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Oblique facial cleft, from an odontological point of view

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Abstract
Introduction: Craniofacial clefts are the result of disruption of the normal developmental program during embryogenesis affecting the face, skull or both.
Objective: To analyze the literature on oblique facial clefting, highlighting the different subtopics such as its etiology and prevalence, clinical manifestations, diagnosis, treatment and dental management.
Methodology: Articles published in databases such as PubMed, SCOPUS and Google Scholar were analyzed, with emphasis on the last 5 years. This was done using keywords such as "oblique facial cleft", with the subtopics "etiology", "oral management", "diagnosis", "epidemiology" and "treatment".
Results: The oblique facial cleft is one of the rarest forms, its incidence is believed to be in the range of 0.24% of all facial clefts. Its etiology is embryologically a failure of the lateral and maxillary process along the naso-optic sulcus. Its clinical manifestations are very variable, but a cleft can be found originating from the Cupid's bow and can involve the alveolus between the lateral and canine, passing laterally from the medial piriform opening to the infraorbital foramen and ending in the lower orbit. Diagnosis is made in the 2nd and 3rd trimester, based on sagittal, coronal and axial views of the fetus. Treatment is based on the protection of vision and corneas, in addition to creating an adequate rehabilitation.
Conclusion: As it is a manifestation that involves the oral region, a dental team is required to complete its general treatment.

Keywords: Oblique Facial Cleft", "Etiology", "Oral management", "Diagnosis", "Epidemiology" and "Treatment

1. Introduction
Craniofacial clefts are the result of disruption of the normal developmental program during embryogenesis affecting the face, skull or both [1]. Oblique facial clefts are among the rarest clefts and few cases have been reported in the literature [2]. These are defined as clefts extending between the nasolateral and maxillary prominences combined with the cleft lip include types 2-6 of the Tessier classification [3, 38, 39, 40]. Most of these arise sporadically and are attributed to a failure of fusion between the medial nasal prominence and the maxillary and lateral nasal processes [4]. They are in the paramedial line of the facial structures, especially in the orbit [5], extending from the nasal ala to the medial canthus, variably affecting the skin, soft tissues, cartilage, bone and mucosa [6]. Generally, there is severe asymmetry and multiple facial areas are affected, with the passage of time the deformities become more obvious and a clear three-dimensional underdevelopment of hard and soft tissues results [7]. Clefts vary in severity and have individual differences, this must be measured, as well as their distribution to correct them surgically [8]. There is some controversy in the treatment options and timing, the priorities in these cases include airway patency, feeding and corneal protection [9].
Therefore, we must be very clear about what this malformation consists of and what it entails, as well as its causes and phenotypic characteristics, to be alert and be able to make a timely diagnosis in patients so that they can be treated appropriately. In the dental area, its importance lies in the affection of the alveolar bone, dental treatments should meet the same objectives as in any other patient, but we must have certain considerations and work in a multidisciplinary

manner with a well stipulated treatment plan. The objective of this review was to analyze the literature on oblique facial cleft, highlighting the different subtopics such as etiology, epidemiology, diagnosis, treatment, and dental management.

2. Materials and methods

Information from articles published in PubMed, Science Direct and EBSCO was analyzed with emphasis on the last 5 years. The quality of the articles was analyzed based on the PRISMA guidelines, i.e., identification, review, choice, and inclusion. The quality of the review was assessed using the measurement instrument for evaluating systemic reviews (AMSTAR-2) [10]. The search was performed using Boolean logical operators AND, OR and NOT. It was realized with the words "oblique facial cleft", "etiology", "epidemiology", "diagnosis", "treatment", and "dental management", in conjunction with logical Boolean operators Or y And.

3. Results & Discussion

3.1 Etiology and Prevalence

Craniofacial clefts have a prevalence of 1.43 to 4.85 per 100,000 births [9, 11, 12]. Less frequently occurring in an oblique manner, therefore, it is the rarest form, its incidence is not estimated, but is believed to be in the range of 0.24% of all facial clefts [13, 14]. Its etiologic factors are not clear, it usually has no familial tendency, syndromic association or gender predominance. Embryologically it is a failure of the lateral and maxillary process, along the naso-optic sulcus [15]. The loss of some elements in craniofacial bone formation, by virtue of its downward cascading nature causes failure of bone formation and fusion, creating a large defect [16].

Its cause is mainly attributed to a primary developmental arrest, neurovascular insufficiency or tears in the developing maxillary process, recent studies suggest that they are caused by a combination of amniotic band migration and increased local pressure that produces cellular ischemia [14]. On the other hand, studies also report that injury or rupture of the stapedial artery early in embryogenesis has been postulated as a potential cause [17]. Genetic studies have revealed that disruption in the SPECCIL protein results in oblique facial clefts, demonstrating that these have a genetic basis [18]. This protein plays an important role in adherens junctions involved in cell adhesion, actin cytoskeleton organization, microtubule stabilization, and bone organization and cytokinesis. Thus, it controls the delamination of cranial neural crest cells during facial morphogenesis [19].

Although the prevalence of the oblique facial cleft is not yet estimated, it is believed to be found in 0.24% of all facial clefts. The etiology is clearly attributed to embryologic failure, as there is a failure of closure of the lateral and maxillary processes.

3.2 Clinical Manifestations

It has a wide clinical presentation [15, 20]; in its phenotype the cleft runs vertically through the lacrimal portion of the lower eyelid, lateral to the lacrimal puncta, through the infraorbital rim and orbital floor, medial to the infraorbital nerve, through the maxillary sinus and cheek [21]. It originates from the Cupid's bow and may involve the alveolus between the lateral and canine, passes laterally from the medial piriform aperture to the infraorbital foramen, the cleft terminates in the lower orbit. Soft tissue and skeletal defects may be observed, in addition the lacrimal apparatus may be involved, the inferior lacrimal canaliculus is usually hypoplastic or absent; the cleft may end in an association of coloboma and the orbital contents may prolapse into the maxilla, in addition to presenting microphthalmos or anophthalmos [20]. In the alveolus, the cleft is in the usual position for a complete cleft palate [21]. Speaking of patients' self-esteem, it has been reported that in addition to facial deformities, patients may present psychological, social and physical stress [17]. Because the face is the primary means by which human beings interact with each other and is the main method of emotional expression and social interaction [22].

Its manifestations are very broad, we can observe a cleft that originates from the Cupid's bow to end in the lower orbit, involving soft tissue and skeletal defects; psychosocial factors are important in these patients by influencing their self-esteem.

3.3 Diagnosis

It has been reported that clefts are usually diagnosed during the second and third trimester of pregnancy, based on sagittal, coronal and axial views of the fetal head and face [23]. Accurate prenatal diagnosis of these is critical to establish long-term treatment planning, prognostic prediction and parental counseling [24]. Imaging and assessment of the deformity play an important role [25]; transabdominal ultrasound represents the first approach [26]. An accurate assessment of these malformations is often seen on ultrasound performed during pregnancy but the accuracy of this depends on the experience of the sonologist, the fetal position, amount of amniotic fluid and the type of cleft we are dealing with [27]. Three-dimensional ultrasound and prenatal MRI improve diagnostic accuracy by providing a more accurate picture of the defect [28]. All this requires a multidisciplinary team involving different areas such as genetics, obstetrics, neonatology, pediatrics, radiology, maxillofacial surgery and medical ethics, in order to proceed with the prognosis [26]. Despite being present before birth, this team continues until the patient reaches skeletal maturity [29].

The diagnosis of these manifestations can be made in the second and third trimester of pregnancy with the help of imaging by the physician, it is important to have a multidisciplinary team from the moment these manifestations are detected in order to issue a prognosis and advise the parents.

3.4 Treatment

Facial clefts are a challenge for plastic surgeons because they can involve soft tissues or facial bones, even both [20]. Their repair is anatomical in all cases [17]; an important step in their treatment would be a better understanding of their genetic pathogenesis [18]. The main objective of its rehabilitation is to protect the vision or corneas; the soft tissue component can be treated in the first month of life; for patients with more accentuated deformities, each facial component should be evaluated in order to restore and give a more natural appearance [13]. Their repair is usually complicated, with more complex surgeries; multiple surgical procedures are required in early infancy, along with lifelong multidisciplinary treatment [11, 18]. Minimally invasive techniques are now included, with minimal incisions to achieve a natural appearance [30]; although this may involve maxillary bone grafting, cheek flaps, rhinoplasty and transcranial correction of orbital dystopia [11], in addition to the use of tissue expanders, along with autologous soft tissue repair and bone grafting [31].

The treatment represents a challenge for surgeons due to the amount of tissue involved; its goal is vision or corneal

protection. Minimally invasive surgery with the aid of bone grafting is currently used to also give a more natural and esthetic appearance.

3.5 Dental Management

In addition to soft tissue and bone abnormalities, the alveolar and palatal regions may be affected, generating a cleft-related maxillary discontinuity with asymmetry and disproportion [32]; collapse of these maxillary elements is frequently observed and demonstrates narrowed and recessed dental arches [33]. For improved esthetics and function, cleft management includes maxillary plastic surgery, which can have complications such as marginal bone destruction, root resorption and lesions in dental organs [34]. To treat such root resorption, endodontic therapy followed by internal dressing with calcium hydroxide is recommended to stop resorption [12]. Calcium hydroxide increases dentin pH and inhibits hydrolase activity while activating alkaline phosphatases [35]. In addition to the above, these patients face risk factors such as dental caries, enamel hypoplasia and structural abnormalities [36]. Another situation that may be present is a diminution of the oral vestibule leading to altered occlusion and articulation along with difficulty in brushing and flossing [29]. The desire to improve facial esthetics is one of the reasons why dental treatment is sought by an orthodontist and a maxillofacial surgeon, depending on the case [37].

When oral regions are affected, a maxillofacial surgeon is involved in multidisciplinary care for the correction of maxillary collapse, in addition to other specialists such as orthodontists for dental alignment and endodontists in cases where there is root resorption.

4. Conclusions

The oblique facial cleft is a rare manifestation with a percentage of 0.24 of all facial clefts, its etiology is a failure in the closure of the lateral and maxillary processes, it is characterized by a cleft that goes from the cupid's bow to the lower orbit with possible involvement of the palate. It is diagnosed using imaging techniques such as three-dimensional ultrasound and prenatal magnetic resonance imaging. Its treatment is complicated and involves a multidisciplinary team for the multiple surgeries it requires. As it is a manifestation that involves the oral region, a dental team is required to complete its treatment.

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