Lymphoepithelial cyst of the nasogenian sulcus: a case report

C. Hupin*, B. Weynand** and Ph. Rombaux*

*Department of Otorhinolaryngology and Head and Neck Surgery and **Department of Pathology, Catholic University of Louvain, University Hospital St. Luc, Brussels, Belgium

Key-words. Cysts; nose; surgery; pathology

Abstract. Lymphoepithelial cyst of the nasogenian sulcus: a case report. A rare case of lymphoepithelial cyst formed in the nasogenian sulcus is reported. Lymphoepithelial cysts are comprised of a stratified squamous epithelial lining above dense lymphoid tissue. They are uncommon in the oral region and, to our knowledge, have never been reported in the nasogenian sulcus. Surgical excision was performed and no recurrence was noted after 6 months. In this report, we describe the etiopathogenesis of lymphoepithelial cysts, as well as the differential diagnosis of nasogenian sulcus swellings.

Introduction

Lymphoepithelial cyst refers to a cyst that is lined by metaplastic squamous or columnar epithelium permeated by lymphoid cells. The cyst wall is formed by lymphoid tissue with germinal centres. Although lymphoepithelial cysts have been reported at various sites, they are uncommon in the facial area and their etiopathogenesis is unclear. The purpose of the present report was to present an unusual case of a woman with a right nasogenian lymphoepithelial cyst.

Case report

A 73-year-old woman with an unremarkable medical history was referred to our department with painless swelling of the right nasogenian sulcus and upper lip. She had noticed the swelling in this area for one month. She also complained of purulent oral discharge.

Clinical examination revealed the presence of a round, elastic-soft mass in the nasolabial region. The floor of the nasal vestibule was elevated, the right upper lip was protuberant, and lateral displacement of the nasal ala was noted. Intraoral examination revealed a large fistula on the right gingivolabial sulcus. Computed tomography showed a cystic formation of $21 \times 27 \times 29$ millimetres in the right vestibule (Figures 1a,1b).

Surgical procedure was carried out under general anaesthesia, with orotracheal intubation. The anterior nasal cavity and upper gum were infiltrated with 1% Lidocaine containing 1:100,000 Epinephrine. Due to the large size of the cyst, we opted for a sparing lateral rhinotomy approach, rather than a vestibular approach (Figure 2). Adherence of the cyst wall to surrounding tissue was noted, although without attachment to the underlying bone. The cyst and oral fistula were removed during the same procedure. The content of the cyst was thick,
purulent, and had a nauseating smell.

The excised material was submitted for histopathological examination. The cyst wall was lined with respiratory epithelium containing focally goblet cells, which was surrounded by lymphoid tissue containing follicles and germinal centres in the submucosal layer (Figures 3a,3b). There were no malignant foci. These findings were consistent with a diagnosis of lymphoepithelial cyst. Bacteriology showed Streptococcus Milleri. The postoperative course was uneventful and local examination, 6 months post-operatively, revealed no recurrence of the lesion.

Discussion

Lymphoepithelial cysts are common in the neck region, considered as branchial cleft cysts. However, there have been many reports of lesions with the histologic features of branchial cysts in other sites, namely salivary glands, lung, mediastinum, tonsils, upper oesophagus, piriform sinus, mouth floor, pancreas, and thyroid. Histopathologically, the cyst lining is squamous or columnar and is surrounded by lymphoid tissue. The lymphoid tissue generally has a follicular pattern with germinal centres or a diffuse band like pattern. The lining epithelium has been described as stratified squamous or low columnar and evidence of keratinisation may be seen when stratified squamous epithelium is present. The presence of hair follicles, sebaceous glands,
sweat glands have been noted within the cyst.¹

To our knowledge, this is the first documented case of a lymphoepithelial cyst located in the vestibular region. On the basis of the histopathological resemblance to branchial cleft cysts, it is postulated this cyst is of branchial origin. It could correspond to a cyst of the first branchial cleft because of its upper position; although, this hypothesis is very unlikely due to its medial position (nasogenian sulcus). The histogenesis of branchial cleft cysts is controversial. Although various theories have been proposed, it is generally accepted that they are derived from the branchial apparatus with cysts arising from incomplete resolution of branchial remnants.¹¹

Anomalies of the first branchial cleft are uncommon and generally present as cysts, swellings, or fistulas located in a triangle defined by the hyoid bone below, the sternocleidomastoid muscle in the back, and the edge of the mandible in the front.¹²,¹³ The etiopathogenesis of lymphoepithelial cysts, located in places other than the neck region, remains unclear.

We suspected the cyst was in communication with the oral cavity because of the Streptococcus Milleri infection, typical bacteria found in the oral cavity. Oral fistula was probably secondary to the cyst’s infection. The differential diagnosis of cysts in the nasolabial region is broad and includes nasolabial cysts, nasopalatine duct cysts, dental or periodontal abscesses, odontogenic or non-odontogenic cysts, soft-tissue masses including benign tumours (schwannomas or adenomas), and malignant tumours of minor salivary glands.¹⁴ Although rare, lymphoepithelial cysts should also be considered.

**Conclusion**

We report here the case of a patient with a lymphoepithelial cyst located in the nasogenian sulcus. Due to its location and histopathology, we suspect it arose from an atypical anomaly of the first branchial cleft, although the exact origin is unknown. In conclusion, although lymphoepithelial cysts of the vestibule are very unusual, this entity should be considered during the differential diagnosis of nasolabial region swellings.

**References**