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Case Report

Sialolipoma of the Right Submandibular Gland: A Case Report

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Abstract: Sialolipoma of the submandibular gland is very rare. We report a case of a 57-year-old woman who presented with a painless, slowly growing, mobile lump of the right submandibular gland. Clinical examination and FNA revealed chronic sialidenitis of the right submandibular gland. Exision was performed with uneventful postoperative course. The histology showed sialolipoma. Review of the 25 reported cases (including our case) of sialolipoma shows that this tumor is more common in the fifth decade of life, on the left side and the superficial lobe of parotid gland. It has a slight preference for men. Surgical excision is curative with minor complications and small recurrence rate. Histological examination is necessary to establish the diagnosis and to exclude malignancy.

Keywords: Sialolipoma, chronic sialidenitis, malignancy, submandibular gland

INTRODUCTION

Lipomatous tumors of the salivary glands are rare, accounting for 0.1-4.4% of all salivary gland tumors (Baker, S. E. *et al.*, 1981; & Nagao, T. *et al.*, 2001). In addition to the standard (pure) lipoma of the gland, other histological subtypes have been described, e.g., angiolipoma, fibrolipoma, sialolipoma, and liposarcoma (Starkman, S. J. *et al.*, 2013). Nagao *et al.*, first coined the term "sialolipoma" in 2001. This tumor is characterized by "a well-circumscribed mass composed of glandular tissue and mature adipose elements" (Nagao, T. *et al.*, 2001). At present, 51 cases of sialolipoma have been reported in the world literature; 25 in the parotid gland (Baker, S. E. *et al.*, 1981; Nagao, T. *et al.*, 2001; Starkman, S. J. *et al.*, 2013; Walts, A. E., & Perzik, S. L. 1976; Hornigold, R. *et al.*, 2005; Michaelidis, I. G. A. *et al.*, 2006; Kadivar, M. *et al.*, 2007; Bansal, B. *et al.*, 2007; Maiorano, E. *et al.*, 2008; Fritzsche, F. R. *et al.*, 2009; Dogan, S. *et al.*, 2009; Kidambi, T. *et al.*, 2012; Qayyum, S. *et al.*, 2013; Agaimy, A. *et al.*, 2013; Khazaeni, K. *et al.*, 2013; & Lee, P. H. *et al.*, 2014), six in the submandibular gland (Khazaeni, K. *et al.*, 2013; & Ahn, D. *et al.*, 2014), and the rest involved the minor salivary glands (Khazaeni, K. *et al.*, 2013; Ponniah, I. *et al.*, 2007; & Binmadi, N. O. *et al.*, 2012). We add one more case of saubmandibular gland sialolipoma.

CASE REPORT

A 57-year-old healthy woman presented with a 3-month history of painless slow-growing lump on the right side of the face. Her past medical history was unremarkable, and she denied any history of trauma or infection of the face. The swelling was 2 cm × 3 cm, nontender, mobile, soft and well-demarcated. The facial nerve was intact, and there were no other salivary swellings or cervical lymphadenopathy. The clinical impression was right submandibular gland sialadenitis. On ultrasound, the lesion was 4.0 cm × 2.7 cm, hypoechoic and well defined at the lateral aspect of the right submandibular gland with minimal vascularity. FNA suggested a suppurative lesion with occasional squamous cells. Excision was performed with uneventful postoperative course. The excised tumor was encapsulated, lobulated, tan-brown and measured 3.5 cm × 2.5 cm × 1 cm. The histological diagnosis was sialolipoma.

GROSS SPECIMEN

We received a nodular ,encapsulated soft tissue mass measuring 3.5x 2.5x 1cms.

Microscopy

Sections show salivary gland tissue with atrophy and numerous ectactic ducts and focal mild

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chronic inflammation along with areas of fibrosis and congestion. (Fig-1).

And hence a histological diagnosis of right submandibular gland sialolipoma was made and the amount of fat was approximately 90%. The patient remaines well 8 months after surgery with no recurrence(Fig-2).

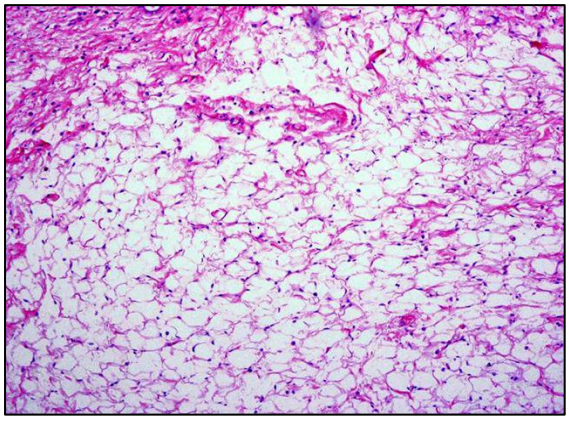

Fig-1.

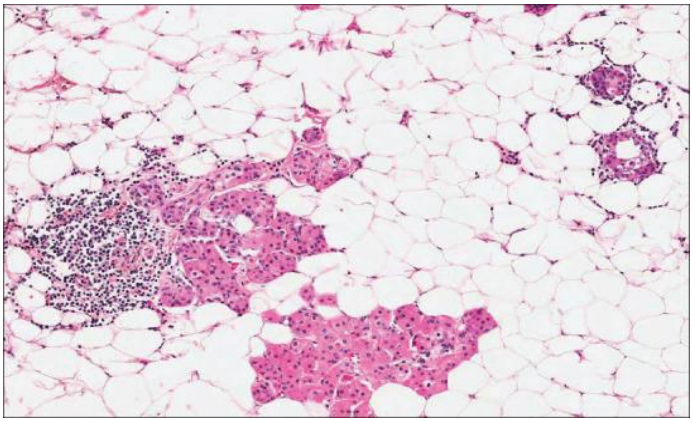

Fig-2.

DISCUSSION

Salivary gland lipomatous tumors are classified into several histological variants. The standard (true) lipoma is the most common type (Starkman, S. J. *et al.,* 2013; Walts, A. E., & Perzik, S. L. 1976; & Agaimy, A. *et al.,* 2013). Sialolipoma, on the other hand, is very rare, accounting for only 0.3% of all salivary gland tumors.[2] It is more frequently seen in the parotid gland (Qayyum, S. *et al.,* 2013; Agaimy, A. *et al.,* 2013; & Khazaeni, K. *et al.,* 2013). These cases are summarized in Table , except the case reported by Starkman *et al.,*. because of insufficient data in the original paper (Starkman, S. J. *et al.,* 2013). Although sialolipoma can affect children and adults, 20 (80%) patients in the literature were adults. Most of the adult patients were in the fifth decade of life (45%) with a mean age of 47 years (range: 18-74 years). The tumor has a small male preference; 52% of the patients were men. Salivary gland sialolipoma usually presents as a painless slowly growing soft mobile well-demarcated lump with intact facial nerve in an otherwise healthy patient. Similar to the pure lipoma, in parotid gland, the most likely preoperative diagnosis is a pleomorphic adenoma versus Warthin's tumor (Baker, S. E. *et al.,* 1981; & Hornigold, R. *et al.,* 2005). Routine laboratory tests are not helpful in establishing the diagnosis. Advanced imagings play the major role in the preoperative diagnosis of these lesions [Table 1]. Today, CT and magnetic resonance imaging (MRI) are the cornerstones for establishing the diagnosis. However, these imagings cannot differentiate sialolipoma from a pure lipoma. Our patients FNA suggested a chronic suppurative lesion . This falsification is supported by the literature, which indicates that in most instances FNA is not helpful in the preoperative work-up (Starkman, S. J. *et al.,* 2013; Michaelidis, I. G. A. *et al.,* 2006; Dogan, S. *et al.,* 2009; & Qayyum, S. *et al.,* 2013). We analysed that excision is the treatment of choice for sialolipomas of the salivary glands. This approach ensures complete excision of the tumor and excludes malignancy. As in other reports, our operative diagnosis was a sialadenitis. However, it was only during histological examination that the diagnosis of a sialolipoma was made. Because of its rarity, certain issues remain unanswered. In addition to the classical histological features of sialolipoma described by Nagao *et al.,* (2012) other features have been highlighted. These include the presence of sebaceous differentiation (Kadivar, M. *et al.,* 2007) nerve bundles (Michaelidis, I. G. A. *et al.,* 2006), oncocytic cells (Agaimy, A. *et al.,* 2013; & Ahn, D. *et al.,* 2014), duct ectasia, lymphocytic infiltration presence of sebaceous differentiation (Kadivar, M. *et al.,* 2007; & Bansal, B. *et al.,* 2007), and periductal fibrosis and inflammation (Agaimy, A. *et al.,* 2013). Therefore, more studies are needed to establish the full histological spectrum of sialolipomas. This is important to (1) explain their pathogenesis, clinical behavior and biological features, (2) establish a standard terminology and (3) determine the extent of surgical intervention.

Table.1 Summary of cases of sialolipoma of the salivary gland reported between 1976 and 2017

Case	Author[a]	Age	Sex	Duration of symptoms (months)	Side	Imaging	Site	Size (mm)	Operation	Follow-up (months)	Recurrence
1	Baker *et al.*[1]	44 years	Male	2	Right	None	SL	10 diameter	SP	30	No
2	Nagao *et al.*[2]	20 years	Male	4	Right	CT	SL	35×30×22	SP	91	No
3	Nagao *et al.*[2]	45 years	Female	120	Left	CT	SL	60×30× 20	SP	85	No
4	Nagao *et al.*[2]	67 years	Male	2	Right	CT	SL	17 diameter	SP	37	No
5	Nagao *et al.*[2]	66 years	Female	5	Left	None	SL	60 diameter	SP	35	No
6	Nagao *et al.*[2]	42 years	Male	120	Left	MRI	SL	60 diameter	SP	20	No
7	Walts and Perzik[4]	48 years	Male	NM	Left	NM	SL	35×25×10	SP	NM	No
8	Walts and Perzik[4]	65 years	Male	2	Left	NM	SL	26 diameter	SP	NM	No
9	Hornigold *et al.*[5]	7 weeks	Female	1.8	Left	US, MRI	SL	30×20	SP	24	No
10	Michaelidis *et al.*[6]	44 years	Male	18	Right	CT	DL	35 diameter	TP	24	No
11	Kadivar *et al.*[7]	3 years	Female	8	Left	NM	SL	30 diameter	SP	NM	NM
12	Bansal *et al.*[8]	11 years	Male	132	Left	US, CT	SL	70×70	SP	12	No
13	Maiorano *et al.*[9]	3 years	Female	36	Left	US, CT, MRI	SL	32×30×24	SP	24	No
14	Fritzsche *et al.*[10]	43 years	Male	NM	Right	US, MRI	SL	65×52	SP	NM	NM
15	Dogan *et al.*[11]	33 years	Male	12	Left	US, CT	SL	26×21×17	SP	NM	NM
16	Kidambi *et al.*[12]	6 week	Male	1.5	Left	US, MRI	SL, DT	50×50	TP	3	No
17	Qayyum *et al.*[13]	69 years	Male	60	Right	CT	SL	20×20	SP	NM	NM
18	Agaimy *et al.*[14]	74 years	Male	NM	Left	NM	NM	15 diameter	NM	23	No
19	Agaimy *et al.*[14]	18 years	Female	NM	NM	NM	NM	40 diameter	NM	93	No
20	Agaimy *et al.*[14]	49 years	Female	NM	Left	NM	NM	43 diameter	NM	NM	NM
21	Agaimy *et al.*[14]	47 years	Female	NM	Left	NM	NM	25 diameter	NM	NM	NM
22	Khazaeni *et al.*[15]	45 years	Female	NM	Right	US	SL	75×50×25	SP	12	No
23	Khazaeni *et al.*[15]	18 years	Female	12	Left	US	SL	50×40×30	SP	8	No
24	Lee *et al.*[16]	65 years	Female	4	Right	CT	SL	30×20	SP	3	Yes
25	Present case	38 years	Female	3	Left	US, CT	SL	28×22×19	SP	8	No

[a]Reference; SL – Superficial lobe; SP – Superficial parotidectomy; CT – Computed tomography; MRI – Magnetic resonance imaging; NM – Not mentioned; US – Ultrasound; DL – Deep lobe; TP – Total parotidectomy

CONCLUSION

Lipoma must be included in the differential diagnosis of a painless, slowly growing, mobile, soft lump of the salivary glands, especially parotid gland. CT and MRI are the cornerstones for establishing the preoperative diagnosis. However, surgical excision and histological examination are necessary to diagnose sialolipoma

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