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Ulectomy on infant patient with microcephaly: a report case

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RESUMO

A microcefalia é uma condição neurológica rara, resultante do fechamento prematuro das fontanelas cranianas. Sua etiologia inclui fatores genéticos, ambientais e infecciosos, a exemplo o Zika vírus, que pode culminar em diversas desordens de desenvolvimento. O objetivo deste trabalho é relatar um caso clínico de atraso de erupção dentária em paciente portador de microcefalia associada à exposição fetal ao Zika vírus, bem como discutir os aspectos inerentes ao diagnóstico e tratamento do caso. Uma pesquisa bibliográfica foi realizada através das seguintes bases de dados: Scielo, PubMed, ScienceDirect, Biblioteca Virtual da Saúde (BVS) e LILACS e vinte e três artigos correlacionados com o objetivo do atual trabalho foram selecionados. Observou-se, com base na literatura e caso em questão, que o atraso de erupção é uma alteração frequentemente identificada em pacientes que sofreram exposição placentária ao Zika vírus e pode estar relacionada a má postura lingual ou falha dos mecanismos responsáveis pelo processo eruptivo. O diagnóstico dessa desordem deve ser realizado de forma precoce por meio do exame clínico e imaginológico. O seu tratamento consiste na incisão e exérese do tecido que recobre a unidade dentária em questão, procedimento denominado de ulectomia. Esse, é de simples execução, baixo custo e apresenta resultados eficazes no tratamento desse distúrbio.

Palavras chave: Anormalidades dentárias. Erupção dentária. Microcefalia. Zika vírus.

ABSTRACT

The microcephaly is a rare neurological condition, resulting from the premature closure of the cranial fontanelles. Its etiology includes genetic, environmental, and infectious factors, for example the Zika virus, which can culminate in several developmental disorders. This work aims to report a clinical case of dental eruption delay in a patient with microcephaly associated to fetal exposition to Zika virus and to discuss inherent aspects to diagnosis and treatment of the case. A bibliographic search was performed through the follow date bases: Scielo, PubMed, ScienceDirect, Biblioteca Virtual da Saúde (BVS) and LILACS and twenty-two articles correlated with this work aim were selected. It was observed that the eruption delay is a commonly observed alteration on patients that had placental exposure to the Zika virus and can be related to bad lingual posture or failure of the mechanisms responsible by the eruptive process. This disorder diagnosis must be performed early through clinical and image examinations. Its treatment consists of the incision and exeresis of the tissue that covers the tooth, a procedure denominated ulectomy. That being a simply executed, low cost and effective treatment for this disorder.

Keywords: Zika virus, microcephaly, dental abnormalities, dental erupti

1. INTRODUÇÃO

According to the World Health Organization (WHO), microcephaly is characterized by the cephalic perimeter (CP) shorter than two or higher diversions on the reference pattern for age, gestational age or the child's sex [1]. Its etiology includes risk factors exposure, genetic causes and, recently, it was observed that the infection by Zika virus (ZIKV) can result in the aforementioned disorder [2].

In March 2015 the first tests for the Zika virus resulted positive, registered initially at the Northeast region of Brazil [4]. In 2015 it was observed that the microcephaly was related to the infection by Zika virus during the gestational period [5]. The Ministry of Health registered in 2016 9.770 cases of microcephaly in the country. Among those, 2.334 were associated with the infection by Zika virus [6].

The microcephaly promotes a disorder in the baby's neurological system, culminating in the premature closure of the cranial fontanel and in the reduction of the brain perimeter [6]. Likewise, children with microcephaly present neuropsychomotor alterations which may result in epilepsy, cerebral palsy, slow cognitive, motor and diction development, as well as hearing and sight

problems. By virtue, some authors suggest the term: "Zika virus congenital syndrome" (ZCS) [5,7]. Beyond the systemic manifestations, individuals affected by ZCS may present oral and craniofacial manifestations [8,9].

The eruptive delay is a clinical finding frequently identified in microcephalic patients. It is defined as the dental unit clinical absence after the development of two thirds of the root. Its cause can be local or resultant of the pathological processes' presence [10]. Recent studies prove that microcephaly associated to the ZIKV may interfere in the dental eruption's physiological process [5,9,11].

The late eruption diagnostic is held by clinical and radiographic examination. In the presence of this chronological alteration, an ulectomy is indicated as a way of treatment [12]. This procedure consists in the resection of the tissues that cover the occlusal face of a deciduous tooth, which has not erupted yet. This method is of a simple technique, low cost and it is made under local anesthesia [12,13].

Therefore, having in sight the increasing of children with ZCS in Brazil, the study aims to relate a clinical case of deciduous dental eruption delay in a microcephalic patient, as well as discuss the inherent aspects to the diagnostic and treatment.

2. CLINICAL CASE REPORT

Male patient, 35 months of age, leucoderma, attended to the Tiradentes University's (UNIT) dental clinic alongside his guardian. She related as a principal complaint "a tooth that did not come up" SIC. During the anamnesis it was obtained the information that the patient's genitor was infected by the Zika virus during the pregnancy's first trimester, resulting in the unborn baby's neurological disorder, named microcephaly. The diagnosis was made at birth, based on the gestational record and the reduced cephalic perimeter of the baby, equivalent to 28,5 cm. Beyond that, misophonia and diction and sight alterations were related, characterizing the ZCS. Due to his neuropsychomotor and phonetic limitations, the patient receives continuous medical and phonoaudiological monitoring.

On the physical exam, it was possible to observe alterations such as: unproportionally big auricular and facial regions, mandibular micrognathia, cap and front of a reduced size and posteriorly projected (Figure 1-a). At the intraoral exam, it was observed the clinical absence of the unit 82 and a volume increase with a pale coloring of the gingival mucosa, at the region of the dental element aforementioned (Figure 1-b), besides an incisal/occlusal abrasion resultant of bruxism and nocturnal clenching. Furthermore, the guardian explained that the baby bites the soft tissues of the oral cavity and she uses bandages as a blocking method of the damaging habits.

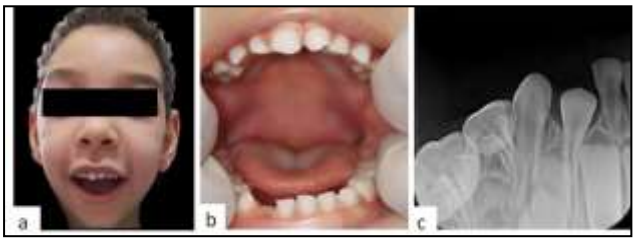

Figure 1- (a) Extra-oral Image; (b) Clinical absence of the unit 82; (c) Periapical radiography evincing unit 82.

On the image exam (periapical radiography) it could be observed suggestive signals of the element 82 in a submucous position, without impaction, with a partially complete root and an open apex, designating Nolla's stage 9. Therefore, it was concluded that the dental unit was retained only by gingival tissue. Regarding this, it was proposed an ulectomy as a treatment plan, expecting the gingival tissue resection and incisal side exposure of the previously referred unit, allowing its spontaneous eruption.

The surgical procedure was initiated after the intra and extra oral antisepsis with chlorhexidine digluconate 0,12% and 2% respectively, under local anesthesia with topical Benzocaine (Benzocaíne®) and lidocaine 2% infiltration associated to adrenaline 1:100.000 (Figure 2-b) at the gingival mucous region regarding the unit in question.

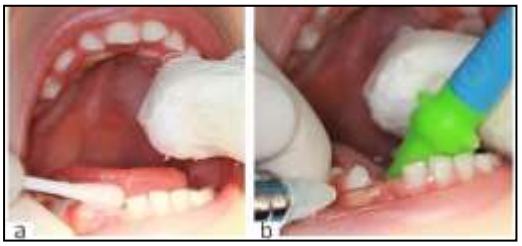

Figure 2- (a) Mucous topical anesthesia; (b) Gingival infiltrative anesthesia.

Following this, the diaeresis process was initiated by an elliptical incision of the hood that covered the dental element, with assistance of a scalpel and a 15° blade (Figure 3-a), proceeding with the tissue's divulsion and resection through a Seldin straight lever, exposing the unit 82's incisal edge (Figure 3-b,c e d).

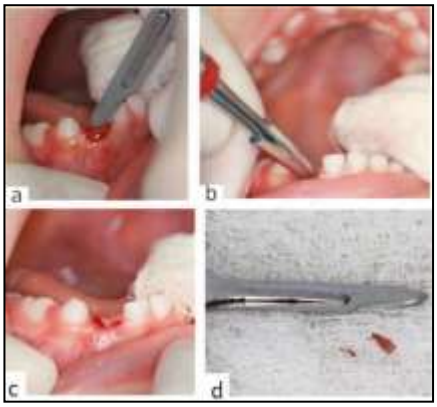

Figure 3 - (a) Gingival hood incision; (b) Tissue divulsion; (c) Exposure of the incisal edge of the unit 82; (d) Removed tissue fragment.

After this, it was conducted the irrigation with saline solution at 0,9% and hemostasis by intermedium of the digital compression and sterile gauze, without the need of synthesis. At the 15-day post-surgery, the spontaneous eruption process and a satisfactory evolution without complications were observed. After 24 months, it was noticed the partially erupted unit in the oral cavity, evidencing the effective result of the procedure on the functional and aesthetic reestablishment and in the prevention of other oral anomalies (Figure 4).

Figure 4 - 24 month post-operative

3. DISCUSSION

Microcephaly is a rare neurological condition, resultant of the cranial fontanelles' premature closing. This alteration, culminates in the brain structures' compression and in the incomplete development of the encephalic mass [14]. Prado (2019) [4] reported that the standard measure of CP to newborns with microcephaly corresponds to approximately 29,12 to 31,52 cm to the male sex, and from 28,85 to 30,99 cm to the female sex. According to Leite (2016) [6], grave microcephaly corresponds to three deviations below the CP rate. In this account the patient in question presented 28,5 cm of cephalic perimeter, an inferior value to the established one by the literature for the male sex, being then diagnosed by the geneticist with serious microcephaly.

Microcephaly possesses a multifactorial etiology, covering genetic and environmental factors, during the pregnancy or right after the birth [15]. Melo (2019) [14], classified gestational microcephaly as primary or secondary. On the primary one, the malformation is a result of genetic hereditary flaws, in which a copy of the defective recessive autosomal gene of the baby's family is inherited. On the secondary one, or by craniosynostosis, the causal factors include alcohol, drug or medication use, placental malformation, maternal non-controlled phenylketonuria, traumatic brain injury, mother's malnutrition, radiation exposure, lack of O2 in the tissues and in the blood during childbirth, infectious diseases, for example Zika, and mercury intoxication. Besides the pre-natal factors, the literature evidences that elements in the postpartum can result in this disorder, such as disruptive traumas, intracranial infections, congenital encephalopathy by HIV and the contact with toxins [15].

The average microcephaly occurrences in Brazil was of 2 cases in each 100.000 alive. Nevertheless, since 2015, there was a sudden increase on the index of born children with this amendment. In which the prevalence was increased to 54,6 infected in each 100.000 live births [2]. According to Marinho (2016) [16], the female sex represents 65 of the cases per 100 thousand habitants, transcending the male sex (43,8 per 100 mil habitants). Consequently, in front of the elevated number of registered cases, especially in the state of Pernambuco which presented a higher percentage, the Ministry of Health declared a nationally important public health emergency. In Sergipe there were 27,62 cases confirmed per 10 thousand live births, becoming one of the states with the highest number of records in Brazilian territory [3].

In November of 2015 the Ministry of Health identified the association between the microcephaly increase in the country, and the infection by Zika virus in the gestational period (CABRAL et al., 2017). This virus belongs to the Flaviridae family and it is classified as a flavivirus. Its transmission occurs through the Aedes Aegypti mosquito bite, thus, can be considered an arbo-

virus [7].

The Zika virus can be detected in the placenta's amniotic liquid of an infected woman and in the blood of newborns. This process is resulting of the capacity of trespassing the placental barrier that the virus presents, causing brain deformities of a congenital nature in the newborn, encompassing microcephaly [3,7]. On the current report, the patient's mother was infected by Zika virus at her 1º trimester of pregnancy. Nonetheless, the child was diagnosed with ZCS only after birth.

The microcephaly carrier patients usually express craniofacial deformities due to the disorder between the cranial and facial growing. The deriving effects are unproportionally large ears and face, head and forehead of a reduced size and posteriorly projected, detached and wrinkled scalp. Microcephaly interferes mainly in the child's cognitive development and may result in alterations such as hyperactivity, dwarfism, epilepsy, brain paralysis, hearing problems and visual, facial, stomatognathic and behavioral alterations [7,14].

Repercussions as deciduous tooth eruption delay, alterations on the irruption sequence, muscular hypotonia or hypertonia are commonly identified on patients affected by ZCS [7,8]. Supporting this, the research made by Carvalho (2019)[5] evidenced that 60% of the 30 chidren analyzed in her study presented reduced lip and tongue breaks, as well as eruption delay (60%), narrow palate (33,3%), bad lingual posture (30%), dental number and/or form anomaly (13,3%), mandibular hypoplasia (6,7%), eruption sequence alterations (13,3%), muscular spasms (56,7%), convulsions (30%) and hearing and/or sight disarrays (16,7%). In conformance with Delgado (2017) [17], beyond these characteristics, children with microcephaly may also present periodontal diseases, dental traumas, bruxism and dysphagia. On the described case and in agreement with the literature, characteristics such as hearing and sight alterations, bruxism and auricular, facial, head and forehead region disproportions were observed.

Dental eruption is defined as the physiological process of the intraosseous element movement towards the oral cavity [18,19]. According to D'Agostino (2019) [9], deciduous teeth erupt between the 6º and 30º months of life, tending to follow a chronological physiological order: central incisors, lateral incisors, first molars, canines and second molars. The eruptive delay is characterized as an alteration of this process. Radiographically is possible to notice root formation of 75% with a clinical absence of the crown [10,19]. This anomaly is constantly related in children with microcephaly and can be associated to a poor lingual posture [5,9,11]. Studies reveal that an inadequate lingual posture is 7,3 times more incident in patients with late dental eruption when compared to those who did not present this disorder. Another possible cause of the eruption delay is the infection of the neural progenitor cells by ZIKA. These cells are responsible for the oral cavity and facial structures' formation. Its commitment may result in microcephaly and in the failure of the mechanisms responsible for the dentition development and for the eruptive process [5,20].

In accord with the current case, the study held by D" Agostino (2020) [9], evidenced that 52,70% of the analyzed patients presented an eruptive delay, with greater rate on unit 82, followed by 72, 52 and 62. In eruption delays that surpass 12 months, a surgical intervention must be established as a treatment method. The ulectomy consists in the excision and removal of the bone and/or gingival tissues which covers the non-erupted dental element occlusal surface, making its path and emergence easier in the oral cavity [10,13,18].

On the described report, it was observed a clinical absence of the unit 82 and the formation of 2/3 of the root radiographically, corresponding to Nolla's stage 9. Based on clinical and imaging signs and the patient's age, it was possible to determine the 22-month eruptive delay. Therefore, the surgical exposure was held intending to easy the spontaneous eruption of the unit in question until it fulfilled its functional position in the oral cavity [12].

Beyond this, current Brazilian literature, in compliance with the global, highlights that the special patients with neuropsychomotor limitations and debilitating characteristics need a continuous dental monitoring. Until this moment, no standard care was postulated and the handling of children with ZCS is highly discussed [15,21].

However, short appointments, thorough anamnesis with records of the administered medication by the patient and contention methods (when needed) are recommended, under the guardian's consent. Besides that, it is of a great importance the bacterial plaque control promotion, due to the higher susceptibility on the development of dental caries and periodontal diseases, resulting of the continuous use of medications that interfere in the saliva flow and the absence of motor coordination in the oral hygiene process, where the use of electrical dental brushes is recommended [6,11,22].

Therefore, it is of a great importance that the patients affected by ZCS are supported by a multidisciplinary health team, aiming the early identification of existing anomalies, as well as the postulation of the needed treatment [8].

4. CONCLUSION

The Zika vírus congenital syndrome has as consequences diverse systemic and stomatognathic manifestations, between those, craniofacial deformities such as microcephaly and eruptive disorders. The eruption delay is frequently reported in patients with this syndrome and may be associated to an inadequate lingual position or a failure of the eruptive process' responsible mechanisms. It is indispensable that the early diagnosis is undertaken by clinical and imaging tests and the adequate treatment is established.

The literature, in concert with the current case, emphasizes the ulectomy as a simple surgical procedure, of a low cost and with effective results in the prevention of functional and aesthetical disorders, helping in the spontaneous eruption process of dental units with eruptive delay.

Until the current moment, there is no care standardization for patients with microcephaly, making necessary studies which aim this aspect, as well as the dentistry professionals' capacitation in the care for patients with neuropsychomotor limitations.

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