

CASE REPORT

Sialolipoma of the Superficial Lobe of the Parotid Gland: A Case Report and Literature Review

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ABSTRACT

Sialolipoma of the parotid gland is very rare. We report a case of a 38-year-old woman who presented with a painless, slowly growing, mobile lump of the parotid gland. Computed tomography revealed lipoma of the superficial lobe of the parotid gland. Superficial parotidectomy was performed with uneventful postoperative course. The histology showed sialolipoma. Review of the 25 reported cases (including our case) of parotid sialolipoma shows that this tumor is more common in the fifth decade of life, on the left side and the superficial lobe. It has a slight preference for men. Its clinical presentation mimics the standard (pure) parotid lipoma and other more common benign parotid tumors particularly pleomorphic adenoma and Warthin's tumor. Surgical excision is curative with minor complications and small recurrence rate. Histological examination is necessary to establish the diagnosis and to exclude malignancy.

Key words: Parotid, sialolipoma, superficial parotidectomy

ملخص البحث :

يعتبر ورم الغدة النكافية من الأورام النادرة. يعرض الباحثون حالة لسيدة في الثامنة والثلاثين من عمرها تعاني من تضخم بطئ النمو وغير مؤلم في الغدة النكافية. وضحت الأشعة المقطعية ورم شحمي في الفص السطحي للغدة النكافية وقد تم استئصاله. أظهر الفحص المجهرى أنه من نوع ورم الغدة النكافية الشحمي. بينت الدراسات السابقة أن هذا النوع من الأورام يكثر في العقد الخامس من العمر وفي الجهة اليسرى ويكثر لدى الذكور. يعالج هذا النوع من الأورام بالاستئصال. ويعتبر الفحص النسيجي ضروريا للوصول للتشخيص النهائي لاستبعاد الحالات السرطانية.

INTRODUCTION

Lipomatous tumors of the parotid gland are rare, accounting for 0.1-4.4% of all parotid tumors.^[1,2] In addition to the standard (pure) lipoma of the parotid gland, other histological subtypes have been described, e.g., angioliipoma, fibroliipoma, sialoliipoma, and liposarcoma.^[3] Nagao *et al.* first coined the term "sialoliipoma" in 2001.^[2] This tumor is characterized by "a well-circumscribed mass composed of glandular tissue and mature adipose elements".^[2] At present,

51 cases of sialoliipoma have been reported in the world literature; 25 in the parotid gland,^[1-16] six in the submandibular gland,^[15,17] and the rest involved the minor salivary glands.^[15,18,19] We add one more case of parotid sialoliipoma and review the literature.

CASE REPORT

A 38-year-old healthy woman presented with a 3-month history of painless slow-growing lump on the left 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 left parotid swelling, most likely to be a pleomorphic adenoma or Warthin's tumor. On ultrasound, the lesion was 2.0 cm × 1.7 cm, hypoechoic and well-defined at the lateral aspect of the left parotid gland

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with minimal vascularity suggestive of pleomorphic adenoma. Computed tomography (CT) with intravenous contrast revealed a 2.8 cm × 2.2 cm × 1.9 cm, well-demarcated, macro-lobulated hypodense lesion with no significant contrast enhancement [Figure 1]. The diagnosis was lipoma of the superficial lobe of the left parotid gland. Left superficial parotidectomy with facial nerve dissection and preservation was performed with uneventful postoperative course. The excised tumor was encapsulated, lobulated, tan-brown and measured 4.8 cm × 4.0 cm × 1.2 cm. The histological diagnosis was sialolipoma [Figure 2] and the amount of fat was approximately 90%. The rest of the superficial lobe was normal. The patient remained well 8 months after surgery with no recurrence.

DISCUSSION

Parotid lipomatous tumors are classified into several histological variants. The standard (true) lipoma is the most common type.^[3,4,14] 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.^[13-15]

All the cases of parotid sialolipoma are written in English,^[1-9,11-16] except one in German.^[10] These cases (including our case) are summarized in Table 1, except the case reported by Starkman *et al.* because of insufficient data in the original paper.^[5] Although parotid 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. Data on duration of symptoms was available for 18 patients; the mean duration was 30 months (range: 1.5-132 months). Information on laterality of the tumor was available for 24 patients; 16 (66%) lumps involved the left side and 8 (33%) the right side. The tumor was located in the superficial lobe in 19 patients, in the deep lobe and both lobes in one patient each, and in four patients the location was not specified. The size was variable. The largest tumor was 75 mm in its greatest dimension, while the smallest was 10 mm in diameter. Follow-up data was available for 17 patients. The mean duration of follow-up was 31.2 months (range: 3-93 months). The outcome of surgery was good with minor complications.^[6,8,12] Table 1 shows that none of the patients but one had recurrence.^[16]

Parotid sialolipoma usually presents as a painless slowly growing soft mobile well-demarcated lump

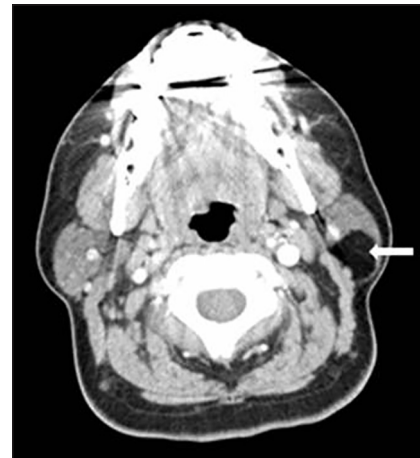


Figure 1: Contrast-enhanced axial computed tomography scan of upper neck showing 28 mm × 22 mm well-demarcated, encapsulated lipoma of the superficial lobe of the left parotid gland (arrow). The right parotid gland is normal

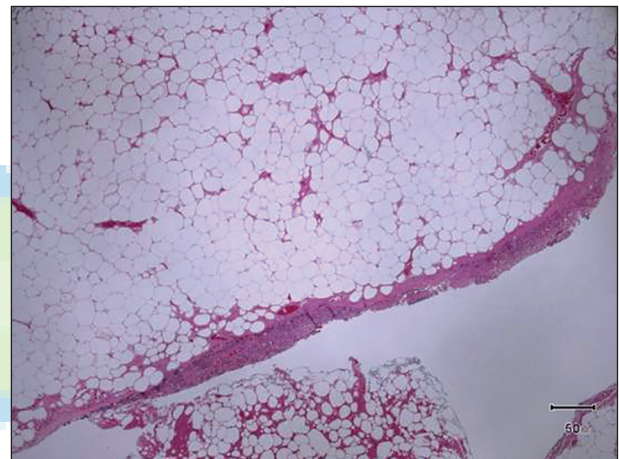


Figure 2a: An outer fibrous capsule and mature adipose tissue (H and E, ×100)

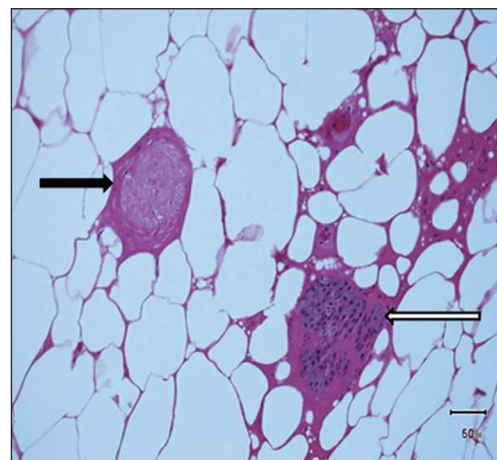


Figure 2b: Mature adipose tissue encircling salivary acini (white arrow) and nerve bundle (black arrow) (H and E, ×200)

with intact facial nerve in an otherwise healthy patient. Similar to the pure lipoma, the most likely preoperative

diagnosis is a pleomorphic adenoma versus Warthin's tumor.^[1,4] 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. In our case, once the CT diagnosis of a lipoma was made, other diagnostic tools, such as fine-needle aspiration (FNA) were not utilized. This approach is supported by the literature, which indicates that in most instances FNA is not helpful in the preoperative work-up.^[3,6,11,13] We believe that superficial parotidectomy is the treatment of choice for sialolipomas of the superficial lobe of the parotid. This approach ensures complete excision of the tumor and excludes malignancy. As in other reports, our operative diagnosis was a lipoma. 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.*,^[2] other features have been highlighted. These include the presence of sebaceous differentiation,^[7] nerve bundles,^[6] oncocyctic cells,^[14,17] duct ectasia, lymphocytic infiltration,^[7,8] and periductal fibrosis and inflammation.^[14] As with Michaelidis *et al.*,^[6] the microscopic features in our case included the presence of nerve fibers. However, in contrast to Michaelidis *et al.*,^[6] we did not encounter any difficulty in the identification of the facial nerve and excision of the tumor. 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.

CONCLUSION

Lipoma must be included in the differential diagnosis of a painless, slowly growing, mobile, soft lump of the parotid gland. CT and MRI are the cornerstones for establishing

Table 1: Summary of cases of sialolipoma of the parotid gland reported between 1976 and 2014

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

^aReference; SL – Superficial lobe; SP – Superficial parotidectomy; CT – Computed tomography; MRI – Magnetic resonance imaging; NM – Not mentioned; US – Ultrasound; DL – Deep lobe; TP – Total parotidectomy

the preoperative diagnosis. However, surgical excision and histological examination are necessary to diagnose sialolipoma.

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