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Understanding the Educational and Social Difficulties in Children with Hemifacial Microsomia – A Literature Review

Michał Pniak M.P. – corresponding author
John Paul II Memorial Masovia Provincial Hospital in Siedlce, Poland
https://orcid.org/0009-0006-2982-6078
e-mail: michal.pniak@tlen.pl

Paweł Miklis P.M.
University Clinical Center of the Medical University of Warsaw, Poland
https://orcid.org/0009-0008-4578-233X
e-mail: pawelmiklis3@wp.pl

Maciej Mawlichanów M.M.
Military Institute of Medicine, Warsaw Poland
https://orcid.org/0000-0002-6543-8105
e-mail: mmawlichanow@gmail.com

Aleksandra Ciesielska A.C.

Mazovian Rehabilitation Center "STOCER" Sp. z o.o. Railway Hospital in Pruszków, Poland
https://orcid.org/0009-0000-5015-6140
e-mail: aleksandra.ciesielska99@gmail.com

Aleksandra Sieradzka A.S.
Medical University of Warsaw
https://orcid.org/0009-0004-2281-7617
e-mail: aleksandra.w.sieradzka@wp.pl

Krzysztof Szerej K.S.
District Hospital in Sochaczew, Poland,
https://orcid.org/0009-0003-7581-4965
e-mail: krzychszerej@gmail.com

Zofia Czaplińska-Paszek Z.C.P.
Mazovian Rehabilitation Center "STOCER" Sp. z o.o. Trauma Surgery Hospital, Warsaw, Poland
https://orcid.org/0000-0003-2429-8262
e-mail: czaplinskapaszek@gmail.com

Alicja Kot A.K.
Medical University of Warsaw
https://orcid.org/0009-0001-2999-6775
e-mail: alicja.kot28@gmail.com

Marta Wojtczak M.W.
County Health Center in Otwock Sp. z o.o. Otwock, Poland
https://orcid.org/0009-0007-0032-7520
e-mail: martaw.1998@gmail.com

Natalia Myśliwiec N.M.
National Medical Institute of the Ministry of the Interior and Administration, Warsaw, Poland
https://orcid.org/0000-0002-4359-9899
e-mail: nataliamysliwiec45@gmail.com

Adrian Różycki A.R.
Medica Plus Family Clinic Sp. z o. o., Gdańsk, Poland
https://orcid.org/0000-0002-9110-5443
adrian.rozycki@gmail.com

ABSTRACT

Introduction: Hemifacial microsomia (HFM) is a congenital craniofacial defect, the second most common after cleft lip and palate. It is characterized by a wide range of symptoms, including mandibular hypoplasia, ear anomalies, and soft tissue defects. Patients may experience difficulties in learning, communication, and social relationships, which affect their quality of life and self-esteem. Study aims to evaluate the impact of defect on the neurodevelopmental and psychosocial outcomes in affected children.

Materials and methods: Materials for the review were selected based on an analysis of publications published in the last 20 years.

Results: In HFM patients with microtia, mandibular hypoplasia, young maternal age, and additional developmental anomalies, worse outcomes were observed in verbal, non-verbal, and academic skills. Studies on emotional functioning demonstrated a similar level of behavioral adjustment compared to control groups. Adolescents with HFM exhibited a lower tendency toward aggression and rule-breaking. Social problems, such as less frequent contact with peers or rejection by the group, were more common. This was particularly true for children with microtia and mandibular hypoplasia.

Discussion: New studies emphasize that although some young children initially do not exhibit neurological or language deficits, there is an increased risk of educational and psychosocial difficulties in later life. Attention should be paid to patients with mandibular hypoplasia, microtia, and additional defects. Negative experiences in peer relationships and hearing loss may further adversely affect self-esteem and social functioning.

Conclusions: HFM can affect the neurological and psychosocial development of children, especially in language and educational areas. Early intervention, assessment of psychosocial problems, provision of appropriate educational resources, and psychological support are crucial to improving patients' quality of life.

Key words: hemifacial microsomia, microtia, psychosocial adjustment, neurocognitive

List of abbreviation: ASEBA - Achenbach System of Empirically Based Assessment, **Bayley-III** - Bayley Scales of Infant and Toddler Development**, CBCL** - Child Behavior Checklist, **ES** - Effect size, **HFM** - Hemifacial microsomia, **OSA** - Obstructive sleep apnoea, **PLS-5** - Preschool Language Scale, **PPVT-III** - Peabody Picture Vocabulary Test, **TRF** - Teacher's Report Form, **VMI-5** - Beery-Buktenica Developmental Test of Visual-Motor Integration, **YSR** - Youth Self-Report

INTRODUCTION

Craniofacial malformations, such as hemifacial microsomia (HFM), pose a significant clinical challenge due to the complexity of the physical manifestations. It is the second most common craniofacial defect after cleft lip and palate. The mechanism of the relationship between the defect and neurodevelopment is still not fully understood [1, 2]. Children with HFM may experience difficulties in learning, communication, and social relationships. These challenges may result from both the physical aspects of the defect and environmental factors [2, 3]. Moreover, the occurrence of problems such as anxiety, depression and low self-esteem emphasize the need for comprehensive care for patients [4].

The aim of this study is to synthesize the existing research on the psychosocial and educational aspects of the lives of patients with HFM. Also, the goal is to provide a comprehensive understanding of how living with this malformation affects individuals' psychosocial experiences, which can inform clinical practice and families. The authors want to show how great the need for further scientific research on this malformation is.

MATERIALS AND METHODS

A systematic search of databases and specialist literature was performed to identify relevant research papers in any language. Several key databases were searched, including PubMed, and Google Scholar. The keywords used included “hemifacial microsomia”, “craniofacial microsomia”, “psychosocial impact”, “quality of life”, “mental health”, “self-esteem”, “body image”. To include the latest research and advances in the subject matter. Priority was given to articles published over the last 20 years. Publications relating to congenital defect called Goldenhar syndrome or oculo-auriculo-vertebral spectrum were not taken into account.

LITERATURE REVIEW

Characteristics of the defect

Hemifacial microsomia (HFM) is a congenital defect resulting from the malformation of the first and second branchial arch [1]. The incidence of HFM in the USA is estimated at 1 in 3,600–5,600 live births. The abnormality is more prevalent in boys (3:2 ratio) and typically affects the right side of the face, moreover, the incidence of bilateral cases is 10% [5]. Mandibular hypoplasia is a characteristic feature of the discussed condition. It is associated with various osseous anomalies (i.a. maxillar hypoplasia, temporal bone and orbital malformations), ear anomalies (i.a. microtia, anotia, preauricular tags, auditory ossicle defects, and external auditory canal atresia) or even soft tissue defects (i.a. hypoplasia of masseter muscles, facial muscles or paratoid gland, and subcutaneous tissue deficiency). More than half of patients with HFM have conductive hearing loss, macrostomia and cranial nerve abnormalities are common as well [1]. This defect is often accompanied by anomalies of other organ systems for instance pulmonological, cardiac, and neurological disorders [6]. It is reported that mandibular hypoplasia results in higher rates of obstructive sleep apnoea (OSA) [7]. The severity of HFM varies greatly, depending on the number of directly or indirectly affected structures as well as on the degree of penetration [8].

Along with the deformities, patients may have problems with learning, mood, social relationships and self-esteem [9].

Assessment tools

To assess developmental delays in infants and toddlers, the Bayley Scales of Infant and Toddler Development (Bayley-III) and the Preschool Language Scale (PLS-5) are used. The Bayley-III provides information on both cognitive and motor abilities [10], while the latter tool evaluates language skills, both expressive and receptive [11].

In older children, the Achenbach System of Empirically Based Assessment (ASEBA) is utilized to assess behavioral and emotional problems as well as social competencies based on reports. The system includes questionnaires such as the Child Behavior Checklist (CBCL), Teacher's Report Form (TRF), and Youth Self-Report (YSR). These are completed respectively by parents or caregivers, teachers, and children aged 11 to 18 years. The reports allow for

comparison of results from different environments like home and school at various stages of a child's development [12]. Another test, the Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI-5), helps assess visual-motor coordination. By drawing shapes of increasing complexity, the precision of movements, the ability to imitate and reproduce patterns are assessed. Impaired coordination can affect skills such as writing, drawing, and other activities requiring precise motor control [13]. Researchers also use the Peabody Picture Vocabulary Test (PPVT-III), which assesses vocabulary comprehension and lexical knowledge. The examinee is tasked with selecting the picture that best matches the word spoken by the examiner [14].

All of the above-mentioned tools assist in evaluating the overall functioning of children, both in terms of emotional and behavioral problems, as well as academic competences. This allows for a better assessment of the challenges faced by children with craniofacial anomalies and leads to a greater understanding of their needs [10-14].

Neurodevelopmental and psychosocial complications

The neurodevelopmental status of children between 12 and 24 months was studied from 2012 to 2017 and included 108 cases. The study participants were evaluated using the FACIAL classification [15]. According to this, a malformation is considered HFM if at least one of the features listed in Table I is present [16].

FACIAL classification
Microtia
Anotia
Facial asymmetry and preauricular tag
Facial asymmetry and facial tag
Facial asymmetry and dermoid
Facial asymmetry and macrostomia
Preauricular tag and macrostomia
Facial tag and dermoid
Macrostomia and dermoid
Preauricular tag and dermoid

Table I. FACIAL classification requires the presence of one of a set of features for the defect to be considered as HFM [16].

Differences were observed in the Bayley-III and PLS-5 results compared to peers, which were insignificant before and after adjusting for variables. The estimated mean differences ranged from –0,23 to 1,79 corresponding to standardized effect sizes (ES) ranging from –0,02 to 0,12 with P values from 0,30 to 0,88. The study showed that neurological development and language were not affected in children in the early stages of growth. Also, hearing impairment or the presence of a hearing aid was not associated with different developmental outcomes. The authors noted the difficulty in obtaining specific vocal or verbal responses. The main reason is that the assessment of problem-solving skills, at such a very young age, relies largely on non-verbal functions [15].

In the USA and Canada, between 1996 and 2002, a study focusing on demographic factors and risk factors for the development of the defect was conducted [17]. The same group was invited to participate in the second phase of the study when the children were 6–7 years old. Thus, a group of 136 children with HFM was compared with a control group of 568 children. Neurological development and psychosocial status were evaluated [2, 4].

Parents and teachers provided information using the CBCL and TFR reports, respectively. Additionally, educators conducted a measurement of children's social acceptance based on ranking the child relative to classmates. Children with HFM performed worse on the VMI-5, PPVT-III, CBCL, and TRF. The differences between groups were even more marked in offspring of young mothers (<25 years old at birth), children with additional defects, and children with speech impairment, hearing loss, and visual impairment. According to the researchers, their results confirm the hypothesis of poorer verbal, non-verbal, and academic skills in individuals with the defect. Additionally, receptive language and scholastic competence were areas of particular vulnerability [2]. The authors who studied the previously mentioned children aged 12–24 months also referred to these results. Similar outcomes at such a young age may indicate that learning problems observed in older children with HFM may develop after the infant period [15]. Patients with HFM require specialized instruction to ameliorate receptive language delays. An increased tendency toward neurodevelopmental problems such as autism, dyslexia, or language delay was also noted, mainly in boys [2].

Psychosocial functioning

We remain in the second phase of the aforementioned study, now incorporating considerations of the emotional side. The results indicate that despite physical differences, parents of children with HFM reported similar levels of behavioral problems and social competences compared to

healthy children. The only exception among parents was a higher average score on the CBCL social problems scale. However, teachers noted a higher frequency of internalizing problems such as anxiety and depression, lower social competencies, and less peer acceptance. Female cases, those with ocular malformations, children of young mothers, and those with additional developmental defects beyond HFM performed worse in teachers' assessments [4].

The third phase of this study took place between 2011 and 2015 when the children were about 13 years old. The aim was to examine behavioral problems using the TRF, YSR, and CBCL. Interestingly, no differences were observed between the study group and the control group on any scales except for disruptive behaviors. Aggression and rule-breaking were less frequently observed or reported by adolescents with HFM. Social functioning differed from children without malformations. Less frequent participation in social activities, fewer friends and less frequent contacts with them, more frequent teasing and rejection by peers were noted. Additionally, according to the researchers, it is difficult to establish a relationship between the extent of facial defects and psychological adjustment [18].

In other studies, significantly lower rates of serious internalizing problems were also observed. Behavioral adjustment was at a comparable level to control groups [19, 20]. Among 35 children between 2 and 3 years old, only one case had a CBCL score outside the norm on the anxiety-depression scale [19]. It is worth mentioning that the child's self-awareness of facial differences in relation to peers most often begins at the age of 3 [3, 21]. Also, in preschoolers, parents reported higher scores on the anxiety-depression subscale, as well as stress problems, anxiety problems, and autism spectrum problems. Regarding the autism subscale, it primarily reflected speech problems. These studies also confirm that more complications occurred in children with extracranial anomalies [20]. However, in the group of 4–11-year-olds, the researchers did not observe an increased risk of internalizing disorders. Researchers stated that there was insufficient evidence for an increased risk of mental disorders in participants. The elevated indicators in the study mainly concerned aspects of peer relationships, such as concentration problems or low academic competences [19].

In Finland, a study was conducted to analyse the specialized healthcare needs of patients with HFM. While the outcome of plastic surgery interventions for 98% of patients may not be surprising, the prevalence of psychiatric care is noteworthy. Nearly 60% of children were given ICD-10 diagnostic codes by child psychiatrists. The most common were Z00.4 - general psychiatric examination, not elsewhere classified - in 31% and Z63.7 - other stressful life events affecting family and household - in 22% of patients. A similar number of diagnoses was

recorded among adolescent and adult psychiatry patients. In patient records, the most common were F40-48 - neurotic, stress-related and somatoform disorders - in 60%, F30-34 and F38-39 - mood (affective) disorders - in 50% of patients. These were the most common codes, ahead of Q67.0 - facial asymmetry - in 43% of patients. Interestingly, the study also determined that the prevalence of HFM patients in Finland was 1:10057 [22].

Problems at school

A study published in 2019 examines speech, language, and communication deficits in adolescents. It highlights the impact of the presence of microtia and mandibular hypoplasia on the outcomes. Group differences were estimated using linear regression analyses with standardized ES adjusted for demographic characteristics or negative binomial regression. Children with HFM scored lower than their peers, with the most significant deviations in patients with mandibular and ear hypoplasia - adjusted ES = -1,15 to -0,18. In addition, there is a difference in the presence of hearing problems - adjusted ES = -0,73 to 0,07 in patients with hearing impairment and adjusted ES = -0,34 to 0,27 in patients without hearing impairment [23].

Regarding learning difficulties, a study involving 142 patients with HFM and 316 in a control group showed that patients with mandibular hypoplasia and microtia, as well as patients with isolated microtia, had lower intelligence test scores. Similarly, they had lower academic achievements compared to the control group. Importantly, the magnitude of differences was small - adjusted ES = -0,2 to -0,04. According to the researchers, there was little evidence that hearing status modified the differences between groups. Twenty-five percent of the control group and 35% of the study group were considered cases having learning problems, more often among boys of Latino origin from bilingual families with lower incomes (below $35,000 per year). Analyses indicated that differential attrition reduced the magnitude of differences between the study and control groups, particularly in areas where cases seemed most vulnerable (vocabulary, reading, and written expression). Furthermore, more than twice as many children with HFM (72%) than controls (33%) received developmental or educational support. Due to the size of the study group, the authors emphasize that it is only a hypothesis that children with microtia and mandibular hypoplasia have a higher risk of learning problems [24]. Supporting arguments for this hypothesis include numerous factors such as hearing impairment, speech difficulties, possible airway obstruction causing OSA [7], multiple surgeries and associated

exposure to anaesthesia [25–28], facial paresis or paralysis [29], and the social-psychological effects of anomalous facial appearance [4, 30].

Craniofacial microsomia: Accelerating Understanding of the Significance and Etiology (CAUSE) is a multicenter observational study designed to assess genetic risk factors for HFM. Data collected via questionnaires between 2018 and 2020 included information on interventions, special education, and bullying. Participants from the USA and Colombia mainly had unilateral defects, microtia, and mandibular hypoplasia. Hearing loss was present in 19%, and 53% used a hearing aid. In the USA, almost half of the participants received special education services, which was rare in South America (only 4%). Teasing was reported in 41% of children, mainly at school, starting on average from 6 years of age. The study suggests that the risk of educational and social problems should be reduced early in childhood - that is, at a time when the child begins to be aware of his or her differences and peer issues begin. It is important to remember the higher incidence of teasing among children with microtia and mandibular hypoplasia, which leads to difficulties in learning and speech. Additional academic support, assessment of psychosocial problems, and providing appropriate resources at an early stage are crucial to improve the quality of life for these children and those close to them [3].

In the United Kingdom and the USA, adolescents and young adults with HFM indicated that bullying was associated with lower self-esteem and feelings of isolation. According to researchers, participants spent considerable time and effort on the contribution of their physical differences to their self-image and identity. These struggles appeared to influence decisions about hiding or revealing their condition through clothing and hairstyle, as well as in the context of surgical reconstruction. Especially during adolescence, support in forming a positive self-image is important. The participants themselves emphasized being more open about their physical differences and accepting themselves. They also admitted to problems with self-esteem. Researchers in this study also noted greater difficulty regarding visual differences among females [31].

DISCUSSION

The described defect, in its complexity, affects not only the external appearance of patients but is also associated with multidimensional challenges. These include psychological, social, and educational development. New findings regarding age and the frequency of teasing among patients may draw attention.

The results presented in this study indicate the diversity of the HFM presentation depending on, among others, the presence of additional malformations, hearing loss, or speech deficits. Similar results concerning neurodevelopment and language development in younger children, and the current differences in older children, can be interpreted in many ways. This may suggest that the primary differences are subtle and only emerge as a result of the accumulation of secondary factors, such as the impact of hearing loss or articulation problems. It may also result from overlapping internalizing complications which affect communication and social interactions. For this reason, pediatricians should be particularly vigilant for signs of developmental delays and quickly refer patients for appropriate specialist consultations. The Finnish study showed that about 6 out of 10 patients had an ICD-10 diagnosis code for a psychiatric unit. The second most common code for psychiatric visits in children was Z63.7 - other stressful life events affecting family and household. [22]. Early interventions can significantly improve the quality of life for children with HFM but also their families.

Despite the valuable findings, the discussed studies have certain limitations. There are very few scientific publications that address the topics presented in this study. It should be noted that most publications are based on small samples, which may affect the generalization of results. An example is the IQ study conducted by Speltz et al., which had a limited sample size, potentially improperly influencing the interpretation of results [23]. Furthermore, methodological diversity and differences in definitions and diagnostic criteria may affect the comparability of results. Discrepancies between studies may arise from differences in methods of determining or defining case samples concerning HFM. For example, Collett et al. excluded children who had unilateral microtia without other features of HFM [2], whereas Speltz et al. included these cases [15]. Large discrepancies in the results will continue until researchers adopt the same diagnostic criteria, e.g. FACIAL or ICHOM [16, 32].

Moreover, the vast majority of studies have taken place in North America. Data concerning children on other continents are fragmentary, which makes it difficult to draw clear conclusions. In the USA, almost half of the children with HFM receive such support, while in South America, this percentage is only 4%. Also, such data highlight the presence of systemic differences in the care of patients with HFM across countries. They can significantly affect long-term educational and social outcomes.

Future research should also analyse in more detail the variability in outcomes depending on gender, and socioeconomic status. Threads of fewer incidents related to rule-breaking or aggression may be continued in subsequent studies. It is also important to examine how genetic

factors and different types of interventions affect outcomes for HFM patients. Conducting research that includes the perspectives of patients and their families will help us better understand their needs and the challenges they face.

CONCLUSION

The accumulated evidence suggests that children with HFM are at greater risk of developing psychological problems, such as anxiety and depression. This is particularly true during early adolescence, when awareness of one's appearance and the importance of social acceptance increase. It is at this time that problems with teasing from peers and exclusion occur. Unfortunately, these issues may intensify as they grow up. Differences in the assessment of behavioral problems between parents and teachers indicate a possible influence of the social context on the perception of these children's difficulties. Parents were more likely to report results within the norm, while teachers were more likely to indicate internalising problems. This is probably due to the greater exposure of children to social situations in the school environment, which highlights their difficulties in interpersonal relationships. The key conclusion is therefore the need for emotional support, especially during adolescence. This is the period when young people confront issues of social acceptance and self-image. It should be emphasized that girls with HFM appear to be more susceptible to emotional problems. Socially imposed expectations on girls regarding aesthetics may increase the pressure they face. In many cases, this affects their self-esteem and social adaptation.

The study also emphasizes paying close attention to the occurrence of additional malformations. Differences in results indicate that patients with a more complex clinical picture require special attention. Although these differences are not always large, their significance increases when considering the higher percentage of children with HFM requiring additional support. The presence of microtia and mandibular hypoplasia, as well as associated problems such as hearing loss, are the main factors increasing the risk of academic challenges. These results confirm the necessity of providing personalized educational assistance and appropriate resources that support the development of children with HFM. Offspring of young mothers and those from families of lower economic status are also more prone to difficulties in cognitive and social development. This indicates the importance of considering these factors in planning support and therapeutic interventions.

Information on aggression and a lower tendency to break rules also seems interesting but requires further research.

The findings highlight the need for early and multifaceted support. This will minimize the negative effects of the condition and improve the quality of life of individuals with the defect, as well as their families. Building social awareness and integrating people with HFM into society can contribute to improving psychosocial well-being and supporting their personal and professional development.

Despite early findings indicating no significant differences in neurological and language development in younger children, long-term studies show difficulties in receptive and expressive language development and academic abilities in older children. Difficulties in subsequent years of life may result both from the defect itself and secondary consequences such as hearing loss.

Authors' Contributions Statement:

Conceptualization: M.P.

Methodology: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Software: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Check: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Formal Analysis: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Investigation: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Resources: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Data Curation: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Writing – Rough Preparation: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Writing – Review and Editing: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Visualization: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Supervision: M.P., P.M., M.M., A.C., A.S., K.S., Z.C.P., A.K., M.W., N.M., A.R.

Project Administration: M.P., M.W., M.M.

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