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Online Case Report

Unilocular cystic sebaceous lymphadenoma: a rare tumour

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A unique variant of the sebaceous lymphadenoma, so-called unilocular cystic sebaceous lymphadenoma, occurred in a 28-year-old male with a painless swelling in the left parotid region. The recognition of key histological features will readily allow differentiation of this unique neoplasm from its benign and malignant mimics. To our knowledge, out of 21 cases of sebaceous lymphadenoma reported, only 3 unilocular cystic variants have been recorded.

Keywords: Sebaceous glands – Parotid – Lymphadenoma

Sebaceous lymphadenoma is an exceptionally rare neoplasm, presenting as a progressively enlarging, painless mass largely in or around the parotid gland.[1] Sebaceous lymphadenoma is also reported in the anterior midline of the neck, lip minor salivary gland, and maxilla.[2,3] We herein describe a unique variant of sebaceous lymphadenoma, so-called unilocular cystic sebaceous lymphadenoma. To our knowledge, out of 21 cases of sebaceous lymphadenoma reported, only 3 unilocular cystic variants have been recorded.

Case report

A 28-year-old man presented with a painless swelling in the left parotid region for the past 5 years which had gradually increased in size. The medical and family history was not significant.

On examination, a diffuse swelling was seen extending from the infra-orbital region to the line extending from the angle of the mouth to the tragus. The swelling was non-tender and freely movable on palpation. Aspiration revealed pus-like material.

Ultrasound evaluation revealed a 3.8 x 2.4 x 3.5 cm, heterogeneous mass in the inner aspect of the parotid region. The mass was excised from an intra-oral approach, under general anaesthesia and submitted for histopathological examination (Fig. 1).

Grossly, the mass was well-circumscribed, tan-to-yellow in colour, ovoid in shape, unencapsulated, and contained irregular, smooth-lined cystic spaces on sectioning.

Microscopic examination revealed a unilocular cyst, with a focal lining of non-keratinising squamous epithelium, along with well-differentiated sebaceous glands in a lymphoid background (Fig. 2). Several areas of adipose tissue and remnants of salivary glands are also seen. There was evidence of moderate vascularity and chronic inflammatory cells along with areas of haemorrhage. Histopathology was suggestive of unilocular cystic sebaceous lymphadenoma.

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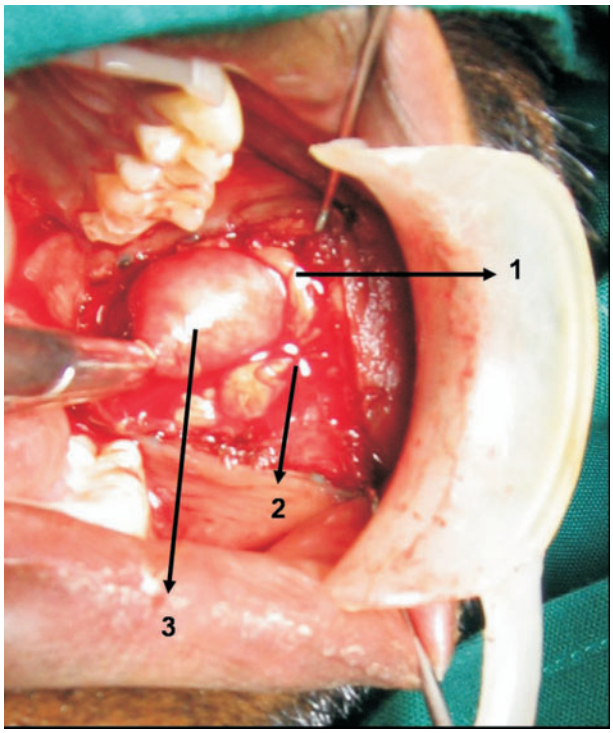

Figure 1 Photograph showing the buccal pad of fat (1), buccinator muscle (2) and the tumour mass (3).

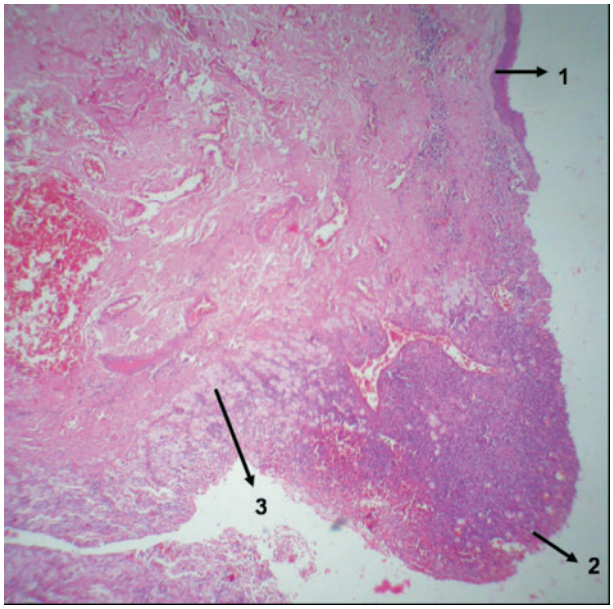

Figure 2 Photomicrograph showing a unilocular cyst, with a focal lining of non-keratinising squamous epithelium (1), along with well-differentiated sebaceous glands (2) in a lymphoid background (3). H&E staining.

Discussion

Sebaceous lymphadenoma is a slow-growing, asymptomatic neoplasm almost exclusively reported in the parotid gland.[4] A single case has been reported in the anterior midline of the neck. Most patients are 50 years or older at the time of diagnosis.[5] Men and women are almost equally affected. Our patient, however, was in his third decade, unlike most reported cases.

Grossly, sebaceous lymphadenoma presents as a well-circumscribed or encapsulated neoplasm, with the size ranging from 1.5–6.0 cm in its greatest dimension. On cross-section, sebaceous lymphadenoma presents as yellow, tan or a grey mass with a solid or microcystic surface.[5] Lesions occurring as solitary cysts are uncommon.[4] Most of these features were noticed in our case with the exception of being an unencapsulated, unicystic mass.

Microscopically, sebaceous lymphadenoma is composed of variably sized and shaped groups of sebaceous cells, salivary ducts and cysts in lymphoid background, the latter often demonstrating lymphoid follicles with germinal centres and sub-capsular sinuses.[6]

The pathogenesis of this lesion and nature of the lymphoid tissue in this lesion is controversial. One theory proposes that sebaceous lymphadenoma appears to arise from ectopic salivary gland tissue entrapped in lymph nodes during embryogenesis.[4] This observation is based on the fact that the ectopic salivary gland tissue in intraparotid or periparotid lymph nodes is a common finding. Additionally, the presence of sebaceous differentiation in some of the intranodal salivary gland inclusions and demonstration of a fibrous capsule with sinuses, lymphoid follicles and germinal centres in most of the tumours, support the presence of an underlying lymph node component.[3] Accepting this lymph node–salivary gland inclusion theory, one can explain the histogenetic relationship between these and several other parotid gland lesions. It is possible that parotid gland tissue entrapped within lymph nodes may undergo cystic degeneration to become a lympho-epithelial cyst, sebaceous differentiation to become a sebaceous lymphadenoma or sebaceous lymphadenocarcinoma, or oncocytic differentiation to become a Warthin's tumour.[6] However, there are some examples of Warthin's tumour and sebaceous lymphadenoma in which the lymphoid component does not appear to represent nodal tissue. It is possible that the lymphoid component represents a secondary reactive response to the epithelial proliferation as seen in other parotid gland tumours. This phenomenon was referred as tumour-associated lymphoid proliferation.

Sebaceous lymphadenoma may be mistaken for a low-grade muco-epidermoid carcinoma, especially in a small biopsy specimen. Mucin, if present, is confined to the ductal epithelial cells and ductal lumina in sebaceous lymphadenoma, and is never present within the sebaceous cells.[5] A foreign body giant cell reaction is also more commonly observed in sebaceous lymphadenoma than in muco-epidermoid carcinoma.

The treatment for sebaceous lymphadenoma is complete surgical excision. Considering the age of the patient and the tumour location, excision of the tumour was done from an intra-oral approach, sparing the parotid gland.

Conclusions

We report a rare case of unilocular cystic sebaceous lymphadenoma. To our knowledge, out of 21 cases of sebaceous lymphadenoma reported, only 3 unilocular cystic variants have been recorded. This particular case is worthy of attention due to its rarity and possible suggestions for the histogenesis of this tumour.

References

1. Boyle JL, Meschter SC. Fine needle aspiration cytology of a sebaceous lymphadenoma: a case report. *Acta Cytol* 2004; **48**: 551–4.
2. Maruyama S, Cheng J, Inoue T, Takagi R, Saku T. Sebaceous lymphadenoma of the lip: report of a case of minor salivary gland origin. *J Oral Pathol Med* 2002; **31**: 242–3.
3. Kurokawa H, Murata T, Fukuyama H, Kajiyama M. Sebaceous lymphadenoma in the midline of the maxilla: report of case. *J Oral Maxillofac Surg* 1999; **57**: 1461–3.
4. Merwin WH, Barnes L, Myers EN. Unilocular cystic sebaceous lymphadenoma of the parotid gland. *Arch Otolaryngol* 1985; **111**: 273–5.
5. Shukla M, Panicker S. Synchronous sebaceous lymphadenoma with squamous cell carcinoma: case report. *World J Surg Oncol* 2003; **24**: 30.
6. Tschen JA, McGavran MH. Sebaceous lymphadenoma: ultrastructural observations and lipid analysis. *Cancer* 1979; **44**: 1388–92.

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