Correlation of Facial, Dental and Oral Functions of Children with Microcephaly: Clinical Case Report

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ABSTRACT

This paper discusses the orofacial manifestations of children diagnosed with severe microcephaly according to the World Health Organization. Microcephaly is a clinical condition made at or after birth that characterizes a significant reduction in head circumference, which may present craniofacial, neurological, and oral alterations as a result of this malformation. As skull growth is linked to facial growth, can patients with microcephaly have structural and functional impairments? Will these possible changes be expressed before skeletal maturity? These issues are discussed through the presentation of two relevant clinical reports of children diagnosed with severe microcephaly with similar clinical facial and occlusion patterns. Studies along these lines should be conducted with a view to future attempts to minimize possible effects of this malformation on the craniofacial complex with preventive and interceptive therapeutics in these individuals.

Keywords: Children, Dentistry, Microcephaly.

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I. INTRODUCTION

Microcephaly is clinically diagnosed by a reduction in the occipitofrontal head circumference of newborns when compared to other individuals of the same age and sex. Depending on its etiology, it may be associated with structural brain malformations or be secondary to different causes. Most children with microcephaly are characterized by motor and cognitive changes that vary according to their abnormal brain development¹². According to the World Health Organization, head circumference measurements in the first 24-48 hours of life should follow the INTERGROWTH-21st reference ranges and defined the classification for newborns with head circumference -2 and -3 standard deviations below the medium for gestational age and sex as microcephaly and severe microcephaly respectively [1]. In addition to this measurement, other tests are also necessary for the diagnosis of microcephaly, such as transfontanellar ultrasonography, magnetic resonance and/or computed tomography [1]-[3]. The neuropsychomotor involvement can cause vision and hearing disorders, epilepsy and changes in orofacial motricity [2], [3].

The stomatognathic system performs important functions for the physical and biological balance of the human being and depends on the craniofacial growth pattern, the central nervous system and dental occlusion for its functionality. Interferences in some organ of this system lead to changes in its dynamics, such as the classic functions of breathing, sucking and swallowing [4]. The impairment of the craniofacial complex, such as the predominance of vertical growth and the inhibition of mandibular growth, can cause alterations in the stomatognathic system [5]. Studies report that in individuals with no craniofacial malformations, genetic determinism prevails for the final configuration of the face and basal bones, maxilla, and mandible. The facial morphology and the growth of the maxilla and mandible, as well as the time when the increments of this growth are manifested, are largely determined by the genome but modified to a lesser degree by environmental components [5], [6]. But what about the child with microcephaly? Is the development of orofacial structures determined by genetics or by local factors such as the presence of reduced head circumference? As skull growth is linked to facial growth, can patients with microcephaly have structural and functional impairments? Future studies may clarify these issues.

Regarding the dental occlusion pattern, maxillary discrepancies can negatively interfere in the orofacial muscles with functional, esthetic, and psychosocial alterations [7]. Recent studies on oral manifestations in children with microcephaly show micrognathia, delayed tooth eruption, dysphagia, dental trauma, bruxism and malocclusion [8], [9]. In this context, the analysis of facial, dental, and skeletal patterns is an important tool for professionals in the field of health and can identify possible alterations in the speech, swallowing and chewing of individuals with microcephaly.

Next, two clinical cases of children with primary dentition, diagnosed with severe microcephaly with similar facial pattern behaviors, relation between dental arches and oral functions, will be clinically described.

II. CLINICAL CASES

Two clinical reports presented below were treated in the Dentistry Faculty of the State University of Southwest Bahia, Brazil, with follow-up by their respective parents. It was necessary to resort to behavioral assertive techniques in order to establish the process of children's adaptation to the dental procedures. For the physical examination, the patient was positioned on the parents' lap in the dental chair, aiming to establish a comfortable and inclined position for better swallowing capacity and stability of movements, in addition to helping to facilitate the conduct of the procedures. In dental care sessions, no sedation was used.

A. Clinical Case 1

Male patient with 4.1 years of age had a medical diagnosis of severe microcephaly without associated syndromes. His medical history included epilepsy, cognitive and neuromuscular disabilities. The father related difficulty of oral hygiene and teeth grinding during the day and at night. In the clinical morphological analysis of the face, frontal view, a narrow and flat forehead was observed, showing the disproportion of the upper facial third in relation to the other thirds. In midface, can observed the absence of zygomatic projection while the lower third is characterized by excessive lip space with lingual interposition between the anterior teeth. Oral breathing was predominantly. It was noted in the sagittal view of the face, nasolabial angle closed and mandibular rotation, showing a possible predisposition to vertical face growth, evidenced by the increase in the lower third of the face in relation to the others (Fig. 1 A-B). Intraoral clinical examination showed the child in the primary dentition with the presence of primate spaces, type I Baume dental arch, with absence of the deciduous upper left canine and deciduous left lower lateral incisor, with no report of extraction. The presence of biofilm, unilateral left chewing, and wear on the occlusal surfaces of the molars and the incisal surfaces of the deciduous upper canines were observed. Occlusal analysis revealed maxillary dental arch constriction and anterior open bite with atypical lingual interposition (Fig. 1 C-E). Due several teeth abrasion, the proposed treatment plan was the oral prophylaxis and application of sealant and fluoride varnish. In addition, the child's father received educational guidance on oral hygiene. The dental eruption, the development of occlusion, as well as the craniofacial growth of this patient will be monitored during the skeletal maturity. According to the patient's psycho-emotional maturity, the installation of a mobile myorelaxant bite plate will be planned to minimize teeth grinding. A report was sent to the speech therapist about the unilateral chewing and the hypotonicity of the orofacial muscles.

Fig. 1. Frontal and lateral extraoral photographs of a child, 4.1 years of age, with severe microcephaly. Predominance of vertical facial dimensions and convex facial profile are observed. Note the absence of passive lip sealing (A-B). Intraoral photographs reveal maxilla atresis associated with anterior open bite (C-E).

B. Clinical Case 2

A 3.6 years old female patient with severe microcephaly without associated syndromes showed medical history included cognitive and neuromuscular deficiency, epilepsy, and visual alterations. The mother related excess of saliva and teeth grinding day and nighttime of her daughter. In the facial analysis, the disproportion between facial thirds were observed. It was also noted the absence of passive lip sealing and atypical lingual interposition with predominantly oral breathing. The lateral facial profile showed disharmony in the sagittal relationship between the apical bases, with a positive sagittal step between the maxilla and the mandible, resulting in an excess of facial convexity (Fig. 2 A-B). Intraoral clinical examination revealed the patient in primary dentition with wear on the occlusal surfaces of deciduous molars due to the tooth grinding, as well as unilateral chewing on the left side. The analysis of the occlusion showed anterior open bite up to the deciduous first molars associated of maxillary dental arch constriction (Fig. 2 C-E). The proposed treatment plan was oral prophylaxis and application of sealant and fluoride varnish on the occlusal surfaces of primary molars. In addition, the mother for the child received instructions to stimulate her regarding the habit of brushing and holding the toothbrush. A report was sent to the speech therapist on unilateral chewing and the hypotonicity of the orofacial muscles. The dental eruption, the development of occlusion, as well as the craniofacial growth of this patient will be monitored. According to the patient's psycho-emotional maturity, the installation of a mobile myorelaxant bite plate will be planned to minimize teeth grinding.

Fig. 2. Frontal and lateral extraoral photographs of a patient with severe microcephaly in the complete deciduous dentition. Note the vertical dimensions of the face prevail over the horizontal and an excess of facial convexity was observed (A-B). Intraoral photographs reveal open bite with atypical lingual interposition (C-E).

III. DISCUSSION

Due to the impairment of psychomotor development, often present in individuals with microcephaly, intellectual and physical disabilities may occur and require differentiated and individualized care, in addition to assistance and supervision in carrying out activities such as toothbrushing. Biofilm prevention and control measures are very important, as they present a high risk of developing caries due to the difficulty in cleaning the teeth and consuming a diet rich in sugar in most cases. A recent study evidenced the importance of information on supervised brushing techniques aimed at parents and caregivers, dietary guidance and topical application of fluoride in order to minimize damage and improve oral health [10]. Instructing parents on how to perform hygiene and teaching how to make mouth openers with wooden spatulas can facilitate brushing and positively impact the improvement of oral health of children with microcephaly [11]. A preventive and educational dental approaches were carried out in this clinical case report corroborating findings in the literature.

The chronology of tooth eruption depends on multiple factors such as genetic, biology, and nutrition during the development of the primary dentition until reaching its functional position of occlusion with its antagonist [12]. Some studies demonstrate that babies with microcephaly have a significant and generalized delay in tooth eruption, suggesting compatibility with the delay in the general development of these children [9], [13]. In the present study, the second clinical case was diagnosed with complete primary dentition within the normal range. However, the first case, 49 months of age, presented an absence of deciduous upper canine teeth and a lower lateral incisor on the left side,

with no report of extraction. It is important to point out that it was not possible to perform an imaging exam, making it impossible to make a differential diagnosis of tooth agenesis.

As for craniofacial growth, it is possible to observe an interconnection between the bones of the anterior region of the face, maxillary bones, and surrounding muscle tissue, with the growth of both the skull base and the mandible, which occurs continuously, starting in the prenatal and remaining in the postnatal period, governed by genetic and environmental factors [5], [6]. During primary dentition, the face has not reached its definitive dimensions yet. The maxilla and mandible did not reach the final dimension and will show considerable growth until skeletal maturity. However, the tendency is for the facial pattern to remain constant during this process, which is why, at this stage, the facial configuration must be identified, defining the individual's facial type [14]. Taking into account that the patients described in this study present a significant occipitofrontal reduction and both show in the morphological analysis a vertical facial skeletal configuration in the primary dentition stage, a central issue that motivated this study is seen. Is there a correlation between facial growth patterns and head circumference reduction? The literature is not yet conclusive regarding the relationship between brain growth and facial growth.

Marques *et al.* [13] analyzed the oral functional characteristics of 26 children aged 12, 15 and 18 months, diagnosed with microcephaly at term birth, without any association with disorders or congenital malformation. The patients showed changes in facial muscle tone, breathing and swallowing. In view of the analysis carried out in the two clinical cases presented in this study, since the children were diagnosed with severe microcephaly at birth, with cognitive and neuromuscular deficit, it is possible to establish the impairment of the orofacial muscles and oral functions. It should be emphasized that the presence of muscle hypotonia in these patients causes difficulties in carrying out oral hygiene habits, since desensitization occurs in neuromotor development [14]. The performance of daily care by the person responsible for the child with microcephaly under the supervision and guidance of qualified professionals, in addition to promoting oral health, also provides a favorable stimulus to the stomatognathic system.

In terms of parafunctional activity observed in the two children in this case report, teeth grinding was present in day and night periods with significant occlusal wear on the posterior teeth evidenced by the flat surfaces of the occlusal faces. Cota *et al.* [15] demonstrate that this deleterious habit, with multifactorial origin, is present in individuals with microcephaly, due to the relationship between the action of grinding teeth and neurological damage. Bruxism may bring about negative consequences for the stomatognathic system, such as pain, excessive tension in the masticatory muscles and occlusal wear in the posterior teeth and in the incisal edges of the anterior teeth [16], [15].

IV. CONCLUSION

Based on the increase in the number of cases of microcephaly in Brazil and grounded on scientific concepts that the facial pattern is maintained during the growth, it

stands out that studies should be conducted with a view to future attempts to morphologically understand the face and dental occlusion of children with microcephaly, once it will significantly contribute for health professionals to adequately perform preventive and interceptive measures in these individuals. Therefore, key points will be analyzed to define a realistic prognosis and the consequent development of a health care protocol in order to promote functional, esthetic and psychosocial improvements.

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