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A rare case of recurrent congenital sialolipoma of parotid gland in a 3-year-old child: A case report and review of literature

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ABSTRACT

INTRODUCTION: Sialolipoma is an extremely rare salivary gland tumor characterized by a well circumscribed mass composed of glandular tissue and matures adipose elements. Herein, the aim of this article is to report the sixth case of congenital sialolipoma and the first case of recurrent congenital sialolipoma in infant, and discuss the clinicopathological and morphological features of sialolipoma and the possible cause of its recurrence.

CASE REPORT: A 3 year-old child presented with a recurrent mass of right parotid gland which progressed from birth, initially treated at the age of 4 months by simple tumorectomy and excision of the surrounding parotid tissue. The tumor recurred 4 months postoperatively. The radiological examination confirmed the parotid origin of the tumor. Histopathology was consistent with a sialolipoma. A superficial parotidectomy with preservation of the facial nerve was performed this time at the age of 3 years. Postoperative recovery proceeded without incident with normal facial nerve function. There was no recurrence at 36-month follow-up.

CONCLUSION: Although it is a very rare benign tumor, congenital sialolipoma should be kept in mind in the differential diagnosis of congenital parotid mass. The recurrence of congenital sialolipoma is dependent on its management, thus complete excision of the mass with the lobes of the salivary glands involved seems to be adequate for definitive management.

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1. Introduction

Tumors of the parotid gland in children are uncommon, and represent only 1.3% of all benign salivary tumors [1]. Lipomas of the parotid are uncommon, accounting for only 0.5% of all parotid gland tumors [2]. In a series of 430 salivary tumors in children less than 15 years of age reported by Krolls et al., just three were of a lipomatous nature [1]. Sialolipoma was firstly described by Nagao et al. [3] as a new variant of salivary gland lipoma. Grossly it is characterized as a well-circumscribed, soft, yellow mass and histologically it contains both mature adipose tissue and entrapped normal salivary glandular components surrounded by a fibrous capsule. Sialolipoma has never before been reported as a congenital lesion, or in a child. The first case of congenital sialolipoma was reported by Hornigold et al. in 2005 in a 7-week-old infant as a parotid gland mass [4]. To date, more than 40 cases of sialolipomas have been presented,

but the majority is in adult patients. Sialolipoma in children is extremely rare, with only five congenital cases depicted to date [4–8]. To the best of our knowledge, our case is the sixth case of congenital sialolipoma, and this is the first case of recurrent congenital sialolipoma in infant. Interestingly, all congenital cases were derived from the parotid gland.

We report a case of congenital sialolipoma in the parotid gland of the 3-year-old male child whose particularity is to be recurrent. The aim of this article is to report one more new case of sialolipoma in infant and discuss the clinicopathological and morphological features of sialolipoma, and the possible cause of its recurrence.

2. Case report

A 3 years-old male child with no specific pathological history, in particular, no family history of congenital abnormalities, referred to our otorhinolaryngology department, for the recurrence of a mass in the right parotid region, which was initially treated in another private medical structure. It is a mass noted since birth, progressively increasing in size, without compressive sign or facial palsy, treated surgically at the age of 4 months by tumorectomy with excision of the surrounding parotid tissue. According to the

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Fig. 1. Preoperative aspect of the mass, filling the right parotid region and extending to the ipsilateral cervical region.

histological report, the histological appearance was in favor of a congenital sialolipoma, and the excision was complete with a clear margin of safety. 4 months later, the mass reappears in the same site (right parotid gland) gradually increasing in size without compressive sign or facial palsy. Physical exam was significant for a soft, non-tender, mobile, 10 cm in diameter mass, interesting the right parotid region and overflowing on the ipsilateral cervical region (Fig. 1), with some centimetric and infracentimetric homolateral lymphadenopathy. On Computed tomography (CT) scan, the mass is subcutaneous, well limited, measuring 90 × 70 × 32 mm, which had fatty density (−108HU), and seat of non-enhanced septa after injection of contrast product, in intimate contact with the superficial parotid lobe (Fig. 2). Faced with this radiological aspect of the mass, the diagnosis of recurrence of congenital sialolipoma was retained.

We have planned to perform a total excision of the mass with superficial parotidectomy. The facial nerve was monitored intraoperatively. A standard modified Blair incision was used, the flaps were lifted (Fig. 3A), then the facial nerve was identified in a stan-

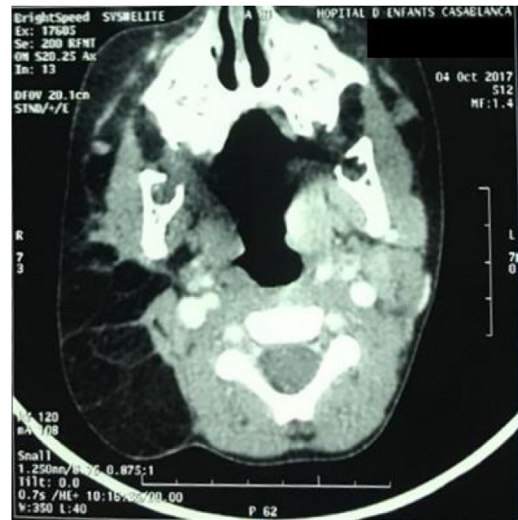


Fig. 2. Computed tomography with contrast injection of the upper neck reveals a subcutaneous low density mass in intimate contact with the superficial lobe of the right parotid gland compatible with a lipomatous tumor.

dard fashion using the tragal pointer and posterior belly of the digastric muscle as landmarks (Fig. 3B). All branches of the facial nerve were identified and separated from the tumor (Fig. 3C). A complete resection of the mass was achieved and normal facial nerve function preserved. The operative blood loss was less than 20 ml. A closed suction drain was put in situ followed by closure in layers. There was no evidence of facial weakness in the immediate postoperative period. Drain removal was done on postoperative day 2.

Histopathological examination of the resected specimen further supported the diagnosis of sialolipoma, showing formation limited by a fibrous capsule consisting of adipose lobules separated by vascular fibrous septa, with local presence of serous acini and excretory tubes (Fig. 4).

The 36 months following the surgery were uneventful. There was no evidence of recurrence Frey's syndrome, or facial weakness.

This work has been reported in line with the SCARE 2020 criteria [9].

3. Discussion

Parotid lipomatous tumors are classified into several histological variants. The standard (true) lipoma is the most common type [10–12]. The term sialolipoma was first used by Nagao et al. it is a mixed tumor consisting of salivary gland elements and mature adipocytes [3]. In their series of 2051 surgically resected primary salivary gland tumors, they reported five cases of well-

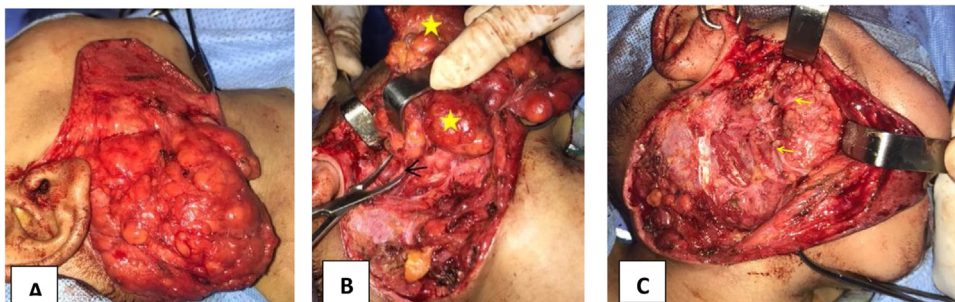


Fig. 3. Intraoperative images of the right parotid mass. After lifting the skin flap, note the polypoid appearance of the tumor which is clearly seen involving the parotid gland (A). The trunk of the facial nerve (black arrow) was meticulously dissected from the tumor (stars)(B). The facial nerve and its two temporo-facial and cervico-facial branches (2 yellow arrows) have been carefully preserved after exofacial parotidectomy with total removal of the tumor(C).

Table 1
Reported cases of congenital sialolipoma of the parotid gland.

author	age	sex	side	imaging	Site	Size (cm)	operation	Follow-up duration (Month)	recurrence
1 Hornigold R et al. (2004) [4]	7 weeks	female	left	MRI	SL	2 diameter	SP	24	NO
3 Bansal B (2007) [5]	11 years	male	left	CT	SL	7 × 7	SP	12	NO
Kidambi T et al. (2012) [6]	6 weeks	male	left	MRI	SL/DL	4.7 × 3 × 4.5	TP	3	NO
4 Mazlumoglu MR et al. (2015) [7]	10 weeks	girl	left	MRI	SL/DL	3,5 × 4 × 2,1	TP	18	NO
5 Erdağ TK (2019) [8]	10 months	male	left	MRI	SL/DL	9 × 8 × 4	TP	24	NO
6 Present case	3 years	male	right	CT	SL	9 × 7 × 3	SP	36	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.

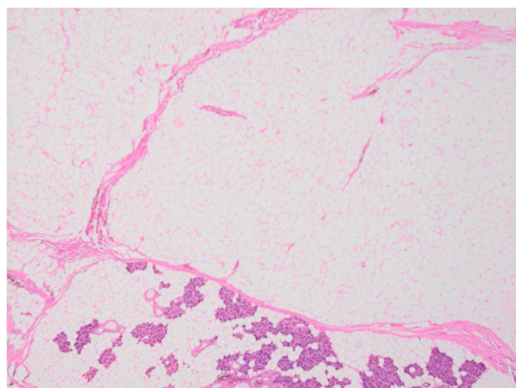


Fig. 4. Histology image of sialolipoma showing large areas Fatty lobules, surrounded by a fine fibrous capsule, with regular acini. (H&E, ×4).

circumscribed parotid tumors, consisting of mature adipocytes and glandular tissue. They proposed that this was a distinct form of salivary gland neoplasm, which they termed sialolipoma. Patients with these lesions have had a mean age of 48.6 years (range 20–67 years). In 2005, the term sialolipoma was accepted in the World Health Organization (WHO) classification of head and neck tumors [13]. It is an extremely rare entity. In one larger study the incidence of sialolipoma was 0.34% of all surgically resected primary salivary gland tumors [3]. Sialolipoma shows a slight male preponderance (male:female ratio = 1.75:1). It is usually seen in adults, the age range spanning 20–75 years {mean, 54 years} [3]. It is more frequently seen in the parotid gland [14–16]. A reported single case each involved, respectively, tumors of the soft and hard palate [3].

Clinically, it presents as a slowly growing, asymptomatic painless palpable mass with a wide range of patients aged from 0 month to 84 years with predominance seen in male. Duration of the lesion in published literature varied from 2 months to 11 years [17].

Computed tomography (CT) or magnetic resonance imaging can be helpful in narrowing the differential diagnosis and is superior to ultrasonography in defining exact location and texture of the lesion. Fine-needle aspiration, which is the first-line procedure in diagnosing major salivary gland lesions, is of little help as its accuracy is <50% in lipomatous tumors [7].

Parotitis is the most common disease of the parotid gland in childhood. Congenital parotid tumours are extremely rare, however, these entities should always be considered in the differential diagnosis if the swelling is persistent. Congenital parotid tumors that can occur in the neonatal period are hemangioma, sialoblastoma, cystic hygroma, branchial cleft cyst, and hamartomas. Epithelial parotid tumors that can occur in late childhood are pleomorphic adenoma, mucoepidermoid carcinoma, and acinic cell carcinoma [4]. Congenital sialolipoma, however, is a newly recognized tumor and only 5 reports are available in the English literature [4–8]. (TABLE below) including 2 girls and 3 boys whose age varies between 7 weeks and 10 months. Coincidentally, the left parotid gland was affected in all 5 cases, while in our case it was located in the right side (Table 1).

It is extremely difficult to confirm a preoperative diagnosis of sialolipoma. Imaging cannot differentiate reliably between a benign neoplasm and one with a more aggressive growth pattern. In this case, the tumor capsule could not be differentiated from the fibrous septa of the subcutaneous tissue of the cheek. Fine needle aspiration cytology may produce adipose and parenchymal tissue only but, as in this case, more aggressive tumors cannot be discounted. Sialoblastoma could well have a similar presentation [18]. It is, therefore, necessary to proceed to total excision of the mass by superficial or total conservative parotidectomy [4].

Pathogenesis of sialolipoma is not completely understood. According to some authors, pathogenesis of sialolipoma may be associated with salivary gland dysfunction, leading to altered salivary gland configuration which can be explained microscopically by replacement of the normal salivary gland tissue with mature adipose tissue admixed with atrophic salivary glandular elements and chronic ductal epithelial cells changes such as oncocytic metaplasia, fibrosis and lymphocytic infiltrate [19,20].

Microscopically sialolipoma is composed of mature adipose tissue along with dispersed glandular elements. The cellular and structural components of glandular tissue are similar to those of the normal salivary gland. There may be slight variations in glandular components, such as duct ectasia, fibrosis, focal oncocytic metaplasia, and sebaceous differentiation [3,21]. The histologic differential diagnosis for a salivary gland tumor with a lipomatous component includes lipomatosis, lipoadenoma, and pleomorphic adenoma with adipose components. Sialolipoma can be differentiated from both lipomatosis and pleomorphic adenoma by the presence of a fibrous capsule and normal salivary gland tissue [3–22]. Lipoadenoma contains a mixture of mature fat cells and glandular epithelial tubules without myoepithelial and acinar cell differentiation. The lack of normal acinar structures distinguishes it from sialolipoma [3,23].

Complete excision of the mass with involved salivary glands lobes appears to be adequate for definite management. Superficial parotidectomy remains the treatment of choice for all superficial lobe benign tumours [24]. Thus most of the tumors in parotid glands are treated with superficial parotidectomy [25]. The extratemporal portion of the facial nerve is most commonly injured during parotid surgery or during the excision of neck lesions originating from or adjacent to the parotid gland. If the tumor is very large, it is safe to approach it by retrograde dissection, by identifying the buccal or marginal branch of the facial nerve. Facial nerve dysfunction may result from accidental transection or compression of the nerve during tissue dissection or wound retraction. Monopolar cautery can also cause thermal and electrical injury to the facial nerve [26]. The patient should be counselled and educated about possible anticipated postoperative complications.

Parotidectomy in small children may present some difficulties. First of all, anatomical structures are much smaller, the facial nerve is located relatively superficially in comparison with adult patients, and the mastoid process is not fully developed. Consequently, operating under magnification with surgical loops or microscopes is mandatory, and facial nerve monitoring may facilitate identi-

fication of the nerve [8]. The most commonly used anatomical landmarks for location of the facial nerve in parotid surgery are the anterior border of the digastric muscle one centimeter below and deep to the cartilaginous pointer of the external auditory canal [4]. In our case, the tumor was carefully dissected from the terminal branches of the facial nerve, continuous monitoring of the intraoperative facial nerve was very helpful to preserve their function.

Several studies have identified no local recurrence, malignant transformation or other complications after conservative surgical excision [19,23,27]. Just one case of recurrent sialolipoma was identified but in adults [28]. Long term data regarding recurrence in the pediatric population is not available, with three years of follow-up being the longest reported thus far [22].

This report highlights an exceptional case of congenital sialolipoma of the parotid gland with short-term recurrence (4 months) following the performance of a simple tumorectomy enlarged to the surrounding parotid tissue. Despite complete resection, the tumor reappeared 4 months later. Recurrence may result from a new and independent sialolipoma in the rest of the parotid parenchyma, because a residual sialolipoma was considered unlikely according to the pathology report, which indicated total tumor exeresis with a clear margin of safety. Whereas, in the recurrence, the complete excision of the mass with the lobe of the salivary glands involved (superficial parotidectomy) allowed definitive management of the tumor; there is no more recurrence after three years of follow-up.

4. Conclusion

Although it is a very rare benign tumor, congenital sialolipoma should be considered in the differential diagnosis of congenital parotid mass. The recurrence of congenital sialolipoma depends on its management. Thus, our case confirms that the complete excision of the mass with the lobes of the salivary glands involved is adequate for definitive management.

Declaration of Competing Interest

The authors report no declarations of interest.

Sources of funding

None.

Ethical approval

The study is exempt from ethical approval in our institution as it is a "Case report" and not a research study.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

K. SALAMA: Investigation, Resources, Writing – original draft, Writing – Review & Editing, Visualization. **M. LAHJAOUJ:** Investigation, Resources. **B. MERZOUQI:** Investigation, Resources, Writing, **Y. Oukessou:** Review & Editing. **S. ROUADI:** Validation, **R. ABADA:** Validation, Supervision. **M. Roubal:** Validation, Supervision. **M. Mahtar:** Validation, Supervision. **KARKOURI. Mehdi, JAMAA. Dounia:** histological examination of the surgical specimen.

Registration of Research Studies

Not applicable.

Guarantor

K. SALAMA.

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