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CASE REPORT

Coexistence of lymphoepithelial and epidermoid cysts on the floor of the mouth: report of a case

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Lymphoepithelial and epidermoid cysts in the oral cavity are uncommon. The coexistence of lymphoepithelial and epidermoid cysts in the oral cavity is extremely rare. Only one case of lymphoepithelial cyst associated with two epidermoid cysts on the floor of the mouth has been reported in the literature and the present report describes a second case where a lymphoepithelial cyst coexisted with an epidermoid cyst on the floor of the mouth. It is likely an accidental trauma that was accompanied by inflammation produced the development of implantation-keratinizing epidermoid and lymphoepithelial cysts.
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Introduction

Lymphoepithelial and epidermoid cysts occur rarely in the oral cavity. Oral lymphoepithelial cysts present as movable, painless submucosal nodules with a yellow or yellow-white discoloration. Occasional cysts are transparent. Approximately half of all intraoral cases are located on the floor of the mouth. When the epidermoid cyst is located on the floor of the mouth its clinical appearance is similar to that of the lymphoepithelial cyst (Bouquot and Nikai, 2001).

The coexistence of lymphoepithelial and epidermoid cysts in the oral cavity is extremely rare. To our knowledge only one case has been reported in the literature (Ahn *et al*, 1996). The purpose of the present study was to present one case of lymphoepithelial cyst that coexisted with an epidermoid cyst.

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Case report

On March 3, 2003, a 27-year-old man was referred to our Department because 1 week ago his dentist on routine practice noticed the presence of a swelling on the floor of the mouth. His medical history and family history were unremarkable. On systematic physical examination no abnormality was detected except a small, round, circumscribed soft mass on the left side of the floor of the mouth adjacent to the ventral surface of the tongue, measuring 6 mm in diameter. The lesion was slightly raised from the surrounding normal mucous membrane, smooth, whitish in color and freely movable from the underlying tissue (Figure 1). The lesion was clinically diagnosed as mucocele and under local anesthesia was totally excised. The wound was closed primarily and healed without complication. One year after surgery, there was no recurrence.

Microscopically, lymphoid tissue exhibiting germinal centers in some areas surrounded the lining epithelium of a cyst. The lining epithelium was thin with lack of rete pegs and was parakeratinized. The lumen of the cyst contained sloughed epithelial cells, lymphocytes and polymorphonuclear leukocytes. Minor salivary glands and salivary ducts were not found adjacent to the lymphoepithelial cyst (Figure 2). A small cyst was observed adjacent to the surface oral epithelium. Examination of serial sections confirmed the presence of this cyst that appeared to be located in the basal layer because of the plane of the sections. There was no continuity of this cyst with either the underlying lymphoepithelial or its mucosal epithelium.

The lining epithelium of the cyst was parakeratinized, not surrounded by lymphocytes and its lumen contained some desquamated parakeratinized cells. Small bud-like projections of the lining epithelium of this cyst into adjacent stroma and lumen were observed (Figures 3 and 4). The case was diagnosed as lymphoepithelial cyst

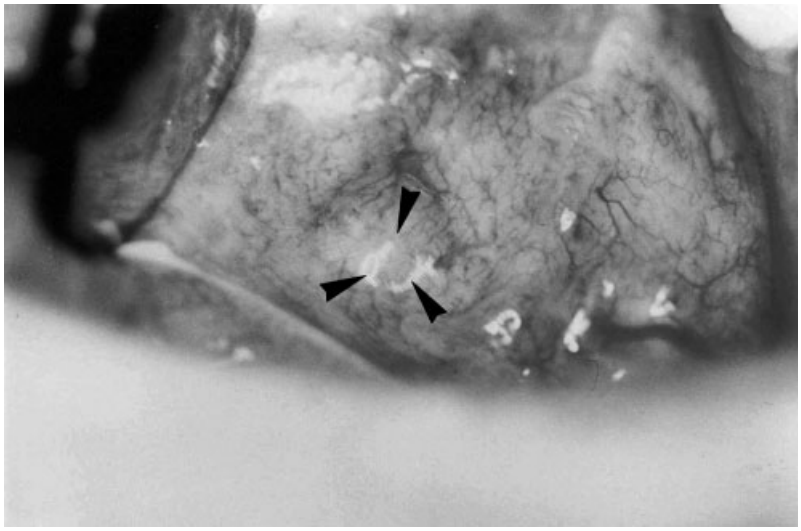

Figure 1 Small swelling on the floor of the mouth (arrowheads)

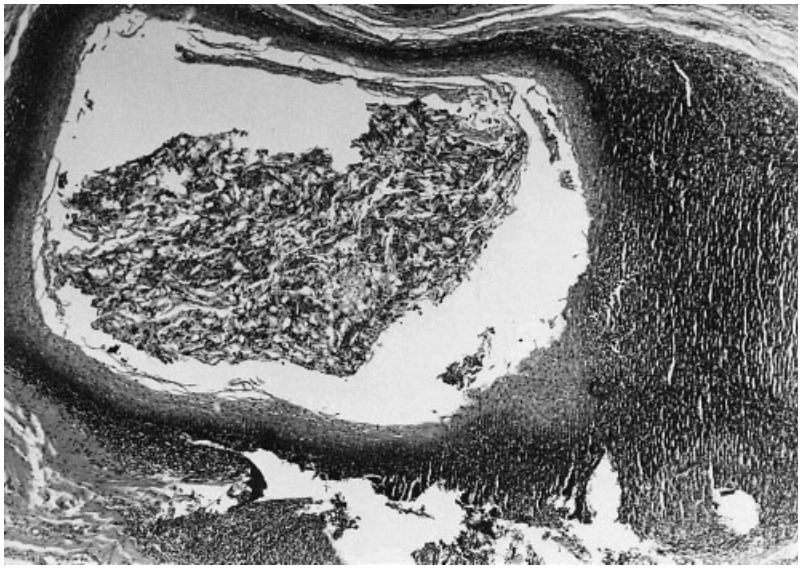

Figure 2 The lining epithelium of the lymphoepithelial cyst is thin without rete pegs and parakeratinized. Desquamated parakeratinized cells, lymphocytes and polymorphonuclear leukocytes fill the lumen (hematoxylin and eosin. Original magnification ×33)

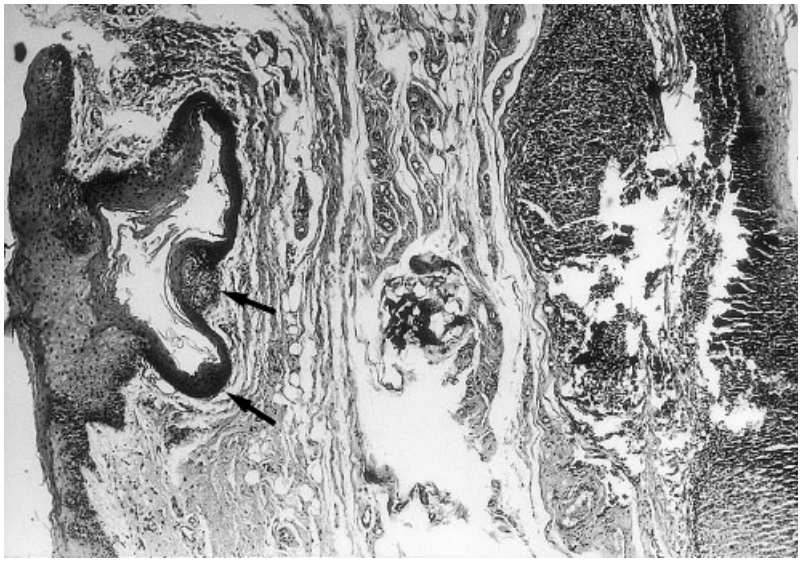

Figure 3 The lining epithelium of the epidermoid cyst is parakeratinized and the lumen contains some desquamated parakeratinized cells. Small bud-like projections of the lining epithelium into adjacent stroma and lumen of the cyst (arrows). On the right side of the illustration lymphoid tissue and part of the lining epithelium of the underlying lymphoepithelial cyst are seen (hematoxylin and eosin. Original magnification ×33)

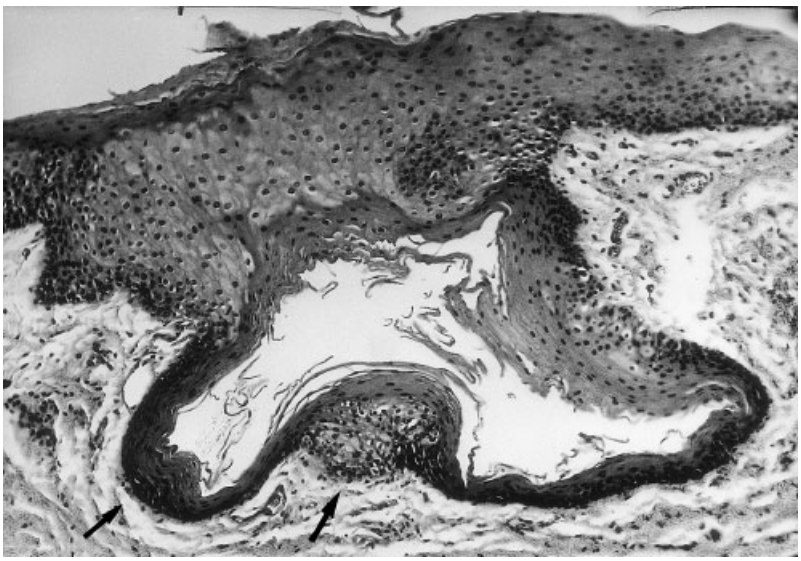

Figure 4 High magnification of the epidermoid cyst (hematoxylin and eosin. Original magnification ×132)

coexisting with implantation-keratinizing epidermoid cyst.

Discussion

The etiology of the epidermoid cyst is basically unknown. The most popular theory was that it can arise by the development of the entrapped ectoderm of the first and second branchial arches. Another theory proposes surgical or accidental events by a traumatic implantation of epithelial cells into deeper tissues. The epidermoid cysts that arise after implantation of the epithelium into deeper structures are called implantation-keratinizing epidermoid cysts (Ettinger and Manderson, 1973), are limited in size and intraorally most often occur on the floor of the mouth (Rajayogeswaran and Eveson, 1989).

According to Knapp's (1970) theory the lymphoepithelial cyst of the oral cavity represents a dilated, obstructed crypt of the oral tonsil in which either purulent material or desquamated epithelial lining accumulate. Buchner and Hansen (1980) supported Knapp's theory and demonstrated all stages in the development of the lymphoepithelial cyst. They suggested that the lining epithelium of the cyst separates from the superficial epithelium at later stages. In the present study serial sections of the lesion did not reveal continuity of the lining epithelium of the lymphoepithelial cyst with the superficial small cyst. Therefore, it is possible that this superficial cyst represents a cyst-like degeneration of a tonsilar crypt opening in which contact with the lymphoepithelial cyst was lost. However, coexistence of the lymphoepithelial cyst with superficial small intraepithelial cyst-like structures such as in the present case has not been reported in the literature.

Another possibility is that an accidental trauma on the floor of the mouth, unnoticed by the patient, accompanied by inflammation led the development of a superficial implantation-keratinizing epidermoid cyst and an underlying lymphoepithelial cyst. In our case, the parakeratinized lining epithelium of the superficial small cyst, the presence of desquamated parakeratinized cells in its lumen and the small bud-like projections of its lining epithelium into adjacent stroma and lumen are histological features supporting evidence of implantation-keratinizing epidermoid cyst. The lining epithelium of the epidermoid cyst is sometimes well formed and consists of a basal cell layer and several rows of prickle cells. Even rete pegs may be recognized at times and the epithelial cells may proliferate in a patchy or papillary fashion to produce bud-like projections into the lumen of the cyst or into neighboring stroma (Asley, 1990). According to Knapp (1970) and Buchner and Hansen (1980) inflammation that accompanies trauma leads to the development of the lymphoepithelial cyst. The presence of polymorphonuclear leukocytes in the lumen of the lymphoepithelial cyst could support evidence of inflammation that led to the obstruction of the crypt of the oral tonsil and the development of the cyst.

The location of a small-sized epidermoid cyst just above the lymphoepithelial cyst produced a unique elevated nodule on the floor of the mouth. Ahn *et al* (1996) reported a case where a lymphoepithelial cyst coexisted with two adjacent epidermoid cysts, producing a unique swelling on the floor of the mouth. They suggested that the two epidermoid cysts developed by implantation of the epithelium into the lamina propria and the coexistence of the two forms of cysts was not uncertain.

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