodontic treatment needs in children and adolescents with FASD have not been described in the literature. The higher prevalence of borderline need for orthodontic treatment in the study sample may indicate the influence of dysfunctions and parafunctions in the aetiology of malocclusion in children with FASD. The results of the study indicate that the state-funded prophylaxis and orthodontic care should include children with FASD, and with FAS in particular, as more occlusal disturbances were recorded in this group.

4.4 | Limitations

The study sample included eleven children and adolescents with PFAS when compared with a three- and two-fold higher number of subjects with FAS and ARND respectively. Also, the number of participants in the national survey who served as the controls was much higher than the number of participants with FASD. These facts have to be accounted for in the interpretation of results.

Different terminology describing disorders associated with the prenatal alcohol exposure was applied and evolved over the years. In 2020, the National Agency for Solutions to Alcohol Problems in Poland initiated an interdisciplinary workshop with the aim to

provide general guidelines for the FASD diagnosis. It is now recommended to differentiate between diagnostic categories such as FAS, ND-PAE (neuro-developmental disorders associated with prenatal alcohol exposure) and FASD risk (non-diagnostic category). The clinical examination of the children participating in the study was performed in the years 2016-2019 when the current guidelines for the FASD diagnosis were not applied. However, all the participants had submitted a written diagnosis confirming the presence of a specific type of the FASD. The process of adoption or allocation to a foster family follows strict rules, also regarding the FASD diagnosis. It could be speculated that the ARND category should now be described as the ND-PAE, but such diagnosis was not possible at the time of the examination. A more detailed description of the physical characteristics of the children with FASD included in the present study could be beneficial in response to the diagnostic changes and evolutions.

The participants represented a wide age range including children with deciduous dentition and late adolescents. Since the study participants were scattered all over Poland, a challenge was encountered in performing full orthodontic diagnostics. When calculating the IOTN index, taking a panoramic radiograph would be recommended to evaluate the presence of missing or impacted teeth. However, obtaining a radiograph was not always possible due to different economic situations in families, or limited access to dental care services.

5 | CONCLUSIONS

In children and adolescents with FASD, a number of systemic anomalies including alimentary, neurological and musculo-skeletal disorders were present. Dental history findings frequently included dysfunctions and parafunctions such as mouth breathing and thumb sucking. Children with FASD have increased prevalence of distal occlusion and crossbites in comparison with the general population. Malocclusions were more often identified in the FAS group. No significant differences in the IOTN between different FASD groups were found. Borderline need for orthodontic treatment was more common in children with FASD compared to controls. Early screening for the presence of dysfunctions/parafunctions and malocclusions in children with FASD is recommended, so that orthodontic prophylaxis and the state-funded orthodontic care programmes for these children are implemented.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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