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REVISTA STRICTO SENSU

Epidermoid cyst in submandibular region: a case report

Cisto epidermoide em região submandibular: relato de caso

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Resumo

O cisto epidermóide (CE) é um tumor benigno raro que pode se desenvolver em qualquer região do corpo humano. Cerca de 7% desses cistos estão localizados na cabeça e pescoço. São entidades pouco frequentes na região crânio-facial e ainda mais raras na cavidade oral 1% estatisticamente. Quase todos ocorrem na região sublingual, mas são raros os relatos de casos de ocorrência em outros locais. Os cistos epidermóides são definidos por um epitélio escamoso estratificado revestido por uma camada externa de tecido conjuntivo rico em colágeno. O desprendimento da queratina do revestimento epitelial cria um material branco macio rico em cristais de colesterol. Este conteúdo rico em lipídios do cisto pode produzir uma reação inflamatória vigorosa se ocorrer ruptura do cisto. Relatamos um caso de cisto epidermóide (CE) localizado em região submandibular e o procedimento cirúrgico de remoção. Este estudo foi aprovado pelo Hospital Regional de Ferraz de Vasconcelos (IRB) e o paciente também assinou o Termo de Consenso Informado. O exame clínico e radiográfico revelou edema móvel, mas endurecido à palpação. A análise histológica da biópsia excisional confirmou as hipóteses iniciais de que se tratava de uma lesão CE. Apesar de a CE ser uma lesão benigna rara, o diagnóstico diferencial é obrigatório e não deve ser subestimado devido à sua baixa mas possível transformação neoplásica.

Palavras-chave: cisto epidermoide, mandibula, diagnostico diferencial.

Abstract
The epidermoid cyst (EC) is a rare benign tumor that can develop in any region of the human body. About 7% of these cysts are located in head and neck. They are not frequent entities in cranio-facial region and in still more rare in oral cavity 1% statistically. Almost all of them occur in sublingual region, but there are rare case reports of occurrence in other sites. The epidermoid cysts is defined by a lined stratified squamous epithelium supported by an outer layer of collagenous-rich connective tissue. The detachment of keratin from the epithelial lining criates a soft white material rich in cholesterol crystals. This lipid rich content of the cyst may produce a vigorous inflammatory reaction if cyst rupture ocurrs. We report a case of epidermoid cyst (EC) located in submandibular region and the surgical removal procedure. This study was approuved by the Ferraz de Vasconcelos Regional Hospital (IRB) and the patient signed the Informed Consensus Files additionally. Clinical and radiographic examination revealed a movable swelling but hardened on palpation procedure. Histologic analisys of the excisional biopsy confirmed the initial hypotheses that it was an EC lesion. In spite of the EC being a rare benign lesion a differential diagnose is mandatory and it should not be underestimated due to its low but possible neoplasic transformation.

Keywords: epidermoid cyst, mandibule, differential diagnosis.

1. Introduction

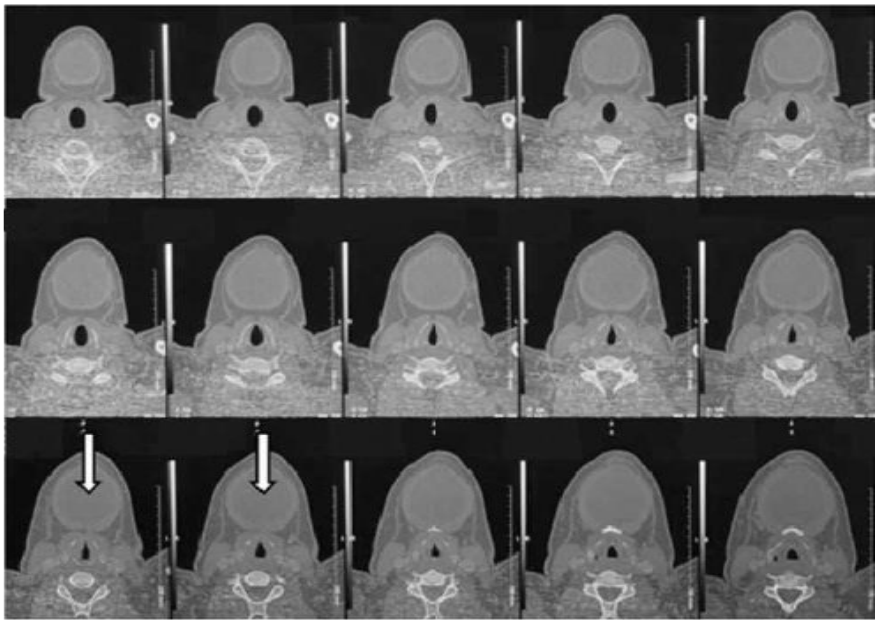
The epidermoid cyst (EC) is a rare benign tumor that can develop in any region of the human body. About 7% of these cysts are located in head and neck. They are not frequent entities in cranio-facial region and in still more rare in oral cavity 1% statistically. Majority of them occur in sublingual region, but there are rare case reports of occurrence in other sites (HOANG et al., 2019; BAREDES et al., 2002). The etiology of EC is still unknown, but it seems to be associated with the remaining of ectoderm trapped on the first and second branchial arches during development, or it occurs as accidental surgical events, in which a traumatic implantation of epithelium cells inside deep structures is achieved (HOANG et al., 2019). EC is commonly associated as a soft floating swelling in the anterior region of the oral floor, displaying slow, progressive and painless growth, with variable dimensions (HOANG et al., 2019; EPLLEY et al., 1985; GIBSON; FENTON, 1982). The intraoral swelling may induce tongue lift. Consequently, speech, chewing and swallowing difficulty are verified. In addition, patient may have a double chin aspect. EC treatment is generally surgical (HOANG et al., 2019). Few cases of EC malign transformation were reported and no recurrence after complete surgical removal (HOANG et al., 2019; DEVINE; JONES, 2000). EC reports in lower jaw region are extremely rare. Surgical approach of encapsulated lesions use to have a good prognosis due to less aggressive procedures (HOANG et al., 2019; MERGIME et al., 2016).

2. Case Report

This Study was approuved by the Ferraz de Vasconcelos regional Hospital IRB and patient signed the Informed Consensus and Informed Consent files as well. A male patient, 19 years old, leukodermata, was admitted in the Surgery-Traumatology Service of Ferraz de Vasconcelos regional Hospital. A Volume increase was observed on the cervical region. Patiente reported in anammesis a increasing volume in the last few

months. No chronic alcohol consumption, no smoking, nor any kind of systemic disease was reported by the patient. The extra-oral physical examination indicated a tumor mass had a considerable extension, hardened on palpation and delimited borders. The intra-oral physical examination evidenced an increased mass mostly in oral floor. Computer Assisted Tomography evaluation (CAT) (IQon Elite Spectral CT, Phillips – Netherland) showed a big abnormal mass in the submental, sublingual and submandibular region with well defined limits, characteristic of cystic lesions images (Figure. 1).

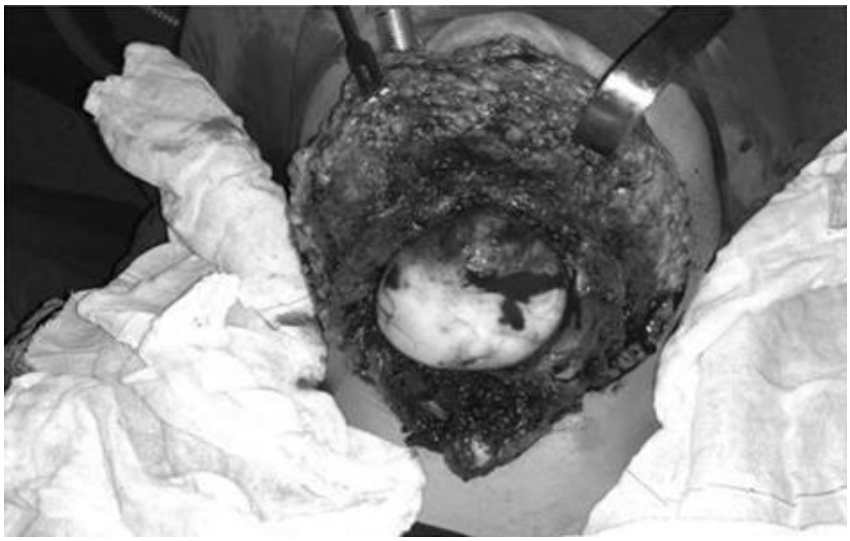
Figure 1: CAT images used to establish the size and limits of the cyst (White arrows).



Source: Own authorship (2020)

The cyst's location is a decisive factor in the surgical technique, indicating an intraoral approach for the sublingual cyst and an extraoral approach for the submental and submandibular ones. The literature reports that jaws EC are very rare. There only few cases described until now and, in some of them, there is a confusion between DC and EC (EPIVATIANOS et al., 2005; SINGH et al., 2012). In our clinical case, the lesion was described as a big dimensions cyst located in the submental, sublingual and submandibular regions. A blood test was carried out at the hospital facilities and didn't show any deviation from normal parameters. After surgical planning a prophilatic antibiotic was injected (Amoxicillin) 2h before surgical procedures. The regional antisepsis procedures was carried out (Poidone-iodine), followed by preanesthesia medication and general anesthesia induction. A nasotracheal intubation were performed and a oropharyngeal plug was introduced to access transcervical region. Surgical incision was done initially at skin level and after that, through the subcutaneous tissue, exposing and dividing the platysma muscle as well as its superficial and deep layer. Additional mouth floor muscles (mylohyoid and digastric anterior bell) had to be dissected to provide enough space for cyst removal. The neurovascular bundles of the region were carefully displaced for cyst dislodgement as well (Figure 2).

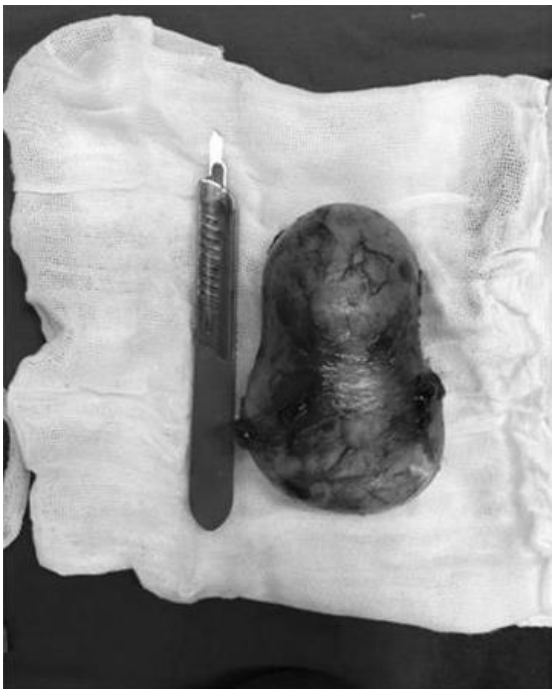
Figure 2: Photography of submandibular region showing soft tissue separation and cyst exposition surgery prior to complete removal.



Source: Own authorship (2020)

Surgical anatomical planes were sutured accordingly and the superficial wound dressing was done. The removed cyst was weighted and measured (160g; 9,0x 5,5x 4,8cm) before been sent to anatomopathological examination. (Figure 3).

Figure 3: Photography of the removed cyst (enucleation technique), prior to histological evaluation.



Source: Own authorship (2020)

Post surgical medication comprised antibiotic (Amoxicilin for 7 days), antiinflamatory (Piroxican for 7 days) and analgesic (Tramadol for 3 days) drugs. The patient remained in the hospital in observation for 3 days after surgry and used chlorhexidine 0,12% for

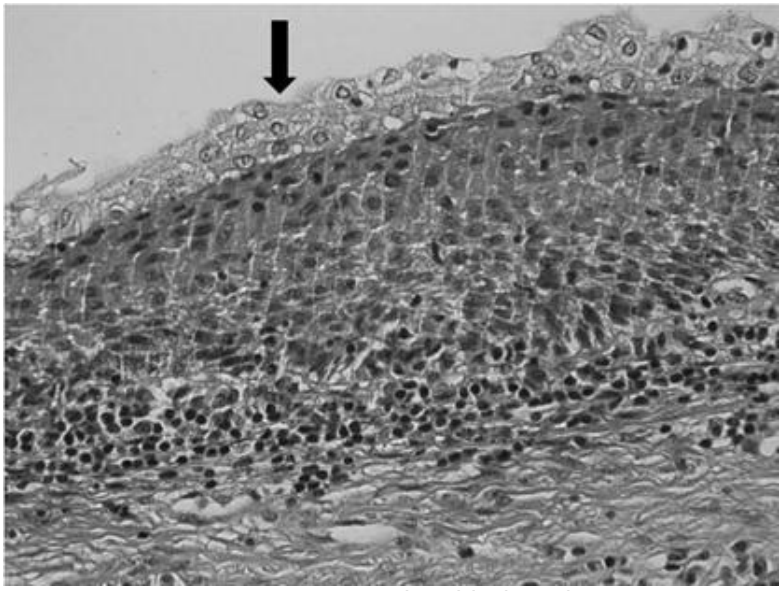
mouthwash. He had no complications on the recovery period. The anatomopathologic evaluation concluded that the specimen presented all the characteristics that define the Epidermoid Cyst. Histopatologic evaluation showed that cyst wall was formed by stratified ephitelium lining cyst content. Ephitelium lining is formed by typical stratified layers (basal, spiny, granular and scamous). However, an addition layer can be observed, composed of bulbous vacuolisated cytoplasm of the ephitelial cells. (Figure 4 and 5).

Figure 4: Photomicrography of the cyst histological stained slide. A leukocyte-rich blood vassel (filled arrow) and the cyst inner superficial ephitelial layer (unfilled arrow). H.E. X100

Source: Own authorship (2020)

The connective tissue just beneath ephitelium evidenced a significant amount of clustered leucocytes, morphologically similar to lymphocytes. The remnant connective tissue is characterized by fibrous connective tissue, commonly found in organs capsules. It is rich in collagen fibers and blood vessels, in addition to fibroblats. (Figures 4 and 5)

Figure 5: Photomicrography of the cyst histological stained slide highlighting the superficial bulbous vacuolisated cytoplasm of the ephitelial cells (arrow). H.E. X400.



Source: Own authorship (2020)

3. Discussion

EC was first described by Cruveilhier and named pearly tumors at 1835. The oral floor is the most common region for the emergence of EC (SAIBABA et al., 2016). A cyst is a space lined by epithelium, and its content is usually a product of its lining, which has no vascular relationship with the wall. Some cysts are of inclusion or retention of normal structures (such as hair follicle-related cysts). The term sebaceous cyst was used for pillar cysts (or trichilemmal cysts, or hairy cysts). Currently, the main cutaneous cysts are epidermal (or epidermal inclusion) and pillars. The cyst is filled with a whitish pasty material, arranged in laminated layers. In the event of an epidermal cyst rupture, the contents are released into the dermal region, which causes a foreign body reaction, in which numerous multinucleated giant cells form a keratin granuloma. This process causes disintegration of the cyst wall and can also lead to a pseudo-epitheliomatous proliferation in residues of this wall, simulating a squamous cell carcinoma. (OHN et al., 2011; DEVINE; JONES, 2000). In fact, it is the better described location of the EC. It can assume one or multiple masses, volume also vary, from small to huge masses distorting neighbor organs and, in some cases, threatening patient's life.(FREITAS et al., 2005; HEMARAJU et al., 2004; CALDERON.; KAPLAN, 1993). In literature we have found a few case reports that associate mandibular cysts with or after extraction of third mandibular molar. Only one case report of bilateral epidermoid associated with impacted mandibular third molar was reported (TOPTAS et al., 2014). According to the literature, etiologic factors of epidermoid cysts are congenital, but there are reports from many authors that trauma was the possible cause of this lesion. These conclusions are based on the theory of traumatic implantation of cystic cells into deeper tissues, and this can be the major etiological factor for the formation of intraosseous epidermoid cysts (ERTEM et al., 2014). Histological analysis shows that epidermoid cysts consist of lined stratified squamous epithelium supported by an outer layer of collagenous tissue. Desquamation of keratin from the epithelial lining produces soft white material rich in cholesterol crystals. The content of the cyst is filled with considerable amount of fat, including some cholesterol which may produce a vigorous inflammatory reaction if the cysts wall disrupts. (LI et al., 2017). On CT scans, the EC appear as moderately thin walled, unilocular masses filled with a homogeneous, hypoattenuating fluid substance with numerous hypoattenuating fat nodules giving the pathognomonic "sack-of-marbles" appearance (PANCHOLI et al., 2006). Due to this particular location, ranula is the first differential diagnostic that comes on mind. In addition

to consistency of tumor mass, image examination provides the most valuable information to clinicians. They can range from radiography to Computer Assist Tomography (CAT), including Magnetic Resonance. Well delimited borders and a cystic cavity direct clinician diagnosis.(ANDERSON; STASSEN, 2014; WORLEY; LASKIN, 1993). The differential diagnosis for sublingual dermoids should comprise ranula, unilateral or bilateral blockage of Wharton's ducts, lipoma, thyroglossal duct cyst, cystic hygroma, branchial cleft cysts, acute infection or cellulitis of the floor of the mouth, infections of submaxillary and sublingual salivary glands, floor of the mouth and adjacent salivary glands benign and malignant tumors, heterotopic gastrointestinal cyst and duplication foregut cyst (KANDOGAN et al., 2007; DAMLE et al., 2002). Anatomically, it is possible to classify these cysts according to their relationship with the musculature of the floor of the mouth. According to D'Antonio (2000), they can be located above the geniohyoid and below the genioglossus muscle, causing a bulging in the floor of the mouth; may be located below the geniohyoid muscle and above the mylohyoid muscle, bulging the submental region, or laterally, in space submandibular, above the mylohyoid muscle and lateral to the base of the tongue. This classification has interest to determine the surgical access route, which can be intraoral, extraoral or mixed. (MOREIRA et al., 1997; D'ANTONIO et al., 2000; KANDOGAN et al., 2007). Lesion removal is mostly determined by cyst volume. Small size cysts are resected via intra-oral surgical access. On the other side hand, big size cysts find their way out via extra-oral surgical access. Removal surgical maneuver usually is not complicated due to a well delimited connective tissue capsule and non-adherence to neighbor tissues/organs.(KOCA et al., 2007; CALDERON; KAPLAN, 1993). Final diagnostic is established by histopathological sample evaluation. (JHAM et al., 2007; OGINNI et al., 2014; WORLEY; LASKIN, 1993). Cyst epithelial cells are positively marked with anti-bodies Against podoplanin. (ASSAF et al., 2012). Caution should be considered in post-operative and patient procession. Literature presents cases of cyst recurrence and cases malignant transformation.(LI et al., 2017; FALTAOUS et al., 2019).

4. Conclusion

The EC is frequently found in the subcutaneous tissue, and usually does not pose great dificulties to resolve, despite the need for a surgical approach. EC of the oral cavity is an uncommon entity. The EC should not be underestimated, despite been considered a rare benign tumor. Clinical and anatomopathological diagnosis are mandatory. The clinical and histopathological details can difficult the diagnosis, mainly because its similarities to dermoid cysts. Literature support the understanding that EC and DC are distinct pathological entities. Lastly, EC treatment is well established, being improbable the relapse after surgery. Ample understanding and vigilance about this slow growing painless mass is essential not only because of the symptoms it produces but also due to its low but possible malignant potential.

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