DE GRUYTER OPEN

10.2478/AMB-2018-0007

CASE REPORTS

SIALOLIPOMA OF THE PAROTID GLAND: AN UNCOMMON LIPOMA VARIANT OF SALIVARY GLAND

M. H. A. Ghafar[1], G. Ju Tuang[2], N. M. Y. Mohammad[2], Ch. Nadarajan[3], B. Abdullah[1]

[1]Department of Otorhinolaryngology – Head & Neck Surgery, School of Medical Sciences, University Sains Malaysia
[2]Department of Otorhinolaryngology – Head & Neck Surgery, Hospital Kuala Lumpur, Jalan Pahang, Malaysia
[3]Department of Radiology, School of Medical Sciences, University Sains Malaysia

Abstract. *Sialolipoma affecting the salivary glands is an uncommon lipoma variant composed of mature adipose tissue mixed with acinar, ductal, basal and myoepithelial cells of normal salivary gland. It has a wide age range at presentation. Generally, the major gland has male predilection while the minor gland has a female preponderance. A diagnostic workup requires CT or MRI to define the exact location and texture of the lesion. Surgical excision is recommended when the tumor progressively increases in size. Prognosis is good whereby no malignant transformation or recurrences has been reported.*

Key words: *parotid, sialolipoma, salivary gland*

Corresponding author: *Prof. Dr. Baharudin Abdullah, Department of Otolaryngology – Head and Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, 16150 Kubang Kerian, Kelantan, Malaysia, Tel: +6097676416, Fax: +6097676424, e-mail: baharudin@usm.my, profbaha@gmail.com*

INTRODUCTION

The term sialolipoma was first introduced by Nagao et al. [1]. In their work, Nagao and colleagues [1] had established that sialolipoma is a distinctive histological variant of lipoma characterized by benign salivary gland parenchyma intimately admixed with mature adipose tissue. The term sialolipoma was accepted in the 2005 World Health Organization (WHO) Classification of Head and Neck Tumors [2]. Since then, there have been reported more than 60 cases [1-11].

Although a lipoma is a common benign soft tissue tumor and frequently developed in the head and neck region, it rarely occurs in the salivary gland. Sialolipoma accounts for less than 0.5% of all neoplasm of the parotid gland [3].

CASE PRESENTATION

A 40 years old female patient presented with swelling over the left parotid region for three years. The swelling had been progressively increasing in size, with no other associated symptoms. She denied dysphagia, odynophagia, hoarseness and facial numbness. There was also the absence of a history of constitutional symptoms, nasal obstruction, epistaxis or hemoptysis. Examination revealed a well-demarcated solitary soft mass of 2 cm x 2 cm in size, which was not tender, mobile on palpation and overlying the parotid tail region. Systemic examination of the head and neck region was otherwise normal.

She underwent two fine needle aspiration procedures, but the cytopathological results were both inconclusive. Computed tomography reveals a fairly well-defined lesion with Hounsfield unit (HU) of fat density at the posterolateral aspect of the left parotid tail (Figure 1). The lesion had no demonstrable contrast enhancement. It, however, exerts mass effect on the underlying deep cervical fascia. No extension to the adjacent structures was noted. Radiological features were consistent with lipoma of the parotid gland.

As she was concerned with the increase in the size of the mass, she was treated with right superficial parotidectomy with facial nerve conservation. Examination of the gross specimen showed a pale yellowish soft mass measuring 38 mm x 30 mm x 20 mm overlying the left parotid tail region. Microscopic examination revealed mature adipocytes surrounding salivary ducts and acini (Figures 2 and 3). The final diagnosis of sialolipoma over the left parotid gland was, therefore, being made.

She was followed-up one week after the surgery and a left facial nerve palsy grade 4 was observed. During the second follow-up one month later, the facial nerve palsy had improved to grade 3. In both of the follow-up sessions, there was no evidence to suggest recurrence of the disease. The patient defaulted her follow-up after that.

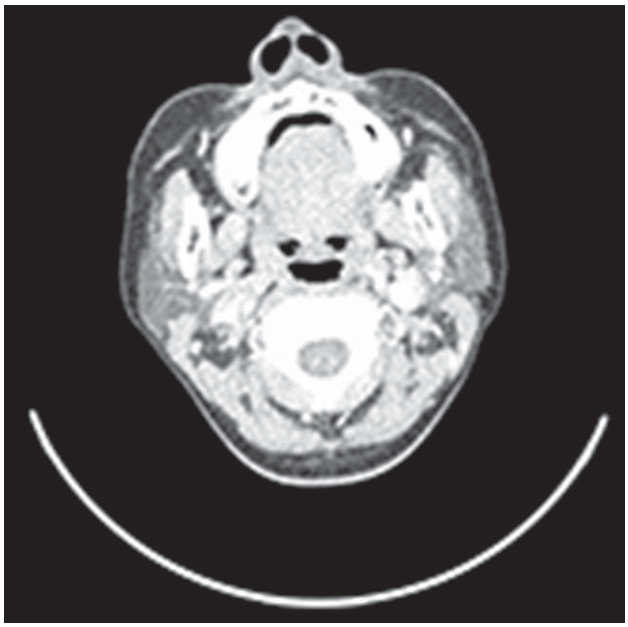

Fig. 1. CT of the neck with contrast revealed increased subcutaneous fat over the left parotid tail with mild mass effect onto the underlying deep cervical fascia

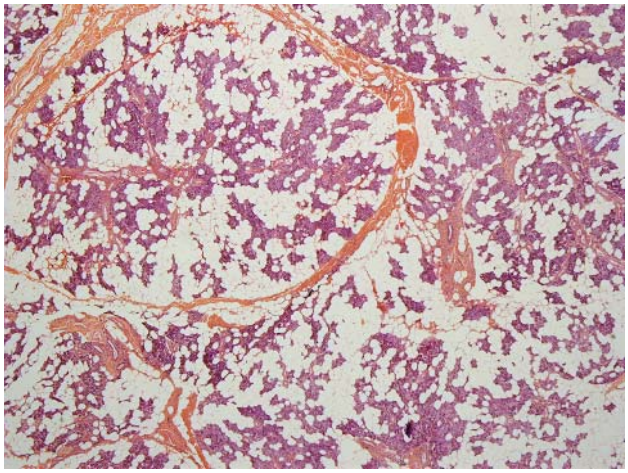

Fig. 2. Image showing glandular element with fat cells [H & E, 40X]

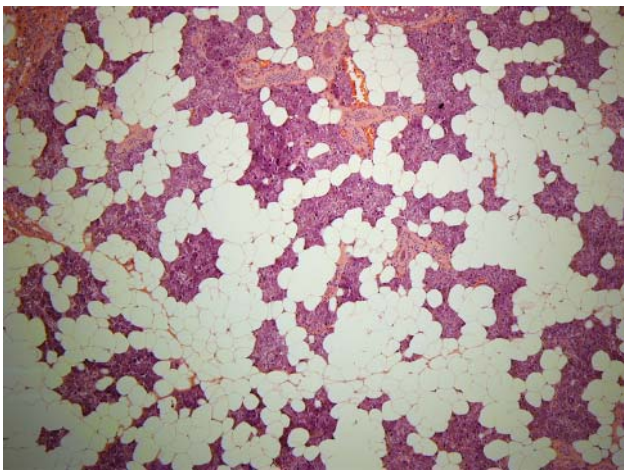

Fig. 3. Image showing adipose cells with salivary gland tissue [H & E, 100X]

DISCUSSION

Earlier work by Nagao et al. [1] demonstrated that sialolipoma shows a slight male predilection (male: female ratio = 1.75:1). Later publications with bigger sample size managed to establish the demographical differences between patients suffering sialolipomas affecting the major and minor salivary glands [4-7]. Major salivary gland sialolipoma is more frequent in males than females (male: female ratio = 2:1.3) with an average age at the time of diagnosis of 42 years (standard deviation ± 25). Minor salivary gland sialolipoma showed female preponderance (1.8:1 female: male ratio) with age average at the time of the diagnosis of 60.3 years (standard deviation ± 15.5) [7].

Generally, sialolipoma is commonly seen in an adult with wide age of distribution between 27 to 84 years (mean, 61.6 years) [4]. Nevertheless, it may also occur in children. Kadivar and colleagues [3] reported the first case of congenital sialolipoma in a 3-year-old female infant who also had a family history of congenital anomalies and a single palmar crease. Bansal B et al. [8] reported the second case of congenital sialolipoma in an 11-year-old boy presented with a gradually increasing mass (since birth) in the left parotid region.

Of all the salivary glands, the parotid gland is the most common site affected. Other anatomic sites include submandibular gland, soft palate, hard palate, tongue, buccal mucosa, floor of the mouth, buccal sulcus, retromolar pad and lower lip [1, 4-7]. There was no report of multifocal sialolipoma or bilateral occurrence.

The typical presentation of sialolipoma is slow-growing, soft, mobile and painless swelling at the affected glands. Symptoms may be present for a long time, from 2 months to 11 years [4-7].

Upon microscopic examination, sialolipoma exhibit a unique feature; a well-circumscribed, often encapsu-

lated mass, composed of benign, neoplastic, adipose tissue with scattered foci of generally atrophic, non-neoplastic, salivary gland acini and ducts contained within the lipomatous proliferation [4]. The fatty component predominates over the epithelial elements (> 20-90%). Another common finding in sialolipoma is foci of sebaceous metaplasia commonly associated with variable periductal inflammation and fibrosis. However, oncocytes are usually absent [12]. All the above characteristics differentiate sialolipoma from other lipomatous lesions such as true lipoma, adenolipoma, lipomatosis, fibrolipoma, spindle cell lipoma and pleomorphic adenoma with an extensive fat constituent [1-8, 11-12].

The pathogenesis of sialolipoma is still not clearly understood. One hypothesis proposed by Akrish et al. [9] is that the pathogenesis of sialolipoma may be associated with some form of salivary gland dysfunction, leading to altered salivary gland configuration.

Both CT and magnetic resonance imaging (MRI) are useful in characterizing lipomatous lesion of the parotid gland and help in differentiating it from common differential diagnosis of sialolipoma such as lipoadenoma, pleomorphic adenoma, and well-differentiated liposarcoma. These imaging modalities are found to be superior to ultrasonography in defining the exact location and texture of the lesion [7, 11]. MRI, however, provides a better soft tissue differentiation. It is also more accurate in the evaluation of the tumoral extension and characterization of uncommon lipomatous lesion of the head and neck. The use of intravenous gadolinium will better depict the margin of the lesion and outlines its vascular supply [10]. Large lesion (> 5 cm), rapid enlargement, significant enhancing soft tissue components, thick or nodular septa, a location deep to the fascia are red flag features of malignant lesion that should be kept in mind in the diagnosis of lipoma [13].

In general, sialolipoma appears as a well-circumscribed lesion during high-resolution CT or a high-intensity MRI [11]. On the other hand, Cappabianca et al. [10] reported 2 cases of sialolipoma of the parotid gland in which both CT and MRI showed no characteristic signs. The radiological interpretation was “simple lipoma”, but histology revealed sialolipoma [10].

Fine needle aspiration, which is the first line procedure in diagnosing major salivary gland lesion offers little diagnostic value, as its accuracy is less than 50% in lipomatous tumors [7]. In addition, the result obtained may not be conclusive and offers little value for the busy clinicians in their decision for any definitive management. Therefore, the combination of fine-needle aspiration (both positive and negative findings) and CT or MRI findings should be taken together for the clinician to consider the probability of a lipomatous tumor of the parotid gland. However, it should be pointed out that both modalities cannot distinguish between benign and malignant lesions with certainty.

The recommended treatment of sialolipoma is surgical excision. Most of the reported cases showed no local recurrence and no malignant transformation [1, 3-11].

CONCLUSION

Sialolipoma of the parotid gland is an uncommon tumor and clinicians may not be aware of this distinct entity. Diagnosis is set mainly by imaging studies, of which CT or MRI has shown an excellent accuracy in defining the location and texture of the tumor. The recommended treatment of sialolipoma is surgical excision whereby no malignant transformation or recurrences have been reported.

Consent: *The authors were not able to reach the patient for the consent. However, none of the images used in the manuscript can be used to identify the patient.*

Conflicts of interest: *There are no conflicts of interest.*

REFERENCES

1. Nagao T, Sugano I, Ishida Y et al.. Sialolipoma: a report of seven cases of a new variant of salivary gland lipoma. Histopathology 2001; 38: 30-6.
2. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. Lyon: IARC Press, 2005; 275.
3. Kadivar M, Shahzadi SZ, Javadi M. Sialolipoma of the parotid gland with diffuse sebaceous differentiation in a female child. Pediatr Dev Pathol 2007; 10: 138-41.
4. De Moraes M, de Matos FR, de Carvalho CP et al. Sialolipoma in minor salivary gland: case report and review of the literature. Head Neck Pathol 2010; 4: 249-52.
5. Khazaeni K, Jafarian AH, Khajehahmadi S et al. Sialolipoma of salivary glands: Two case reports and review of the literature. Dent Res J (Isfahan) 2013;10: 93-7.
6. Qayyum S, Meacham R, Sebelik M, Zafar N. Sialolipoma of the parotid gland: Case report with literature review comparing major and minor salivary gland sialolipomas. J Oral Maxillofac Pathol 2013; 17: 95-7.
7. Huerta EL, Rivera DQ, Rocha FT et al. Sialolipoma of minor salivary glands: presentation of five cases and review of the literature with an epidemiological analyze. Indian J Otolaryngol Head Neck Surg 2015; 67: 105-9.
8. Bansal B, Ramavat AS, Gupta S et al. Congenital sialolipoma of parotid gland: a report of rare and recently described entity with review of literature. Pediatr Dev Pathol 2007; 10: 244-6.
9. Akrish S, Leiser Y, Shamira D, Peled M. Sialolipoma of the salivary gland: Two new cases, literature review, and histogenetic hypothesis. J Oral Maxillofac Surg 2011; 69:1380-4.
10. Cappabianca S, Colella G, Pezzullo MG et al. Lipomatous lesions of the head and neck region: imaging findings in comparison with histological type. Radiol Med. 2008; 113: 758-70.
11. Lee PH, Chen JJ, Tsou YA. A recurrent sialolipoma of the parotid gland: A case report. Oncol Lett. 2014; 7: 1981-3.
12. Agaimy A. Fat-containing salivary gland tumors: a review. Head Neck Pathol 2013; 7: 90-6.
13. Razek AA, Huang BY. Soft Tissue Tumors of the Head and Neck: Imaging-based Review of the WHO Classification. Radiographics. 2011; 31: 1923-54.