



## Oral lymphoepithelial cyst in a young child

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### Abstract

*The oral lymphoepithelial cyst is an uncommon lesion in children with rather distinct clinical features. This case report describes the pertinent characteristics of this entity, involving the floor of the mouth in a 4 year-old boy. (Pediatr Dent 22:422-423, 2000)*

The oral lymphoepithelial cyst (LEC) is an uncommon developmental lesion that is frequently an incidental finding during a routine dental examination. An example of a symptomatic oral LEC in the floor of the mouth in a young child is described. The distinguishing features of this oral cyst are compared with other yellowish-white submucosal nodules that occur in this age group.

### Case history

A 4 year-old African-American boy was evaluated for a white enlargement of the ventral surface of the tongue, which occasionally was tender without an identifiable triggering factor. The soft tissue entity was first noticed a year ago when the child protracted the tongue (Fig 1). Minimal increase in lesion size was observed during that time period. The yellowish-white submucosal nodule, measuring 1 cm in diameter, had a smooth surface and was decorated with fine superficial capillaries (Fig 2). This freely movable, spongy enlargement had well defined borders and was located just left of the midline, involving the papilla, which was overlying the orifice of Wharton's duct. Palpation of the area produced mild tenderness without suppuration. There was no obvious obstruction of salivary flow from the ductal orifices, although mild lymphadenopathy of the mental and right submandibular nodes was detected. A cross sectional occlusal radiograph did not demonstrate a radiopaque focus in the floor of the mouth. Under deep sedation, an excisional biopsy was performed and the lesion was submitted for histopathologic examination.

### Clinical impression

Based on the clinical appearance, minimal growth and lack of significant ductal obstruction, this submucosal nodule is most consistent with an oral LEC. The etiopathogenesis of this cystic lesion is either developmental or reactive in nature (1, 2). Entrapment of the surface or ductal epithelium during embryogenesis within a lymphoid aggregate is thought to be the primary cause for this lesion. Another explanation includes the development of a cyst from an excretory duct of the sublingual or minor salivary glands, which stimulates a local immune response, resulting in lymphoid hyperplasia adjacent to the duct.



Fig 1. The lymphoepithelial cyst is clearly visible when the child protracted the tongue.

Most oral LECs are described as yellowish-pink to white submucosal nodules with a smooth surface that are 1 cm or less in size but range in size from a few millimeters to 2 cm. Typically asymptomatic, some lesions are tender, fluctuate in size and drain periodically. The contents of the cystic cavity vary from a creamy to cheesy white material. Although the oral LEC may develop wherever normal or accessory lymphoid tissue is found, the most common sites are the floor of the mouth, followed by the ventral and posterior lateral border of the tongue. This cystic entity is usually diagnosed in late adolescence or young adulthood; however, any age group may be affected. Between 8 to 20% of the biopsied cases from several studies were less than 18 years old, while the youngest child was reported to be 7 years old (1-4).

### Treatment and prognosis

Conservative surgical excision is usually recommended if the diagnosis is uncertain or if the lesion is symptomatic. Small lesions with a characteristic appearance may be periodically monitored and do not always require surgical removal. Recurrence is not expected and neoplastic transformation has not been reported in these oral lesions. In this particular case, the cystic nodule was removed because of periodic tenderness and the potential for partial obstruction of Wharton's duct due to its size and location.

### Microscopic findings

The histopathologic features of this lesion include a keratin-filled cyst that is lined by parakeratotic stratified squamous

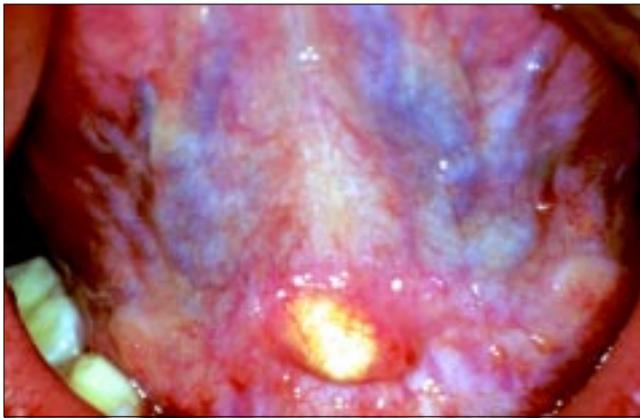


Fig 2. The submucosal nodule has a distinct creamy white surface with fine, superficial capillaries.

epithelium and occasional mucous cells. The wall of the cyst contains well demarcated lymphoid tissue with germinal centers and scattered macrophages and plasma cells.

### Differential diagnosis

Several yellowish-white submucosal nodules may occur in children in this location, including reactive, cystic and neoplastic conditions. Due to the lesion site and presence of symptoms, a sialolithiasis may resemble an oral LEC. A sialolith represents an organized structure within the salivary duct that develops due to the deposition of calcium salts around a nidus of mucus, bacteria, sloughed epithelial cells or foreign body. This entity presents as a white, submucosal nodule with a smooth surface that is often inflamed. The area is tender and hard to palpation and since it resides within the ductal structure, the salivary flow from the site is partially or totally obstructed. Wharton's duct of the submandibular gland is most often affected because of the thick mucoid secretions and anatomical position of the duct. Pain and swelling of the gland during mealtime are common complaints. In many cases, an occlusal radiograph is helpful in identifying this structure when it is located in the anterior floor of the mouth.

A soft tissue abscess frequently produces a pinkish-yellow soft tissue enlargement. Unless there is entrapment of a foreign body, suppurative sialadenitis or evidence of nonvital anterior teeth, this is an uncommon site for this inflammatory lesion. The sudden onset of pain with a purulent exudate and fluctuation in the size of the lesion help to distinguish this entity from most oral LECs.

The lipoma is a benign tumor of fat that infrequently occurs in the mouth. This submucosal nodule is characterized by a smooth surface with a yellowish hue. Similar to the oral LEC, superficial capillaries are easy to identify because they reflect through the pale background of this exophytic lesion. The most common intraoral site for the lipoma is the buccal mucosa, although the tongue and floor of the mouth may be involved. Lipomas are uncommon in children unless they are associated with Mohr syndrome, an orofacial-digital syndrome (5). These congenital lesions represent fatty hamartomas and occur on both the dorsal and ventral surface of the tongue.

The small dermoid cyst is included in the differential diagnosis because this developmental cyst has a marked predilection for the midline floor of the mouth. These pale, rubbery submucosal swellings may occur in infants and children but are most common in adults. Typically these lesions are asymptomatic, slow-growing enlargements that have a wide range in size. Secondary infection, both intraoral and cutaneous drainage, and dimpling with pressure are characteristics of large lesions. Continued growth of the dermoid cyst is an important distinguishing feature of an oral LEC that occurs in the midline floor of the mouth.

### Pediatric significance

Although the oral LEC is a rather distinct lesion, there are several pale, submucosal entities that may mimic this developmental condition. This case was interesting because it is one of the youngest reported cases and was associated with occasional tenderness. Partial obstruction of Wharton's duct was the suspected cause for the periodic pain, thus mimicking sialolithiasis. For this reason, surgical intervention of the cyst was recommended despite the young age of the child.

### References

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