Sebaceous Lymphadenoma of the Parotid Gland in a 13-Year-Old Girl: A Case Report

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Abstract Sebaceous lymphadenoma is a benign rare tumor of the salivary gland. It typically presents in the 6th decade of life as a slowly growing parotid mass. Pathologic examination reveals a well circumscribed mass composed of epithelial islands set in a lymphoid background. The epithelial nests are composed of sebaceous cells and ductal structures. Surgical excision is curative. Prognosis is good with rare recurrence or malignant transformation. Currently, there are less than 50 reported cases in the English literature. We report the second case of sebaceous lymphadenoma in the pediatric population and the youngest reported so far that presented in a 13 year old girl with a parotid mass.

Keywords Sebaceous lymphadenoma · Pediatric salivary tumor

Introduction

Sebaceous lymphadenoma (SLA) is a rare benign neoplasm of the salivary gland first described by Rawson and Horn in 1950 [1]. Less than 50 cases have been reported in the English literature. The great majority of these cases arise within the parotid gland, with rare reports involving minor salivary gland, and maxilla.[2–4] Clinically, it presents as a

slowly growing painless mass which may be present for many years, but has also been reported as an incidental finding [2]. The reported age at diagnosis ranges from 25 to 89 years with no sex predilection. There has been only one previous case reported in the pediatric population, occurring in a 16 year old boy [5]. Here we report a case in a 13 year-old girl, the youngest known case and only second case in a teenager.

Case Report

Clinical History

A 13-year-old Caucasian female with a history of cervical lymphadenopathy was referred to Nationwide Children's Hospital for excision of a left parotid mass. The patient had been seen approximately 5 months prior in the infectious disease clinic for evaluation of cervical lymphadenopathy. At that time, the patient also reported a 30 lb weight loss (over approximately 5 months). The patient denied any associated symptoms. The patient had no past history of hospitalization, serious infection, or other chronic illness. She reportedly had streptococcal pharyngitis in the remote past (about 4 years prior). There were no sick contacts or suspicious environmental exposures. The patients' medications were non-contributory, and she had no known drug allergies.

Physical exam was significant only for a left sub-mandibular mass (2×2 cm), without erythema, warmth, or tenderness to palpation; no other anterior or posterior cervical lymphadenopathy. There was no significant cervical, axillary, or inguinal lymphadenopathy. A complete blood count (CBC) showed no abnormalities. A monospot test was negative. EBV panels, performed on two separate

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occasions, were consistent with past EBV infection (i.e. EBV IgM negative but VCA IgG and nuclear antigen IgG positive). A sedimentation rate was normal. An extensive infectious disease workup revealed no evidence of an infectious etiology. Tests included a negative tuberculin skin test and negative serology for cat scratch disease (*Bartonella*), toxoplasmosis, aspergillus, *Blastomyces*, coccidioides, and histoplasma. Serum LDH was mildly decreased (388 U/L; reference range 400–1,250). Uric acid level was in the expected range. The clinical impression was that of a non-specific reactive left cervical submandibular lymphadenopathy.

In the interval period, the patient developed an enlarging left preauricular mass accompanied by ear pain, for which the patient was referred to Nationwide Children's Hospital. At this time there was no clinical evidence of the cervical submandibular lymphadenopathy described 5 months prior. A Computed-tomography (CT) scan of the head and neck demonstrated two predominantly solid well circumscribed enhancing masses: one in the superficial left parotid gland (3.9×2.9 cm) and one in the most superior portion of the gland (1 cm). There were small non-enhancing cystic foci scattered within the larger lesion (1–2 mm). The radiologic impression was “suggestive of a Warthin's tumor” (see Fig. 1).

The patient underwent left superficial parotidectomy with removal of a left jugular vein lymph node. The patient experienced no adverse events following surgery, and was discharged in stable condition.

Fig. 1 CT scan of the head and neck showing left parotid gland mass (arrow)

Pathologic Findings

Gross Examination

Three specimens were received: a partial parotidectomy specimen ($4 \times 2.7 \times 1.9$ cm) and two grossly unremarkable lymph nodes ($1.5 \times 1 \times 0.4$ and $1.3 \times 0.9 \times 0.5$ cm). The parotidectomy specimen had a pink lobular outer surface, which was inked black. Sectioning revealed a round well circumscribed homogenous white mass ($2.4 \times 2.3 \times 2.0$ cm) with a surrounding thin rim of fibrous tissue (0.1 cm in thickness) which grossly approximated the margin. A representative portion of the mass was snap frozen. The remainder of the specimen was entirely submitted in a sequential fashion.

Microscopic Description

One of the lymph nodes was submitted for frozen section and was histologically unremarkable. The second lymph node was also histologically unremarkable. Histologic sections of the parotid mass demonstrated a well circumscribed lesion with a rim of fibrous tissue and surrounding unremarkable serous salivary gland tissue. In some areas this rim of tissue was very thin, and focally the lesion was present at the inked margin in one slide. The lesion was comprised of lymphoid tissue with small epithelial islands of varying size and shape, some of which formed small cysts (see Fig. 2a, d). Within rare epithelial islands, there were groups of sebaceous cells (see Fig. 2a, c). The background lymphoid portion of the lesion had the appearance of a lymph node being comprised mainly of small lymphocytes with occasional germinal center formation. These findings are diagnostic of sebaceous lymphadenoma. There was no evidence of atypia or malignancy. Also, present within adjacent salivary gland parenchyma were two histologically unremarkable lymph nodes.

Discussion

Sebaceous lymphadenoma (SLA) is a rare benign salivary gland tumor, arising most commonly in the adult parotid gland. The current case is the second report of SLA in a teenager, and youngest case reported thus far. Grossly, these lesions are well circumscribed, 1–6 cm in diameter, with solid white-yellow cut surfaces. Microcystic or a unicystic lymphadenoma can be seen [2, 6]. Histopathologic examination reveals a well demarcated biphasic lesion composed of variably sized epithelial islands set within a diffuse lymphoid background with occasional germinal centers. The epithelial nests have small dark basally located cells and larger clear luminal sebaceous

Fig. 2 Sebaceous Lymphadenoma: **a** Low power view demonstrating lymphoid tissue with epithelial islands and cystic spaces at upper right (H&E. 12.5x). **b** Edge of lesion, with normal salivary gland at upper right (H&E. 50x). **c** Sebaceous cells within epithelial islands (H&E. 100x). **d** Cystic epithelial islands with scattered lining sebaceous cells (H&E. 100x)

cells with central nuclei and a vacuolated cytoplasm. Central cystic degeneration may be seen. Some of the epithelial nests represent salivary duct [2].

The etiopathogenesis of SLA is somewhat controversial. Postulated theories include origin from branchial cleft remnants, from ectopic salivary gland inclusions within intraparotid or periparotid lymph nodes, or from sebaceous metaplasia of the epithelial component of Warthin tumor [2, 5]. Gnepp et al. stated that the presence of germinal centers within the lymphoid background made it more likely that SLA derives from ectopic salivary gland tissue within lymph nodes [8]. Though, of course, the presence of germinal centers within a given lesion at any site does not necessarily equate to origin from a lymph node. There has been a single report of a combination tumor having features of both Warthin tumor and sebaceous lymphadenoma [9]. Sebaceous differentiation in salivary glands can be seen in up to 24.8% of normal parotid glands. The presence of this differentiation is thought to be related to hormonal influences as the frequency is much lower in childhood [1, 2]. This hormonal influence may also account for the rarity of SLA in the pediatric population.

The differential diagnosis for SLA includes mucoepidermoid carcinoma, sebaceous adenocarcinoma, lymphoepithelial carcinoma and lymphoma. The lymphoid component is an integral part of SLA and a diagnostic prerequisite, as in Warthin tumor. Mucoepidermoid carcinoma may have a background lymphoid component, but

the presence of mucinous and intermediate cells as well as the infiltrative nature rules out sebaceous lymphadenoma. Sebaceous carcinoma is also invasive, and lacks a prominent lymphoid component, being composed of sheets and/or nests of atypical and pleomorphic sebaceous cells. Unlike, Lymphoepithelial carcinoma, SLA is circumscribed with lack of mitotic activity or invasive growth. Lymphoma is differentiated by lack of circumscription with infiltration of surrounding tissue and displacement of normal ducts and acini [5, 7].

Sebaceous Lymphadenoma should be considered in the differential diagnosis of a salivary lesion with a predominant lymphoid background even in the pediatric teenage population. We present the youngest reported case and only the second reported case in a teenager. Complete surgical excision is the modality of therapy for SLA. Recurrence is rare following complete removal. Transformation to a sebaceous lymphadenocarcinoma has been reported but is extremely rare [10]. Synchronous acinic cell carcinoma, Warthin tumor and squamous cell carcinoma within parotid gland are rare reported occurrence [1, 5].

References

- Hayashi D, Tysome JR, Boye E, et al. Sebaceous lymphadenoma of the parotid gland: report of two cases and review of the literature. *Acta Otorhinolaryngol Ital.* 2007;27:144–6.

2. Maffini F, Fasani R, Petrella D, et al. Sebaceous lymphadenoma of salivary gland: a case report and a review of the literature. *Acta Otorhinolaryngol Ital.* 2007;27:147–50.
3. Maruyama S, Cheng J, Inoue T, et al. Sebaceous lymphadenoma of the lip: report of a case of minor salivary gland origin. *J Oral Pathol Med.* 2002;31:242–3.
4. Kurokawa H, Murata T, Fukuyama H, et al. Sebaceous lymphadenoma in the midline of the maxilla: report of a case. *J Oral Maxillofac Surg.* 1999;57:1461–3.
5. Sun G, Hu Q, Huang X, et al. Sebaceous lymphadenoma of parotid gland in a child. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2009;107(2):253–5.
6. Chandradekar T, Ramani P, Anuja N, et al. Unilocular cystic sebaceous lymphadenoma: a rare tumor. *Ann R Coll Surg Engl.* 2007;9(4):1–3.
7. Auclair PL. Tumor-associated lymphoid proliferation in the parotid gland. A potential diagnostic pitfall. *Oral Surg Oral Med Oral Pathol.* 1994;77:19–26.
8. Gnepp DR, Brannon R. Sebaceous neoplasms of salivary gland origin: report of 21 cases. *Cancer.* 1984;53:2155–70.
9. Dreyer T, Battmann A, Silberzahn J, et al. Unusual differentiation of a combination tumor of the parotid gland. A case report. *Pathol Res Pract.* 1993;189(5):577–81. discussion 581–585.
10. Croitoru CM, Mooney JE, Luna MA. Sebaceous lymphadenocarcinoma of salivary glands. *Ann Diagn Pathol.* 2003;7:236–9.