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Sialolipoma of the Lower Lip: Case Report and Literature Review

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Abstract: Sialolipoma is a relatively rare and fairly recently described as a variant of lipoma with salivary elements. Any site within the oral and maxillofacial region may be involved with the parotid gland being the most common location. Herein, we present a case of silaolipoma in lower lip. The clinical and histological features and differential diagnosis are discussed.

Keywords: Lipoma, Salivary gland, Oral cavity.

INTRODUCTION

Sialolipoma is a new histological variant of salivary gland lipoma, which is composed of adipose and glandular tissues. It was first described by Nagao *et al*., in 2001 [1]. The etiology of sialolipoma is not completely understood. It typically arises within the major salivary glands and the minor salivary gland of oral cavity. To the best of our knowledge only 35 cases of sialolipoma have been reported in English literature (Table**1** [1-21]) including the present case.

CASE REPORT

A 54-year-old Caucasian female was seen by her general dentist for evaluation of a painless swelling in her lower lip. There was no history of trauma or infection and the patient's medical history was unremarkable. An intra-oral examination revealed a 0.6x0.6cm soft tissue mass with normal overlying mucosa in her left lower lip, while an extra-oral examination revealed a normal facial morphology. The clinical differential diagnosis included mucocele, fibroma, lipoma, and salivary gland neoplasm. An excisional biopsy was performed and submitted to the Oral and Maxillofacial Pathology Department at University of Maryland, Baltimore. On gross examination, the mass was well-circumscribed, tan in color, soft in consistency and measured 0.6cm at its largest diameter. The histological examination revealed a mass of mature adipose tissue completely encapsulated by a fibrous band. Islands of salivary gland acini and ducts were located within the tumor. Neither atypia nor mitotic figures were observed in either the salivary glandular type tissue or the adipocytes. Mild lymphocytic infiltration and ductal dilation were seen (Fig. **1**). Consequently, the lesion was diagnosed as sialolipoma and no further treatment was required. The patient has been followed for 3 years without evidence of recurrence.

DISCUSSION

Sialolipoma, an uncommon variant of head and neck lipoma, is composed of proliferative adipocytes with entrapped normal salivary gland islands [1]. Almost any site within the oral and maxillofacial region may be involved with the parotid gland being the most frequently reported location [1-3, 6, 7, 9, 14, 21]. To our knowledge, 34 cases of sialolipoma have previously been reported in the English literature and eighteen of them were found in minor salivary glands [seven on the palate [1, 8, 10, 16, 17, 19], three in buccal mucosa [4, 10, 20], three on floor of the mouth [5, 11, 20], two on the tongue [4, 20], two in lower lip [12,the present case], and one on retromolar pad [20].

Clinically, sialolipomas usually present as a solitary painless palpable mass with an average size of 2.74 cm in diameter. Females are affected slightly more than males (with ratio 1.1:2). Patient's ages range from 6 weeks to 84 years, with average of 47.6 years. The duration of the lesion range from two months to ten years, with average of three years. In the present report, the lesion is in the lower lip and the diameter is 0.6 cm. Because lower lip is a preferable site of mucocele, it is probable that superficially located sialolipoma might be misdiagnosed clinically as mucocele. The other most common preoperative diagnoses are fibroma and salivary gland tumor. There is no distinguishable radiographic sign for sialolipoma in either computed tomography scan (CT) or magnetic resonance imaging (MRI) compared to a typical fatty lesion in the head and neck region [14].

Histological findings of haematoxylin and eosin staining in previous studies include a well circumscribed mass surrounded by a delicate fibrous tissue. The tumors are composed of mature adipose elements mixed with salivary gland

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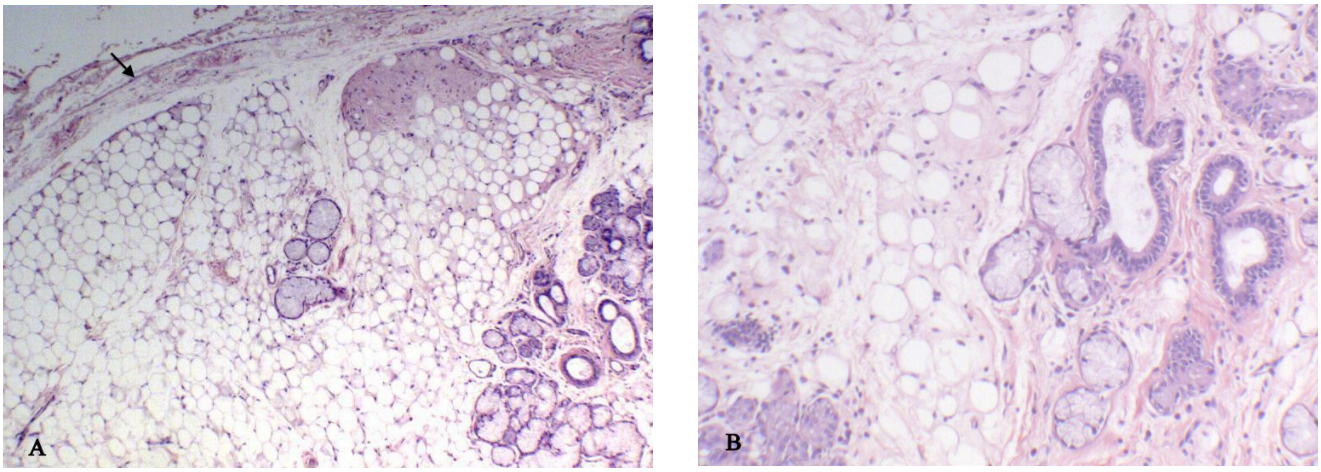

Fig. (1). (**A**) Photomicrograph showing islands of salivary gland tissue present within an adipose tissue tumor encapsulated by thin fibrous tissue (arrow) (hematoxylin and eosin, original magnification 4x); (**B**) Higher magnification revealing mild ductal dilatation with fibrosis within the tumor mass (hematoxylin and eosin, original magnification 10x).

Table 1. Clinical Features of 36 Cases of Sialolipoma

Author	Age (years)	Sex	Location	Size in cm	Duration	Treatment	*Follow-up*
Walts and Perzik, [2]	48	M	Parotid gland	3.5x2.5x1	NA	Superficial parotidectomy	*NED*
Walts and Perzik, [2]	65	M	Parotid gland	2.6 diameter	2 months	Superficial parotidectomy	*NED*
Baker *et al.,* [3]	44	M	Parotid gland	1.0 diameter	2 months	Superficial parotidectomy	*30 mo; NED*
Nagao *et al.,* [1]	20	M	Parotid gland	3.5x3.0x2.2	4 months	Superficial parotidectomy	*7 yr, 7 mo;NED*
Nagao *et al.,* [1]	45	F	Parotid gland	6.0x3.0x2.0	10 years	Superficial parotidectomy	*7 yr, 1 mo;NED*
Nagao *et al.,* [1]	67	M	Parotid gland	1.7diameter	2 months	Superficial parotidectomy	*3 yr,1mo; NED*
Nagao *et al.,* [1]	66	F	Parotid gland	6.0 diameter	5 months	Superficial parotidectomy	*2 yr,11mo;NED*
Nagao *et al.,* [1]	42	M	Parotid gland	6.0 diameter	10 years	Superficial parotidectomy	*1 yr 8 mo; NED*
Nagao *et al.,* [1]	66	M	Soft palate	2.2x1.5x1.5	6 years	Surgical excision	*11 mo; NED*
Nagao *et al.,* [1]	75	M	Hard palate	1.0 diameter	3 years	Surgical excision	*NA*
Fregnani *et al.,* [4]	NA	NA	Tongue	NA	NA	Surgical excision	*NED*
Fregnani *et al.,* [4]	NA	NA	Buccal sulcus	NA	NA	Surgical excision	*NED*
Lin *et al.,* [5]	67	F	Floor of the mouth	3.0x2.0	1 year	Surgical excision	*2 yr; NED*
Hornigold *et al.,* [6]	7 wk	F	Parotid gland	2.0x1.7x1.1	10 weeks	Surgical excision	*2 yr; NED*
Michaelidis *et al.,* [7]	44	M	Parotid gland	3.5 diameter	1 .5 years	Total parotidectomy	*2 yr; NED*
Sakai *et al.,* [8]	60	F	Hard palate	1.8x1.2x1.0	10 years	Surgical excision	*NED*
Kadivar *et al.,* [9]	3	F	Parotid gland	3.0 diameter	8 months	Superficial parotidectomy	*NA*
Ramer *et al.,* [10]	84	F	Buccal mucosa	1.0x1.0	NA	Surgical excision	*11 mo; NED*
Ramer *et al.,* [10]	43	F	Soft palate	2.0x2.0	NA	Surgical excision	*NA*
Ponniah *et al.,* [11]	70	M	Floor of mouth	2.0 diameter	NA	Surgical excision	*2 yr; NED*
De Freitas *et al.,* [12]	38	M	Lower lip	1.0 diameter	NA	Surgical excision	*NA*
Parente *et al.,* [13]	77	F	Submandibular gland	3.0x2.0x1.8	NA	Surgical excision	*22mo; NED*
Dogan *et al.,* [14]	33	M	Parotid gland	2.0x2.0	1year	Superficial parotidectomy	*NED*
Jang *et al.,* [15]	62	F	Submandibular gland	5.0 diameter	2-3years	Surgical excision	*17mo,NED*
Okada *et al.,* [16]	66	F	Hard palate	0.8 diameter	10 years	NA	*NA*

Table 1. contd…

Author	Age (years)	Sex	Location	Size in cm	Duration	Treatment	*Follow-up*
De Moraes *et al.,* [17]	72	F	Hard palate	2.0 diameter	2 weeks	Surgical excision	*8 mo; NED*
Sato *et al.,* [18]	3	M	Submandibular gland	4.0x3.0	NA	Surgical excision	*3 yr; NED*
Akrish *et al.,* [19]	52	M	Submandibular gland	3.5x2.0x1.5	NA	Surgical excision	*1yr; NED*
Akrish *et al.,* [19]	67	F	Hard Palate	5.0x4.0x4.0	NA	Surgical excision	*1yr; NED*
Nonaka *et al.,* [20]	27	F	Tongue	1.0x1.0	5 years	Surgical excision	*NA*
Nonaka *et al.,* [20]	73	F	Floor of mouth	4.0x1.0	NA	Surgical excision	*NA*
Nonaka *et al.,* [20]	65	F	Buccal Mucosa	2.0 diameter	2 years	Surgical excision	*NA*
Nonaka *et al.,* [20]	68	F	Retromolar pad	0.9 diameter	NA	Surgical excision	*14mo; NED*
Kidambi *et al.,* [21]	6 wk	M	Parotid gland	4.7x4.5x3.0	4 wk	Total parotidectomy with facial nerve dissection	*3 mo; NED*
Case report*,	54	F	Lower lip	0.6 diameter	NA	Surgical excision	*3 yr; NED*
Total number of cases=35	*Avg: 47.6*	*M:F 15:18*	*Parotid gland:13, Hard palate:5, Soft palate:2, Tongue :2, Floor of mouth:3, Buccal mucosa:3,Lower lip :2,submandibular gland:4, retromolar pad: 1*	*Avg:2.74*	*Avg: 3.04 years*		

*Present case report; NA, not available; NED, no evidence of disease.

tissues. The glandular components, consisting of acinar cells and ductal components, may be scattered through out the tumor or located in the periphery of the tumor [1, 5]. The 80 % of sialolipomas in major salivary gland are composed of adipose tissue while in minor salivary gland the glandular elements are clustered and evenly distributed around fat tissue [10,11]. No mitosis is seen in adipocytes or acinar and ductal cells [5]. The glandular components may be showed ductal dilation, oncocytic changes and squamous ductal metaplasia [1, 5-10]. In some cases areas of fibrosis are seen while myxoid changes are reported only in one case [9, 15]. Additionally, lymphocyte infiltration and enlarged congested vessels are reported [1, 10, 15].

The pathogenesis of the sialolipoma is not completely understood. However, immunohistological and ultrastructural studies confirmed that the glandular elements of the lesion could arise from entrapment of minor salivary gland during lipomatous proliferation rather than representing neoplastic process [1, 4, 5].

The morphologic differential diagnosis includes a variety of entities. Adenolipoma has histologic characteristic similar to sialolipoma; but it is composed of adipocytes and duct elements without acinar cells. Adenolipoma also differs from sialolipoma by the lack of organoid arrangement of the ductal type tissue [1, 5, 8]. Lipomatosis which typically occurs in older patients can be excluded by the microscopic lack of the fibrous capsule in addition to the absence of any medical condition associated with lipomatosis, for instance diabetes mellitus, malnutrition, chronic alcoholism and liver cirrhosis [1, 7, 9]. The distinction from pleomorphic adenoma is made by the presence of extensive fatty elements within the normal salivary gland tissue and lack of ducts and strands of dark-staining myoepithelial cells in sialolipoma [1, 5, 10].

Sialolipoma in the minor salivary glands is treated by complete surgical excision. However, most of tumors in parotid glands are treated with superficial parotidectomy. A complete parotidectomy with preservation of the facial nerve has been reported in two cases [7, 21] (Table **1**). Malignant transformation of sialolipoma has not been reported yet in the literature [5]. The follow up period ranged from 2 months to 10 years and there is no evidence of recurrent sialolipoma.

CONFLICT OF INTEREST

The authors confirm that this article content has no conflicts of interest.

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