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Bilateral Intraosseous Lymphoepithelial Cyst of the Mandible

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Abstract. *A 29-year-old healthy patient was submitted for exploration of intraosseous, unicystic lesions of the mandibular angle distal to the wisdom tooth of both sides. Histological investigation of the enucleated cystic lesions revealed bilaterally developed lymphoepithelial cysts (LEC). Healing was uneventful and re-ossification of the bone complete. LEC are rarely diagnosed in oral tissues and usually located in the soft tissues. This report details the clinical, radiological and morphological findings of intraosseous LEC of the mandible.*

Lymphoepithelial cysts (LEC) are rare findings in the oral cavity and maxillofacial regions (1-6). LEC represents a lesion developing in soft tissues (1-6). In the maxillofacial region, the parotids are the predominant organs giving rise to LEC (7), frequently associated with other findings in the context of acquired immunodeficiency syndrome disease (AIDS) (8, 9). Inside the oral cavity, LEC have occasionally been noted, predominantly diagnosed in specimens from the floor of the mouth (10, 11), the tongue (12) and in Waldeyer's ring (12). LEC is not expected in osseous lesions of the jaws. This report adds the unexpected findings of bilateral intraosseous LEC to the literature of LEC of the oral region.

Case Report

Patient. A 29-year-old, healthy female was submitted by her dentist to our outpatient clinic to clarify the incidental finding of bilateral cystic lesions of the mandible that were depicted on a routinely performed radiograph. The HIV-negative patient had no history of earlier surgical interventions of the mandibular retromolar region. Inside the oral cavity, the teeth had emerged normally and were in their anatomic positions, including the fully erupted lower third molars. The oral mucosa was intact and showed no irregularities, including the retromolar pads.

Radiology. On the orthopantomogram (Figure 1), an oval radiotranslucency was depicted on both sides of the mandible that was located close to the distal interface of the third molars. These obviously osteolytic lesions were sharply demarcated to the mandible. Cone-beam tomograms were obtained to analyse the dimensions of the lesions. The lesions were almost completely located inside the mandible. The bone was reduced to the cortical borders lateral to the lesion and outreached the mandibular canal. The upper side of the cystic lesions was focally not covered by bone (Figure 2).

Therapy. The cysts were excised following local anaesthesia. The tissues obtained at excisional biopsy were fixed in formalin immediately after surgery and sent to pathology. The subsequent healing was uneventful and radiographs obtained 2 years after surgery showed complete ossification of the regions (Figure 3).

Histopathology. The tissues were cut in parallel slices and completely embedded in paraffin wax and stained with hematoxylin-eosin and periodic acid Schiff (PAS). Both lesions displayed identical morphological patterns of a cyst lined with non-keratinized squamous epithelium and fibrous cyst wall. The epithelium displayed no atypia. Furthermore, neither respiratory epithelia nor other cell type was present within the epithelial surface. The subepithelial stroma was densely infiltrated with lymphocytes and focal lymphocyte diapedesis into the squamous epithelium. The fibrous cyst wall did not contain any glandular structures or odontogenic-type tissues. The histopathological diagnosis was a bilateral LEC without formation of lymph follicles containing germinal centers (Figure 4).

Discussion

The radiographic appearance of these unilocular lesions was indistinguishable from several other odontogenic or osseous tumours or lesions known to have a preference for developing

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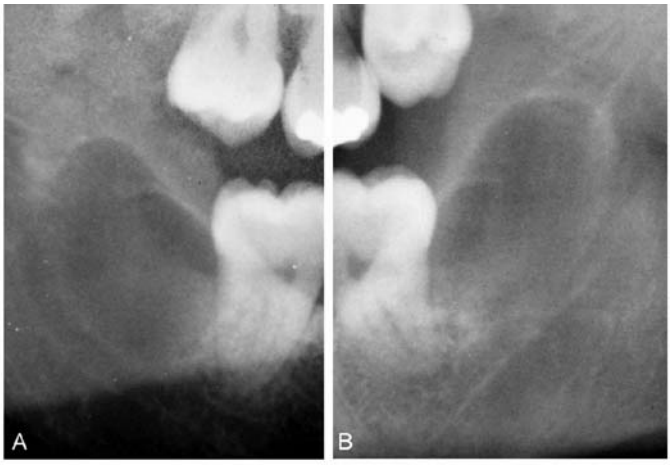

Figure 1. *Orthopantomogram at the time of admission, depicting the mandibular wisdom tooth and mandibular angle: (A) right side, (B) left side. Distal to the lower wisdom teeth, a bean-shaped translucency is depicted extending into the anterior part of the ramus on both sides. The lesions contain no radiopaque structures.*

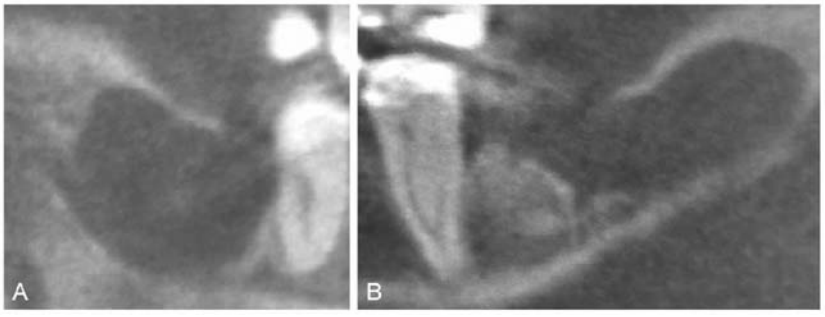

Figure 2. *Representative sagittal tomograms from cone beam tomography of the (A) right and the (B) left side. The radiotranslucency is sharply demarcated. A: Only the caudal border of the mandibular canal is depicted in continuity. B: The tomogram from a far lingual aspect (tooth 38 is not depicted), showing that the cyst is incompletely covered by bone.*

in the mandibular angle, such as keratocystic odontogenic tumour (5), ameloblastoma (13), myxoma (14), and even very early stages of a developing (supernumerary) tooth (15). However, neither odontogenic nor fibro-osseous structures were found in these truly cystic lesions. The cone-beam tomograms revealed a close connection between the teeth. However, these cysts did not show the radiographic appearance of dentigerous cyst (14). Furthermore, the dentition of the patient showed no apparent alteration, the inferior third molars had completely emerged and were situated in the expected position. The calculated centers of the cysts supported the hypothesis that the lesions originated in the anterior ramus distant to the crown of the third molars despite their narrow location at the distal roots of the lower third molars. Indeed, on both sides the re-ossification of the surgical defects following enucleation showed no alteration of the distal parts of the alveolar limbus.

Interestingly, these lesions developed symmetrically. Both the topography and the size of the lesions were almost equal. These findings supported the hypothesis of a developmental disorder. Indeed, epithelial tissue can be entrapped in the mandible during ontogenesis, derived from odontogenic epithelia (16) or other embryonic structures (17).

Lymphoid tissue adjacent to the lining stratified epithelium is a defining feature of LEC (12). The lymphoid tissues encircle the lesion in the majority of cases. However, only partial covering of the cysts by lymphoid tissues may also be noted (12, 18). Germinal centers are facultative findings in LEC of the oral cavity (12).

The pathogenesis of LEC has been discussed for a long period of time and is presently still debateable. The earlier terms of LEC, branchial cyst, branchial cleft cyst, branchiogenic cyst or pseudocyst (19) were abandoned in favour of the term 'lymphoepithelial cyst' following Bernier and Bhaskar's report on their observation that lymph nodes may contain inclusions of epithelial tissue that might undergo a cystic transformation resulting in a so-called

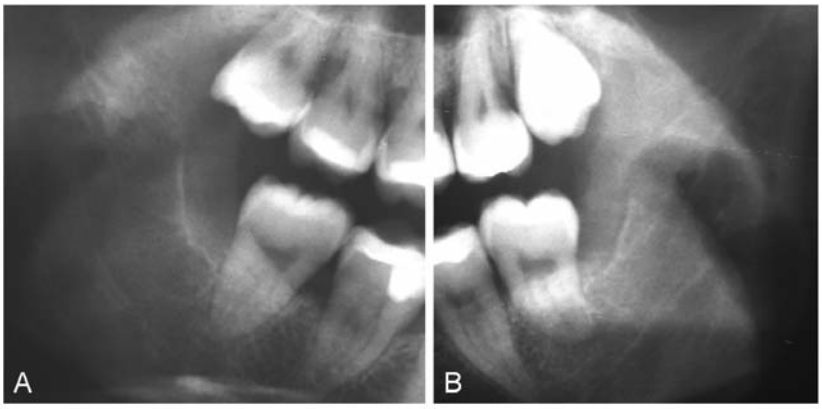

Figure 3. *Postoperative orthopantomogram, 2 years after surgery: (A) right side, (B) left side. The lesions are completely ossified.*

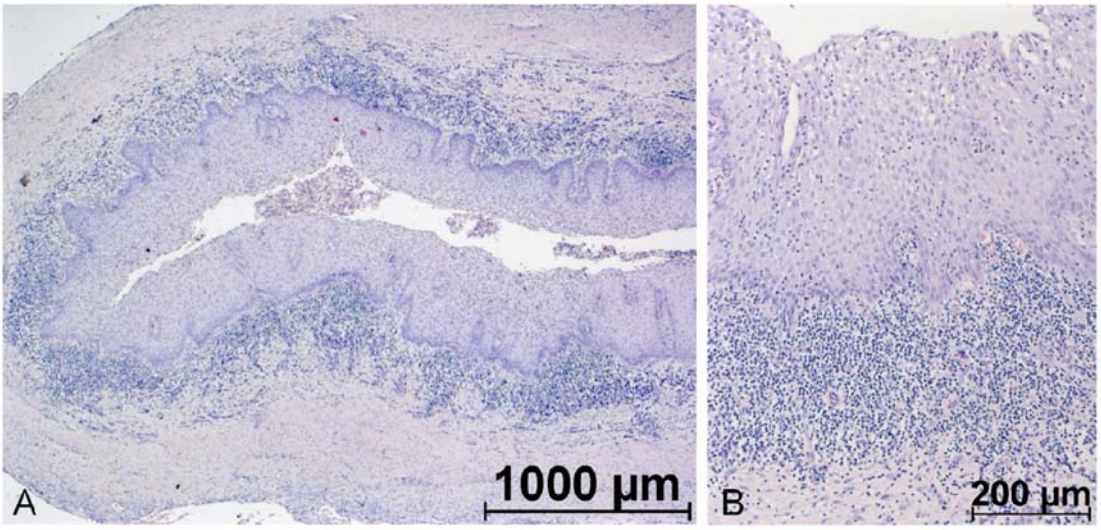

Figure 4. *Intraosseous lymphoepithelial cyst. A: Cystic cavity lined with squamous epithelium without atypia. Goblet cells or respiratory epithelium not present. No glandular or odontogenic structures within the cyst wall detected (original magnification: ×25). B: Dense subepithelial, band-like lymphocyte infiltration and scattered intraepithelial lymphocytes (original magnification: ×100). Both cysts showed identical histopathological structures.*

branchial cyst of the neck (19). Experimental studies supported the hypothesis that epithelia transplanted into lymph nodes are capable of cystic transformation (20, 21). However, this pathomechanism was not applicable to the current case of intraosseous LEC. Knapp suggested that an oral cavity LEC was a result of cystic obstruction of oral tonsils (22). However neither epithelial entrapment nor the obstruction theory was applicable in the current case.

Burkhardt (4) judged the term 'lymphepithelial cyst' to be unspecific due to the multiplicity of entities that might give rise to this phenotype, referring to the literature (12, 23). Consequently, alternative terms were proposed, such as benign cystic lymphoid aggregates (2). However, this terminology of benign oral epithelial lesions with prominent lymphoepithelial infiltrates is not widely used and is, at least in the presented case, a misnomer of a cystic lesion.

To the best of our knowledge, this report is the first detailing the occurrence of bilateral lymphoepithelial cysts in the mandible with no evidence of interference with odontogenesis.

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