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Two Cases of Epidermoid Cyst of the Buccal Floor Simulating: A Tumour of the Submandibular Gland

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Abstract

Dermoid cyst of the oral floor is rare benign tumour, who having three histological aspects: dermoid, teratoid and epidermoid. This one is characterized by the presence of a squamous stratified epithelium with cutaneous remnants. It may occur in any part of the body, however their frequency in the ENT sphere is relatively scarce. Seven per cent (7%) only of epidermoid cysts occur in the cervico-facial area, 1.6% of which locate at the floor level. When they are located submandibular, they can pose diagnostic difficulties and look like a tumour of the submaxillary gland. We report two cases of epidermoid cyst of the floor. Both patients suffered from swelling of the submandibular gland. Magnetic Resonance Imaging was not requested due to lack of resources. However, surgery allowed in both cases the excision of a cyst next to a normal submandibular gland. Patients did well post operatively.

Keywords

Epidermoid Cyst, Mouth Floor, Submandibular Gland, Surgery, Tumour

1. Introduction

Epidermoid cyst is uncommon developmental cystic malformations termed dysontogenetic cyst [1]. They are related to an abnormal localization of the ectodermic tissue.

In 1955, Meyer updated the concept of dermoid cyst to describe three histological variants: The true dermoid cyst, the epidermoid cyst, and the teratoid va-

riant [2]. True dermoid cysts are cavities lined with epithelium showing keratinization and with identifiable skin on the cyst wall. Epidermic cysts are lined with simple squamous epithelium with a fibrous wall and no attached structures. The lining of teratoid cysts varies from simple squamous to a ciliate respiratory epithelium containing derivatives of ectoderm, mesoderm, and/or endoderm. All three histological types contain a thick, greasy-looking material [2].

Epidermoid and dermoid cysts constitute 1.6% to 6.9% of all cysts in the head and neck area. They can be found anywhere in the body. Common location sites are the orbit, calvarial diploic space and intracranially. The incidence in the floor of the mouth of the oral cavity is rare [1] [2] [3] [4] [5] and represented less than 0.01% of all cysts of the oral cavity. Sublingual, submaxillary and submandibular spaces are common localization in the floor of the mouth [2].

Epidermoid cysts generally present slow and progressive growth, and even if they are congenital, the diagnosis is commonly possible in the second or third decade of life. They appear as painless, asymptomatic mass, slowly increasing in size, usually located in the midline, above or below the mylohyoid muscle.

Treatment of epidermoid cysts of the floor of the mouth is surgical and can be intraoral or extraoral according to the localization and the size of the lesion [6].

We report two cases of floor cysts which clinically looked like a submandibular gland tumour and whose anatomo-pathological examination led to an epidermoid cyst.

2. Clinical Cases

We report the cases of two patients with no pathological history. The first, a 23-year-old woman with AD as initials and the other, a 21-year-old man with MD as initials.

They suffered from swelling of the submandibular region, on the right in AD and on the left in MD. Clinical examination found in both cases a firm, painless and non-inflammatory mass bringing upwards the lateral part of the floor but with no functional discomfort associated. The symptomatology had been evolving respectively for eight years and two years.

The two ultrasound results suggested a hypertrophy of the submandibular gland. Further magnetic resonance imaging (MRI) could not be performed because patients didn't have medical insurance that covers the costs. Therefore, we decided to do an exploration in the operating room under general anesthesia.

The patients were admitted to the operating room and during surgery, we carried out a complete excision of a deep cystic pocket independent of the submandibular glands that looked healthy (**Figures 1-7**). Incision of the surgical specimen left sebum to weld (**Figure 4**, **Figure 8**).

The post-operative period was uneventful and they were released two days after surgery. The anatomo-pathological examination of the two specimens concluded to an epidermoid cyst, with, the presence of a cystic wall made up of dense fibrous tissue lined with a squamous epithelium surmounted by lamellae

of keratin (Figure 9).

Figure 1. Intraoperative discovery of a cyst next to the submandibular gland.

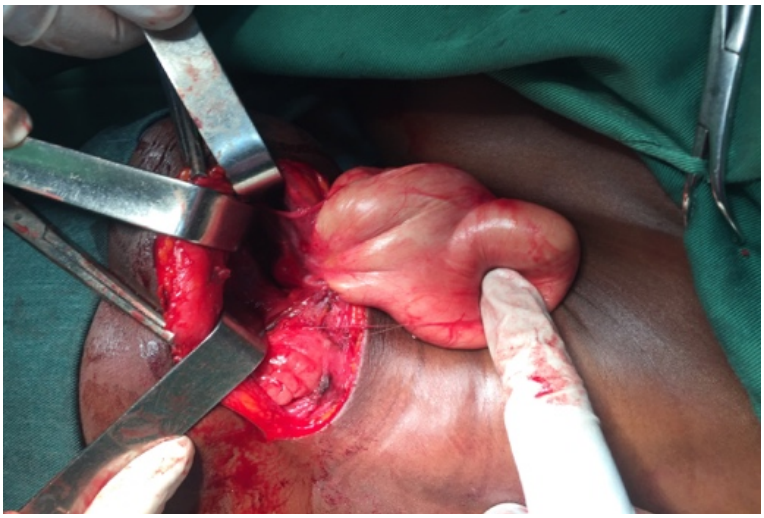

Figure 2. Progressive cyst excision.

Figure 3. Complete excision of the cystic pocket.

Figure 4. Incision of the cystic pocket revealing thick yellowish contents.

Figure 5. Discovery of a cyst during surgery.

Figure 6. Progressive cyst excision.

Figure 7. Complete excision of the cystic pocket.

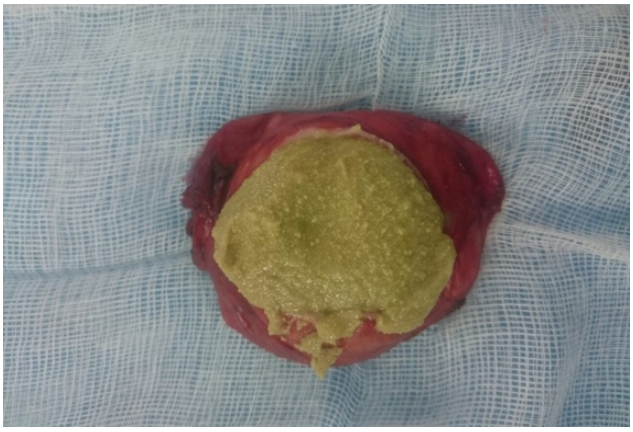

Figure 8. Incision of the cystic pocket revealing thick greenish contents.

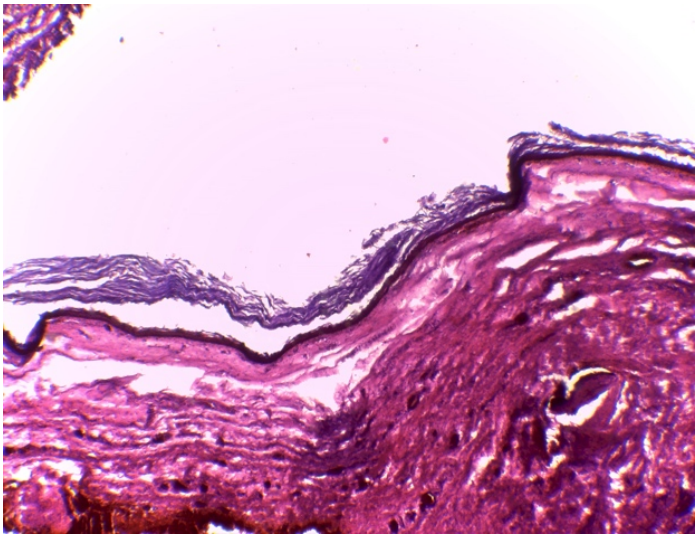

Figure 9. Histological examination: presence of a cystic wall made up of dense fibrous tissue lined with a squamous epithelium surmounted by lamellae of kerat.

Patients were monitored regularly until complete recovery and no recurrence was noted.

3. Discussion

The floor of the mouth and submandibular area may be affected by numerous pathologic conditions, which can be broadly classified as developmental, inflammatory, obstructive, or neoplastic in origin. Lesions in this area may be present for a prolonged period of time before the patient seeks medical advice, usually as a result of interference with swallowing or speech. The floor of the mouth is the most common intraoral location for developmental lesions of the oral soft tissues, particularly dermoid and epidermoid cysts, lipoma, branchial cleft cysts, and thyroglossal duct cysts [6].

Epidermoid cyst is one of the three congenital cysts of embryogenesis [3]. The other two are the dermoid cyst and the teratoid cyst. During the closure of the first and second branchial arch, the inclusion of epithelium remains leads to the occurring of epidermoid cysts [3].

Epidermoid cysts are a rare occurrence at the cervico-facial. They represent only 7% of the cystic masses in the region, of which 1.6% occur in the oral cavity [1] [3] [4] [5]. New and Erich (1937) reported 24 (1.6%) epidermoid cysts occuring at the floor of the mouth out of 1495 cases of dermoid cysts seen at the Mayo Clinic [6].

Epidermoid cysts can be congenital or acquired (post traumatic) with no clinical or histological difference between the two [3].

Anatomical classification divides the epidermoid cysts of the buccal floor into three categories depending on their location in relation to the muscles of the floor of the mouth. The sublingual cyst or median genio-glossus sitting above the genio-hyoid muscle; the median genio-hyoid cyst located at the submental area between the genio-hyoid and the mylo-hoid and the lateral dermoid cyst at the submandibular region.

They occur at all ages, but are more common in young adults between the second and third decades of life, with a male predominance [3]. Most patients with epidermoid cyst are in the range between 10 and 35 years of age. In a series of 16 cases, the mean age is 27.8 years and the ratio of men/women is 3:13, although previous papers have found no difference by gender while others have found predominance of women. Growth of the cyst may be constrained by hormonal stimulus during puberty, producing a hypersecretion of fat, which would explain the greater incidence in the young adult stage (16 - 40 years of age) [6].

Clinically, it takes the shape of a well-limited painless mass covered by a healthy mucosa that grows gradually. Some cysts can get infected and associate with skin fistulas. In children, some big cysts can cause respiratory discomfort, swallowing difficulties and speech disorders [2] [6] [7]. The cystic mass can vary in size from a few millimeters up to 4.72 in diameter [7].

Indeed, the MRI, the examination of choice, confirms the cystic nature by

putting into evidence a lesion with hyposignal T1 and hyposignal T2 not evolving after injection of contrast media [1] [3] [4] [5] [7]. MRI also shows with accuracy the very localization of the cyst and its relationship with the genio-hyoid and mylo-hyoid muscles. This allows to choose the best surgical approach particularly for extensive lesions [8] [9].

Treatment of epidermoid cyst is exclusively surgical. A complete excision is necessary to avoid recurrences [8] [9]. The presence of a fibrous capsule allows easy enucleation of the cyst. Two possibilities have been described: the intraoral route and the cervical route. The choice of surgical approach depends on the location of the cyst on the floor of the mouth as well as its extent.

The intraoral route is generally considered for sublingual cysts. The cervical route is primarily considered for cysts exceeding the limit of the genio-hyoid muscle as was the case in our patients. When lesions are extensive, both approaches can be combined [8] [9].

Histologically there exist three subtypes: epidermoid cysts, dermoid cysts and teratoid cysts or teratomas [3] [4] [5]. Real dermoid cyst contains squamous epithelium with dermal derivatives (hair follicles, swet and sebaceous glands). When the latter are not present it is then called epidermoid. Teratomes contain, in addition to epithelial elements, bone and cartilaginous structures.

Prognosis is very good, with a very low incidence of relapse, usually related to bone remnant to the genial tubercles or to the hyoid bone. Malignant changes have been recorded in dermoid cysts by New and Erich but not in the floor of the mouth, although a 5% rate of malignant transformation of oral dermoid cysts has been reported by other authors, but only for the teratoid type [6].

4. Conclusion

Cervical epidermoid cyst is rather rare. It can be difficult to properly diagnose particularly at the submandibular area. Imaging is of crucial importance in the diagnostic procedure and in the surgical planning.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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