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Epidermal cyst: a rare case report in a 5-month-old child

Cisto epidérmico: relato de caso raro em criança de 5 meses

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ABSTRACT

The epidermal cyst is a rare, benign lesion, usually asymptomatic and slowly enlarging, more frequently located in the submandibular region. Its etiology is believed to be associated with the epithelial residual tissues retained in the midline during the closure of the first and second brachial arches, in the third and fourth weeks of intra-uterine life. Its clinical characteristics are of a rubbery consistency to palpation, well defined borders, with the absence of skin appendages within their squamous epithelium lined walls. The final diagnosis is confirmed through histopathological examination with stratified squamous epithelium findings in the cystic fluid lumen or keratin. The treatment is usually surgical and without recurrence and the final diagnosis is confirmed through histopathological examination. This paper aims to present a case report of a congenital epidermal cyst in a 5-month-old female patient located in the mouth floor near the sublingual gland duct. The case was treated with excisional biopsy and after follow-up, there was no recurrence.

Indexing terms: Surgery oral. Epidermal cyst. Nonodontogenic cysts. Diagnosis, oral. Pathology, oral.

RESUMO

O cisto epidérmico é um cisto de desenvolvimento, benigno, raro, localizado com maior frequência na região submandibular, geralmente assintomático e de evolução lenta. Acredita-se que a sua etiologia esteja associada aos restos epiteliais retidos na linha média durante o fechamento do primeiro e segundo arcos braquiais, na terceira ou quarta semana de vida intrauterina. Suas características clínicas são de uma consistência borrachóide à palpação, bordas delimitadas e sem apêndices de pele no seu interior. O diagnóstico é confirmado através do exame histopatológico com achados de epitélio escamoso estratificado e no lúmen fluído cístico ou queratina. O tratamento é geralmente cirúrgico e sem recidivas. Este trabalho relata um caso de cisto epidérmico congênito de uma paciente pediátrica, do sexo feminino, de cinco meses de idade e localizado no assoalho de boca próximo ao ducto da glândula sublingual. O caso foi tratado com a enucleação cirúrgica e em seis meses de acompanhamento não apresentou recidiva.

Termos de indexação: *Cirurgia oral. Cisto epidérmico. Cistos não odontogênicos. Diagnóstico bucal. Patologia bucal.*

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INTRODUCTION

Epidermoid cysts are benign, painless lesions of congenital or acquired origin, slowly increasing in size [1,2]. Epidermal and dermoid cysts are found throughout the body. Epidemiologically they represent 7% of the head and neck cysts [2,3-5] and 0.01% of those within the oral cavity [1,5,6]. Although rare in the mouth, most of them occur in the midline, in the sublingual region [2,7], but may also appear in other parts of the oral cavity [5,8-12].

The etiology is uncertain and may be associated with remains of the ectoderm retained in the first and second brachial arches [3-6,11-14] accidental or surgical events which lead to a traumatic implantation of the epithelium within deep structures [4,11,13,14]; or a variation of the thyroglossal cyst [4,13,14].

Clinically, epidermoid cyst present rubbery consistency to palpation, defined borders and variable size. Depending on their location and size, it may cause dysphagia, dyspnea, and speech difficulty [9]. However, the final diagnosis is only obtained through the histopathological examination.

The treatment is surgical excision and the location of the cyst, whether above or below the mylohyoid muscle, is a determining factor in the choice of the appropriate intra or extra-oral treatment [4] approach.

These cysts are rarely seen in the first years of life [15] and can often be seen in young adults [1] or in the third or fourth decade of life [16]. Therefore, the objective of this study is to report a rare case of epidermoid cyst in a 5-month-old patient focusing the clinical and histopathological aspects.

CASE REPORT

Approval of the Ethics Committee of Faculdade Educacional de Francisco Beltrão: 2.162.971

A five-month-old female patient was taken by the parents to a dental pediatric appointment when the parents complained of a "ball" on the floor of the mouth floor, which had been present since the baby was born. According to the parents, the pediatrician believed that the lesion was not malignant and referred the patient to the dentist. The first dental appointment was performed at the health clinic, where the dentist suggested, as a diagnostic hypothesis, the presence of a ranula and referred the patient to a pediatric dentist. Anamnese showed the baby's good health condition and breastfeeding was maternal.

The clinical examination revealed the presence of a circular-shaped smooth, whitish surface of fibrous consistency on the left side, near the sublingual gland duct, measuring 0.3 x 0.3 x 0.2 cm, sessile base (figures 1). The parents were advised of a possible cystic lesion diagnosis which should be confirmed by a histopathological examination after excision.

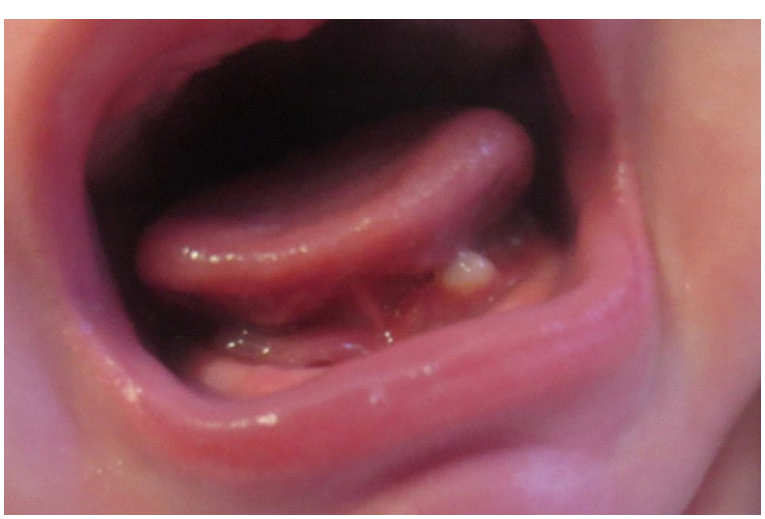

Figure 1. Papule on the mouth floor.

Three days after the first appointment intraosseous antisepsis was performed with a gauze soaked in 0.12% chlorhexidine solution and the child was anesthetized with one third of the anesthetic tube containing 2% Lidocaine solution with Epinephrine 1: 100,000 (DFL - Brazil) by the infiltrative technique, on the floor of the mouth. Clamping was performed using a 15C scalpel blade and the lesion was nucleated (figure 2). Once the lesion was superficial with little bleeding there was no need for suturing, and the specimen was stored in 10% formalin solution. The child was medicated with analgesic after the procedure and was scheduled for another appointment.

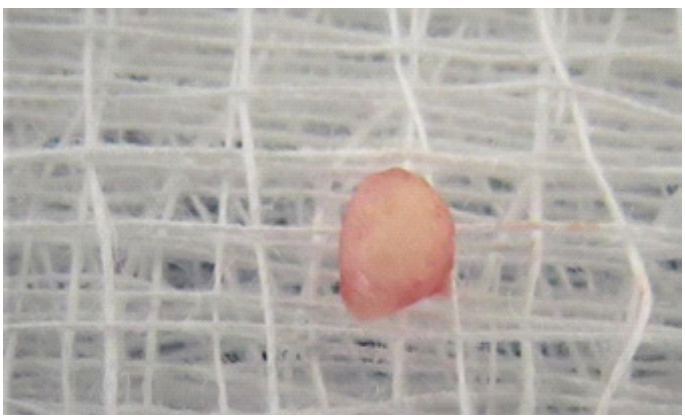

Figure 2. Nucleated lesion.

The histopathological analysis revealed fragment of oral mucosa constituted by parakeratinized epithelial showing areas of acanthosis and atrophy. In the lamina propria, constituted by dense connective tissue, it can be observed a cystic capsule coated by parakeratinized stratified squamous epithelial tissue presenting remnants of keratin in its lumen (figure 3 A, B, C and D).

Several scaly cells were observed in the cystic lumen. Presence of salivary duct and small blood vessels led to an epidermal cyst diagnosis.

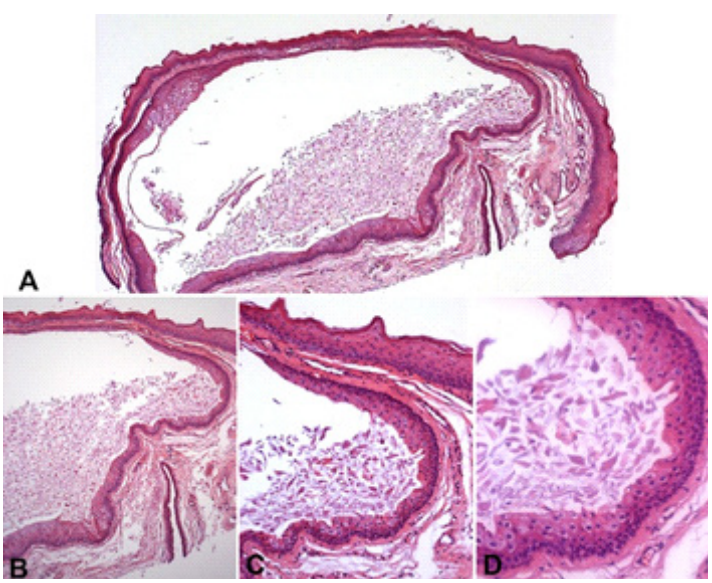

Figure 3. Histopathological exam image.

The case was reassessed in one week and 45 days when the parents were informed of the diagnosis (figure 4). At the six-month follow-up the child did not present a recurrence and the case has been under follow up since then.

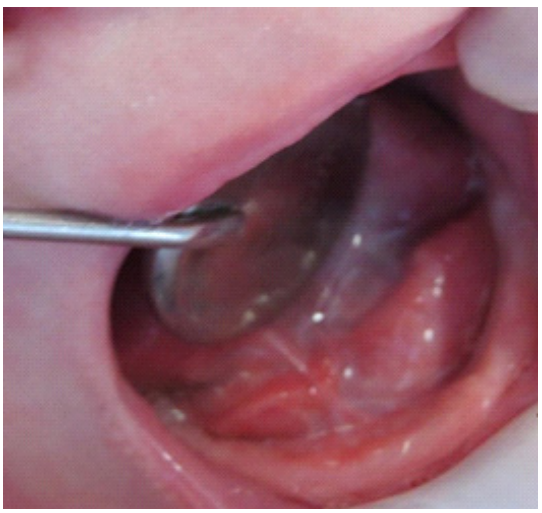

Figure 4. Healing after 45 days.

DISCUSSION

The epidermoid cyst, a part of the cystic teratoma spectrum3, can be according to Meyer (1955) histologically differentiated into three types, as follows: epidermoid (simple): when it presents stratified squamous epithelium and no skin appendages; dermoid (compound): besides the typical squamous epithelium it also encloses skin appendages such as hair, hair follicle as well as sebaceous and sweat glands; terathoid (complex): a cavity lined with epithelium containing mesodermal and endodermal origin such as muscles, intestinal mucosa, respiratory mucosa, fibrous tissue, bone tissue, blood vessels and skin appendages which are typical of dermoid cysts [1,4,5,14].

The etiology of this case is probably related to the entrapment of ectodermal tissue of the first (mandibular) and second (hyoid) brachial arches remanants during fetal development [1,3,5,11,17] during the third and fourth week of intrauterine life. It is a congenital cyst. once it was present since birth. The development of these cysts in the oral cavity is extremely rare [4,5,13,15], although they have been reported in the mouth floor [2,7,14], on the tongue11, lips, oral mucosa [18], pharynx, soft palate [19] and uvula [12].

They may occur during childhood [15,16] and in young adult patients, with age preponderance in patients between 35 and 45 years of age [13]. In the literature, cases of epidermal cyst in childhood (0-10 years) have been reported in the floor of the mouth [2,6,7], on the tongue [6,11], on the uvula [12], in the soft palate [14] and in the pterygopalatine fossa [14]. Studies have shown that few cases occurred in early childhood (0-3 years) [6,15], on the floor of the mouth5, on the tongue [6], on the uvula and in the soft palate [15].

Some authors have found no gender predilection [5,16], others however state either male predilection [17,18] or female [18].

Clinically, the lesions are of a rubbery, soft, palpable consistency, mobile and well- defined borders, separated from the underlying skin [16], asymptomatic, presenting a slow and progressive growth [4,16,17], without lymphadenopathy or signs of infection [13]. If large they can interfere with feeding or speech and tongue displacement (up or behind) [4,17].

Some cases in the literature have used ultrasound [2,5,7,13,16,17], computerized tomography scan [1,10,14,16] and magnetic resonance imaging [1,3,10,14,17] for the diagnosis and treatment planning.

Depending on the location, the differential diagnosis can lead to a dermoid cyst, epidermoid [1,14], infectious processes, ranula [1,3-6,13,14,17], cyst of the thyroglossal duct, cystic hygroma [3,6,13,17], accumulation of adipose tissue, unilateral or bilateral blockage of the Wharton ducts, cyst of the branchial cleft, submandibular and sublingual gland infection, benign and malignant tumors of the mouth floor and adjacent salivary glands [3-5,17], hemangioma [3], lymphoepithelial cyst [4,13,14], lipoma [3,13,17], neurofibroma and lymphangioma [3]. In newborns, it is essential that professionals are able to identify the differences in transient cysts such as Epstein's Pearls, Bohn's Nodules and dental sheet cysts in order to avoid unnecessary therapeutic procedures.

The anatomical classification is based on its relation with the geniohyoid, milo-hyoid and genioglossus muscles, determining whether the lesion is sublingual (located above the geniohyoid muscle); submental (located between geniohyoid) or milo-hyoid or submandibular (located on the lateral aspect of the floor of the mouth). When the lesion is above the geniohyoid muscle, intra-oral swelling may cause tongue elevation and retraction, making it difficult to speak, chew, swallow [2], causing dysphagia, dysphonia and dyspnea [4,5]. If the location is between or below the geniohyoid and milohyoid muscles, swelling may give the patient the appearance of a double chin [4,5]. Most reports of cysts in the oral cavity describe cysts in the submental region.

The epidermoid cyst treatment is surgical without rupture of the cyst, since its content may irritate the surrounding fibrovascular tissue producing postoperative [1,3,5,16-19] inflammation [3]. Surgery can be performed by transcutaneous approach in the upper neck or by intraoral access1. The choice of the type of surgical approach will depend on the location of the cyst in relation to the muscles of the mouth floor [2,4] and the size of the mass [2]. Intraoral access should be chosen whenever the lesion is above the geniohyoid and milohyoid muscle, due to its lower morbidity, absence of dermal scar [3] and faster recovery [3,4]. Large cysts should receive an extraoral approach, due to better visualization of surrounding structures, control of bleeding and intrabuccal contamination [13]. However, the treatment of choice should be the one allowing better visualization of the operative field in order to faciliate the removal of the lesion [17]. Due to the age of our patient, size and location of the lesion, the intrabuccal access was the choice for surgical excision. For larger lesions marsupialization can be performed [3]. Recurrence rarely occurs and it has been reported in less than 3% of cases [19], however it may occur if the cyst is not completely removed [4,13,17,18]. Malignant alterations are also rare [3,18,19], in a proportion of 5% [3,5].

CONCLUSION

A diagnosis of Epidermal cyst was made based on clinical and histopathologeatures. The six-month follow-up the child did not present a recurrence. This case report also highlights the importance of differential diagnosis and management of oral benign lesions in children.

Collaborators

E Stringhini Junior, performed the treatment, writing the original draft– proofreading e editing. DG Kegler, writing the original draft. VAM Montalli, performed the histopathological analysis and proofreading and editing, LB Oliveira, writing the original draft, proofreading and editing.

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