Rare disease

A rare case of congenital epidermoid cyst of the hard palate

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Correspondence to Professor Lucio Montebugnoli, lucio.montebugnoli@unibo.it**Summary**

Epidermoid cysts are benign conditions that are thought to derive from abnormally situated ectodermal inclusions in the oral cavity. They are generally found in hands, fingers, feet, ovaries and testicles but in oral cavity they represent a very rare event. This is the first case of an intraosseous epidermoid cyst situated in the hard palate. Healing was uneventful and there was no sign of recurrence in 2-years follow-up.

BACKGROUND

Epidermoidal oral cysts (EOC) are rare conditions in the head and neck region that are derived from abnormally situated ectodermal tissue.^{1 2} They are inclusion cysts lined by ectoderm³ without any annexed structures, consisting of squamous epithelium that may contain cheesy keratinous material.⁴

They represent less than 0,01% of all oral cavity cysts and mainly affect the floor of the mouth, lateral tongue, lateral pharyngeal wall and soft palate.⁵⁻⁷ There is only one report case in the literature of EOC localised in the hard palate that presented as a lobulated mass without any bone involvement.⁶ We report the first case of EOC localised in hard palate with bone involvement.

CASE PRESENTATION

In April 2008, a 26-year-old woman presented to our department for a routine dental check-up. Incidental intraoral radiographs showed a radiolucent rounded lesion between

upper left premolars' roots appearing with well-defined margins and a small sclerotic hem delimiting the cavity (figure 1). The patient was asymptomatic and family and personal medical history revealed nothing meaningful.

INVESTIGATIONS

Electric pulp test ascertained the vitality of adjacent teeth, and periodontal probing depths were less than 3 mm. CT scan of the upper maxillary bone allowed to measure a diameter of the lesion of 7,5 mm and to identify a partial interruption in the palatal cortical bone.

DIFFERENTIAL DIAGNOSIS

The proximity with premolars' roots imposed EOC to be distinguished from lateral radicular cyst resulting from an inflammatory stimulus through a lateral root canal of a non-vital tooth, lateral periodontal cyst, odontogenic keratocyst along the lateral root surface and radiolucent odontogenic tumours.

Figure 1 Radiographic examination shows a radiolucent rounded lesion of 7–8 mm in diameter between upper left premolars' roots.

Figure 2 Surgical excision of the lesion: raising of palatal mucosa of premolar region in a full-thickness flap (A); enucleation of the cyst (B) and bone defect immediately after surgery (C).

Figure 3 On histology, the cystic cavity was lined by orthokeratinised squamous epithelium. The cystic wall was composed of fibrous connective tissue. There was no evidence of skin appendages (as sebaceous glands, hair follicles or sweat glands). Focal chronic inflammatory infiltrate was observed (white arrow) (A). At higher power the orthokeratinised squamous epithelium (indicated by the white arrows) is devoid of cytological atypia. The cystic lumen is filled with keratin debris (B).

TREATMENT

Treatment consisted of local excision and curettage of the socket. For the operation, a sulcular incision was performed to raise palatal mucosa of premolar region in a full-thickness flap. Palatal cortical bone appeared to be partially reabsorbed from the expansion of underlying lesion (figure 2A). The cystic lesion was easily enucleated since her wall was not very adherent to the surrounding bone (figure 2B). Finally, inner walls of the bone defect were

cleansed with curettage (figure 2C). The cavity was filled with fibrinoid material to help regeneration of the tissues and to avoid the collapse of the same ones. Healing after surgery was uneventful and no sign of recurrence was noted in 2-years follow-up.

Macroscopically, the surgical specimen consisted of 1 cm diameter yellowish, firm in consistency mass. Microscopic examination showed cystic lining composed of stratified squamous epithelium, focal hypergranulosis and focal

Figure 4 Radiographic examination at 2-years follow-up shows a complete healing of the site.

chronic inflammation, without any cytological atypia, surrounded by fibrous tissue. Cystic lumen contained keratinous material and no annexed ectodermal appendages such as hair, sweat glands, sebaceous glands, and so on were noted (figure 3A,B). Histological diagnosis was of epidermoidal cyst.

OUTCOME AND FOLLOW-UP

Healing after surgery was uneventful and no sign of recurrence was noted in 2-years follow-up.

A complete healing of the site was achieved 2 years after surgery (figure 4).

DISCUSSION

EOC are rare, benign conditions in the head and neck region that are derived from abnormally situated ectodermal tissue.^{1 2} Epidermoid cysts can occur anywhere in the body; about 7% of them can be found in the head and neck region, and only 1.6% within the oral cavity.^{5 7} In the oral cavity, they mainly affect the floor of the mouth, lateral tongue, lateral pharyngeal wall and soft palate, and only a case report was reported by Ravindranath in the hard palate.^{6 7} Differently from that case where EOC presented as a lobulated mass, brownish pink in colour, without any bone involvement, the case here presented proved to have an intraosseous localisation. The unusual localisation of the lesion, and the proximity to the roots of upper premolars teeth, put the problem of a correct differential diagnosis with other similar lesions most commonly found in the same area. Lateral radicular cyst was excluded by vitality pulp test, while a histological examination following excision of the lesion was required to exclude lateral periodontal cyst, odontogenic keratocyst and radiolucent odontogenic tumours. Histologically, EOC can be divided into epidermoid, dermoid and teratoid.⁸ In our case, the lesion showed simple squamous epithelium without skin appendages, characterising it as

an epidermoid cyst. Pathogenically, EOC can be divided into congenital and acquired types, without any clinical or histologic differences.⁹ Acquired factors include aberrant epithelial components in deep tissue due to trauma, inflammation or surgical complications which may be mechanisms of onset,¹⁰ while congenital EOC develop from inclusion of ectodermal tissue during embryogenesis.⁶ Previous trauma or surgical intervention was not encountered in our case suggesting a congenital epidermoidal cyst. EOC are characterised by a slow and painless growth, attracting little attention until their size gives annoyance.^{4 6} Surgical excision is the treatment of choice and the tool for a definitive diagnosis.¹ Prognosis is good, malignant transformation is widely reported as a very rare event,^{11–14} and recurrences have been described in a recent review as less than 3%.¹⁵

Learning points

- ▶ Epidermoid oral cysts are rare lesions in the oral cavity.
- ▶ Prognosis is good: relapse is uncommon and malignant transformation is very rare.
- ▶ They should be considered in the differential diagnosis with odontogenic cysts.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Ozer E**, Kanlikama M, Bayazit YA, *et al*. A unique case of an epidermoid cyst of the pterygopalatine fossa and its management. *Int J Pediatr Otorhinolaryngol* 2003;**67**:1259–61.
2. **Marsot-Dupuch K**, Leroux C. [Epidermoid cyst of the base of the skull]. *Ann Radiol (Paris)* 1992;**35**:472–6.
3. **Ulku CH**, Uyar Y, Kocaogullar Y, *et al*. Iatrogenic epidermal inclusion cyst of the parapharyngeal space: unusual complication of ear surgery. *Skull Base* 2004;**14**:47–51.

4. **Makarand V**, Dinaz K, Navin IL. Epidermoid cyst of the floor of the mouth. *BHJ* 2002;**44**:267.
5. **De Ponte FS**, Brunelli A, Marchetti E, *et al*. Sublingual epidermoid cyst. *J Craniofac Surg* 2002;**13**:308–10.
6. **Ravindranath AP**, Ramalingam K, Natesan A, *et al*. Epidermoid cysts: an exclusive palatal presentation and a case series. *Int J Dermatol* 2009;**48**:412–5.
7. **Turetschek K**, Hospodka H, Steiner E. Case report: epidermoid cyst of the floor of the mouth: diagnostic imaging by sonography, computed tomography and magnetic resonance imaging. *Br J Radiol* 1995;**68**:205–7.
8. **Jham BC**, Duraes GV, Jham AC, *et al*. Epidermoid cyst of the floor of the mouth: a case report. *J Can Dent Assoc* 2007;**73**:525–8.
9. **Sánchez Torres J**, Higa TT. Epidermoidal cysts in the oral cavity. Report of three cases. *Oral Surg Oral Med Oral Pathol* 1970;**30**:592–600.
10. **Suga K**, Muramatsu K, Uchiyama T, *et al*. Congenital epidermoid cyst arising in soft palate near uvula: a case report. *Bull Tokyo Dent Coll* 2010;**51**:207–11.
11. **Yilmaz I**, Yilmazer C, Yavuz H, *et al*. Giant sublingual epidermoid cyst: a report of two cases. *J Laryngol Otol* 2006;**120**:E19.
12. **Pancholi A**, Raniga S, Vohra PA, *et al*. Midline submental epidermoid cyst: a rare case. *Internet J Otorhinolaryngol* 2006;**4** http://www.ispub.com/journal/the_internet_journal_of_otorhinolaryngology/volume_4_number_2_33/article/midline_submental_epidermoid_cyst_a_rare_case.html (accessed 30 June 2011).
13. **Hoeffel C**, Heldt N, Chelle C, *et al*. Malignant change in an intradiploic epidermoid cyst. *Acta Neurol Belg* 1997;**97**:45–9.
14. **Wong TH**, Khoo AK, Tan PH, *et al*. Squamous cell carcinoma arising in a cutaneous epidermal cyst—a case report. *Ann Acad Med Singap* 2000;**29**:757–9.
15. **MacNeil SD**, Moxham JP. Review of floor of mouth dysontogenic cysts. *Ann Otol Rhinol Laryngol* 2010;**119**:165–73.

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