

Sebaceous lymphadenoma of parotid gland: A case report of a unique presentation in an immunocompromised patient

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ABSTRACT

Sebaceous lymphadenoma is a rare, benign tumor of the parotid gland accounting for only 0.196% of all adenomas of the parotid gland. Our aim is to present a case of sebaceous lymphadenoma, which has been rapidly enlarging over a period of few months in an immunocompromised patient. This presentation is unusual for a benign salivary gland neoplasm. A 55-year-old female who is a known case of systemic lupus erythematous, antiphospholipid syndrome, and lupus nephritis, which have been treated by cyclophosphamide, presented with a 2-year complaint of fluctuating painless right parotid swelling, over the last 3 months the swelling started to progressively increasing in size. Physical examination showed a 4 × 3 cm firm, nontender mass in the right parotid gland. The facial nerve was intact and no cervical lymphadenopathy. Fine-needle aspiration cytology (FNAC) revealed marked chronic inflammation and was not helpful for diagnosis. Right superficial parotidectomy was performed without complications and there was no recurrence after 24 months of careful follow-up. In patients presenting with a rapidly enlarging parotid mass associated with an intact facial nerve, the possibility of sebaceous lymphadenoma should be considered as an important differential diagnosis in addition to other benign tumors of the parotid gland. The role of FNAC in this neoplasm is controversial.

Keywords: Diagnosis, FNA cytology, lymphadenoma, pathology, sebaceous lymphadenoma

Introduction

Sebaceous differentiation in salivary glands was first described by Hamperl in 1931.^[1] Neoplasms showing sebaceous derivation include sebaceous lymphadenoma, sebaceous carcinoma, sebaceous adenoma, and sebaceous lymphadenocarcinoma.^[2] Sebaceous lymphadenoma is a rare benign tumor of the parotid gland accounting for only 0.196% of all adenomas of the parotid gland. It is characterized clinically by slow-growing salivary gland mass and histologically by islands of epithelium showing sebaceous differentiation, which are distributed in a

hyperplastic lymphoid tissue. We present a case of sebaceous lymphadenoma of parotid gland, which has been rapidly enlarging and masquerading as a malignant growth, after taking the approval from the institutional review board in King Saud University Medical City.

Case Presentation

A 55-year-old female who is a known case of systemic lupus erythematous, antiphospholipid syndrome, and lupus nephritis, which have been treated by cyclophosphamide, presented with a 2-year complaint of a right parotid swelling, fluctuating in size and painless, over the last 3 months the swelling started to increase in size. Furthermore, there was no history of cutaneous neoplasms, visceral malignancies, or family member with similar presentation. Physical examination showed a 4 × 3 cm firm, nontender mass

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in the right parotid gland [Figure 1]. The facial nerve was intact and no cervical lymphadenopathy was appreciated.

Computerized tomography of the neck with contrast showed a well-defined oval-shaped heterogeneous and predominantly hypodense lesion arising from the right parotid gland and showing several internal small cystic areas with multiple foci of low-density enhancement which is likely due to the presence of fat [Figure 2 a and b]. Fine-needle aspiration cytology (FNAC) revealed marked chronic inflammation with florid histiocytic collections. Right superficial parotidectomy was performed without complications and there was no recurrence after 24 months of careful follow-up.

Gross inspection of the surgically excised specimen showed a parotid gland tissue measuring $5 \times 3 \times 2$ cm and weighing 16.7 g. The cut surface revealed a well-circumscribed, heterogeneous, solid, and cystic mass measuring $4 \times 3 \times 2$ cm, yellowish in color with focal sebaceous material [Figure 1]. Microscopic examination showed nests and clusters of benign sebaceous epithelium intermixed with a reactive lymphoid tissue [Figure 3].

Discussion

Sebaceous lymphadenoma is a rare benign salivary gland tumor. The name “sebaceous lymphadenoma” was given to this rare entity by McGavran *et al.*, 30 years after its first description.^[3] Parotid gland is the most common salivary gland site affected by this disease process. Rarely, other tumors have been reported to occur synchronously with sebaceous lymphadenomas; these include Warthin’s tumors, pleomorphic adenomas, oncocytomas, acinic cell adenocarcinomas, and basal cell adenomas.^[4,5] Sebaceous lymphadenoma is characterized histologically by the presence of islands of epithelium showing sebaceous differentiation, and these islands are surrounded by hyperplastic lymphoid tissue.

Sebaceous lymphadenoma of the parotid gland typically presents as a painless mass, with equal distribution in both genders and



Figure 1: Physical examination showed a 4 × 3 cm firm, nontender mass in the right parotid gland

favorable prognosis without recurrence after total excision. The age of presentation of sebaceous lymphadenomas ranges from 25 years to 89 years with the majority discovered after the age of 50 years. Neoplasms with coexisting areas of Warthin’s tumor and sebaceous lymphadenoma have been described, supporting the theory of common pathogenesis for these neoplasms.^[6] The origin of both sebaceous and oncocytic differentiation is the salivary ducts; both of these neoplasms occur mainly in the parotid glands and their lymphoid stroma contains well-developed follicles with germinal centers.^[6,7]

Sebaceous lymphadenoma is not usually correctly diagnosed by preoperative cytologic investigations in the majority of cases. A single case report suggested that the FNAC findings in a sebaceous lymphadenoma have accurately reflected the histological picture, although the neoplasm is rare to the point that the diagnosis might be missed.^[8] On rare occasions, sebaceous lymphadenoma can transform into a sebaceous lymph-adenocarcinoma; this is an extremely rare event and only five cases have been reported to date.^[9] To the best of our knowledge, our case is the only case in which the FNAC showed inflammatory process instead of neoplasm [Table 1]. The majority of published cases showed that sebaceous lymphadenoma rarely changed in size and most of the patients were immunocompetent. In our case, the parotid mass was rapidly enlarged over few months in an immunocompromised patient.

Conclusion

In patients presenting with a rapidly enlarging parotid mass associated with an intact facial nerve, the possibility of sebaceous lymphadenoma should be considered as an important differential diagnosis in addition to other benign tumors of the parotid gland. The role of FNAC in this neoplasm is controversial.

List of abbreviations

FNAC: fine-needle aspiration cytology.

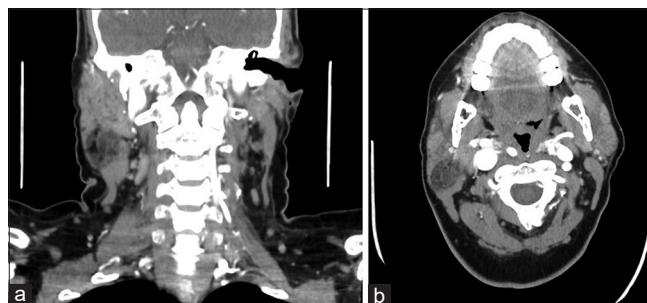


Figure 2: (a) Computerized tomography (axial) showed well-defined oval-shaped heterogeneous and predominantly hypodense lesion arising from the right parotid gland, showing multiple internal small cystic areas. (b) Computerized tomography of the neck with contrast showed a well-defined oval-shaped heterogeneous and predominantly hypodense lesion arising from the right parotid gland and showing several internal small cystic foci of low-density enhancement which is likely due to the presence of fat

Table 1: Characteristic of study participants in different studies

Preoperative diagnosis	Age (years)/Sex	Author	Preoperative investigation	Surgery	Follow-up
chronic inflammation	55/F	Our case (2015)	*FNAC and **CT scan	Superficial parotidectomy	NER at 24 months
Sebaceous lymphadenoma	56/F	Firt <i>et al.</i> (2000)	FNAC and CT scan	Superficial parotidectomy	NER at 12 months
Sebaceous lymphadenoma	75/M	Boyle and Meschter (2004)	FNAC	Excision (unspecified)	Not stated
Warthin's tumor	72/M	Hayashi <i>et al.</i> (2007)	FNAC	Superficial parotidectomy	NER at 16 months
Pleomorphic adenoma	57/M	Hayashi <i>et al.</i> (2007)	FNAC	Superficial parotidectomy	NER at 12 months
Pleomorphic adenoma	53/F	Kwon <i>et al.</i> (2002)	CT scan	Superficial parotidectomy	Not stated
Pleomorphic adenoma	68/F	Shukla and Panicker (2003)	FNAC	Total conservative parotidectomy	Not stated
Acinic cell adenocarcinoma	78/F	Mayorga <i>et al.</i> ^[5] (1999)	FNAC	Superficial parotidectomy	NER at 13 months
Mucoepidermoid carcinoma	65/F	Assor (1970)	Needle biopsy (unspecified)	Total parotidectomy	NER at 6 months
Warthin	16/M	Sun <i>et al.</i> (2009)	FNAC	Superficial parotidectomy	Not stated
Granulomatous inflammation	67/F	Maffani <i>et al.</i> (2007)	FNAC	Simple parotidectomy	Not stated
Lymphoid cell	80/F	While <i>et al.</i> (2010)	FNAC and CT scan	Surgical Excision	Not stated
Differential diagnosis included nonHodgkin lymphoma					
Insufficient for diagnosis	60/M	Majeed <i>et al.</i> ^[3] (2008)	FNAC and 28-02-2020 MRI	Superficial parotidectomy	Not stated
Aspiration revealed pus-like material	28/M	Chandrasekar <i>et al.</i> (2007)	FNAC	Not stated	Not stated
Warthin	60/M	Banich <i>et al.</i> (2007)	FNAC and MRI	Superficial parotidectomy	Not stated
Sebaceous lymphadenoma	57/M	Jazaerly <i>et al.</i> (2014)	FNAC and MRI	Superficial parotidectomy	NER at 20 months
Pleomorphic adenoma	73/M	Liu <i>et al.</i> (2014)	FNAC	Superficial parotidectomy	NER at 24 months
Pleomorphic adenoma	60/M	Liu <i>et al.</i> (2014)	FNAC	Superficial parotidectomy	NER at 36 months
Lymphadenoma	72/F	Liu <i>et al.</i> (2014)	FNAC	Superficial parotidectomy	NER at 36 months

*FNAC: fine-needle aspiration cytology; ** CT scan: computed tomography; 28-02-2020 MRI: magnetic resonance imaging

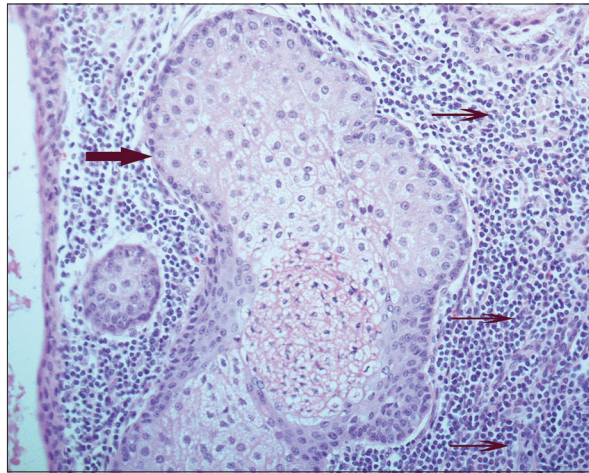


Figure 3: Photomicrograph: the sebaceous lymphadenoma shows benign nests and islands of bland sebaceous epithelium (thick arrow) intermixed with a prominent benign lymphoid component (thin arrows). Hematoxylin and eosin stain, original magnification 400x. Fine-needle aspiration cytology (FNAC) revealed marked chronic inflammation with florid histiocytic collections. Right superficial parotidectomy was performed without complications and there was no recurrence after 24 months of careful follow-up

Declarations

Disclosures

This paper was presented at the 7th Emirates Otorhinolaryngology

Audiology and Communication Disorders Congress in January 2107 as a conference talk with interim findings.

Ethical approval and consent to participate

Ethical approval for this case was not required according to the guidelines stated by the King Saud University Research Ethics Board.

Consent for publication

Consent to publish this work was obtained from the patient in writing and is available upon request.

Availability of data and materials

The data used in construction of this case report was obtained directly from the clinical chart. No software or databases were used. Anonymized raw data is available upon request.

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Competing interests

The authors declare that they have no competing interests.

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