Case Report

Sialocyst of the Parotid Gland in a Child

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ABSTRACT: Non-neoplastic cysts localized in salivary glands are quite rare and represent approximately 2-5% of all salivary gland lesions. Salivary duct cyst, also known as sialocyst is a true cyst with the epithelium lining the inner side of its walls. It is generally observed in minor salivary glands and it rarely involves the parotid. Patients are mostly affected between the ages of 30 and 40 and the lesion is rarely seen in children. The current case is a rare report, as the patient was a child. The diagnosis has been difficult to ascertain due to the presence of pain, symptom characteristic for inflammatory lesions.

KEYWORDS: *Salivary duct cyst, sialocyst, parotid gland, child*

Introduction

Cystic lesions localized in salivary glands are commonly of neoplastic origin, whereas non-neoplastic cysts are much rarer, and constitute approximately 2-5% of all salivary gland lesions [1,2]. Salivary duct cysts, also referred to as mucus retention cyst, mucus duct cyst or sialocysts, are true cysts that have a congenital or acquired origin [1-3], and duct obstruction seems to be the main cause of the lesion [3]. Such cystic masses in parotid gland may lead to clinical and radiographic confusions and multiple differential diagnoses [1].

Salivary duct cyst most frequently involves the minor salivary glands (usually those on the floor of the mouth, buccal mucosa and lip). They rarely affect the major salivary glands, in which case, they are multiple and commonly found in the superficial lobe of the parotid gland [4].

Salivary gland neoplasms are a very complex and diverse group of tumors in the head and neck region. The diagnosis and management of such lesions is complicated by their relative reduced frequency (1% of head and neck tumors). Less than 5% of all salivary gland tumors are found in patients younger than 16 years old, but only 35% of them are malignant [5].

Patients affected by cystic lesions of salivary glands are usually between 30 and 40 years old. Such lesions are rarely seen in children [6,7]. We report here a case of a salivary duct cyst of the left parotid gland found in a 14-year-old child.

Case Report

A 14-year-old female patient presented at the Maxillo-facial department polyclinic from the Emergency County Hospital of Craiova, with a swelling in front of her left ear that had persisted for 2 months. The swelling that was initially small had gradually increased in size.

On extra-oral examination, a single, well-defined, localized, round shape swelling was present in the left preauricular region, measuring 2x2cm. On palpation it was soft in consistency, mobile, fluctuant and painful. The lesion was also painful spontaneous.

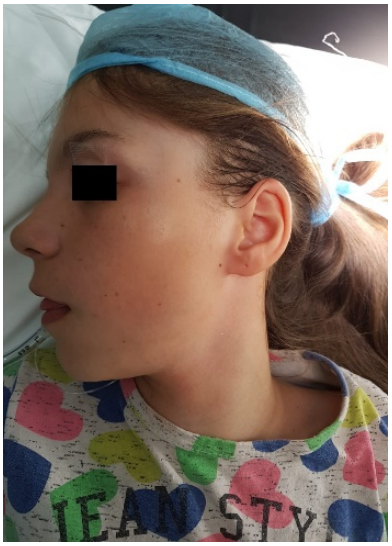

Figure 1. Patient's photo before the surgery (lateral view): a single, well-defined, localized, round shape swelling was present in the left preauricular region, measuring 2x2cm.

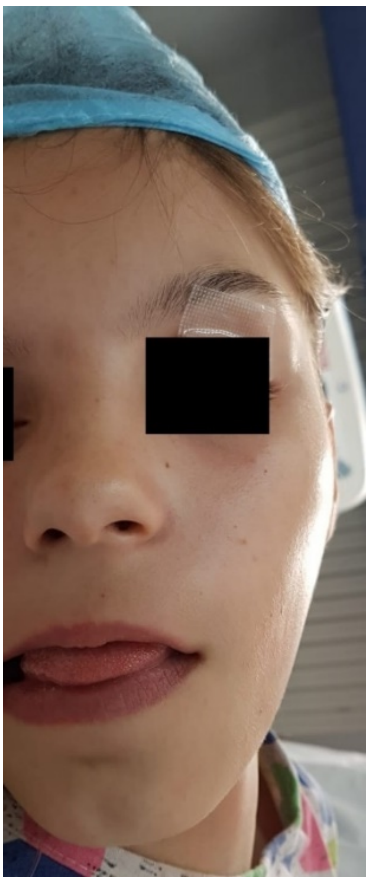

Figure 2. patient's photo before the surgery (anterior view): a single, well-defined, localized, round shape swelling was present in the left preauricular region, measuring 2x2cm.

Considering that the presence of pain is not characteristic for cysts and the rarity of the sialocyst, the initial diagnosis has been of intraparotidian adenitis (Figures 1-2).

Differential diagnoses included neoplastic lesions as pleomorphic adenoma, Warthin's tumor, mucoepidermoid carcinoma, low grade papillary cystadenocarcinoma, metastatic squamous cell carcinoma and non-neoplastic pathologies, especially cysts (lymphoepithelial cyst) [1].

Ultrasonography has been the first intention imaging approach, and it has revealed a well-defined lesion, with a diameter of 2x2cm.

An increase of echogenicity in the superior half of the cyst was noticed, possibly caused by a previous infection.

There was blood flow on Doppler test only in the cyst's wall and not within the lesion (Figure 3).

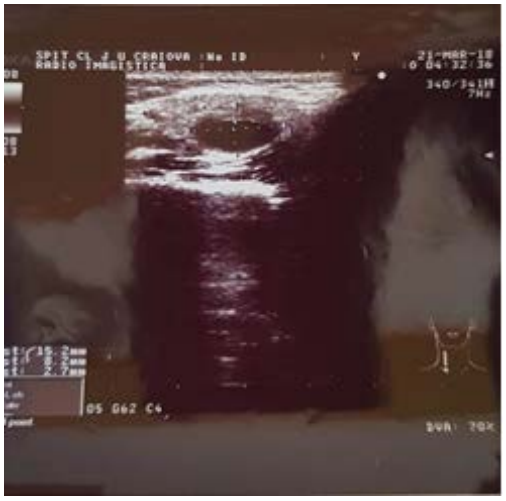

Figure 3. Ultrasound image of the cyst: well-defined lesion measuring 18/11mm in the superficial part of the left parotid gland, with anechoic center and acoustic enhancement in the superior half of the lesion (possibly caused by a previous infection), blood flow present on Doppler test only in the cyst's wall and not within the lesion.

Antibiotic, anti-inflammatory and antispastic treatment has been administered to the patient with the role of a therapeutic test, but the cyst didn't resolve.

Needle aspiration has been performed next, and a serous fluid similar to saliva has been evacuated (Figures 4-5).

Figure 4. Needle aspiration of the cyst content.

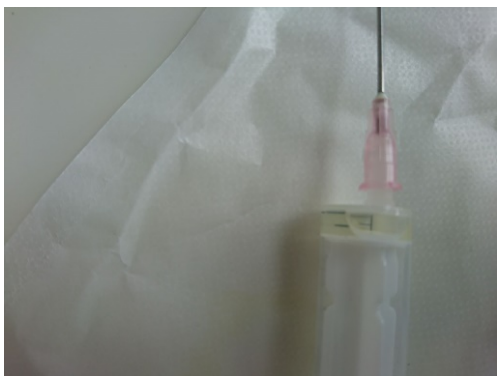

Figure 5. Cyst content: a serous fluid similar to saliva has been evacuated.

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Our findings after clinical and paraclinical examination (ultrasonography, therapeutic test with antibiotics, anti-inflammatory and antispastic medication, needle aspiration) have been corroborated with the data from the existing literature, suggesting most probably a diagnosis of sialocyst. The cyst has been enucleated under general anaesthesia and a superficial parotidectomy has been performed, preserving the facial nerve (Figures 5-11).

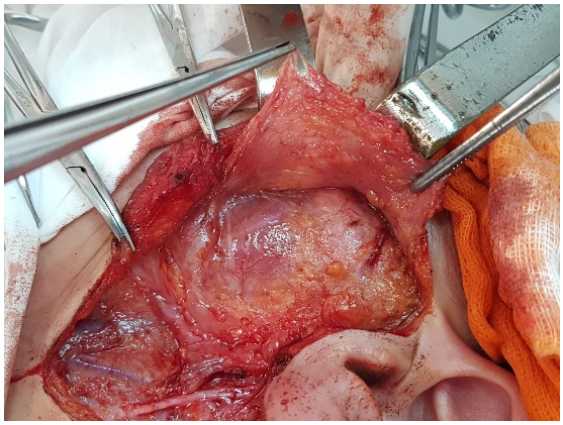

Figure 6. Intraoperative image of the sialocyst: through the membrane's transparency of the cyst the blue color of the liquid content can be observed.

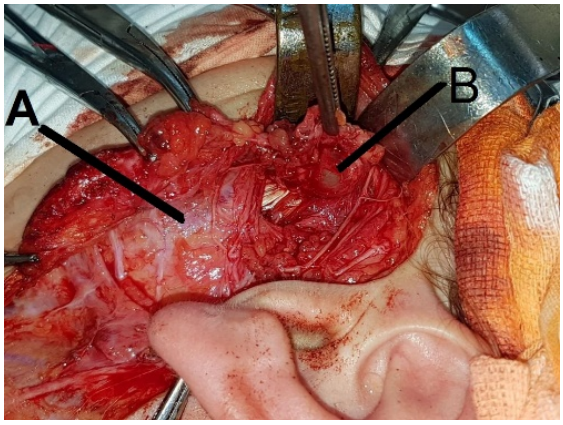

Figure 7. Intraoperative image: A=masseter muscle, B=retention cyst.

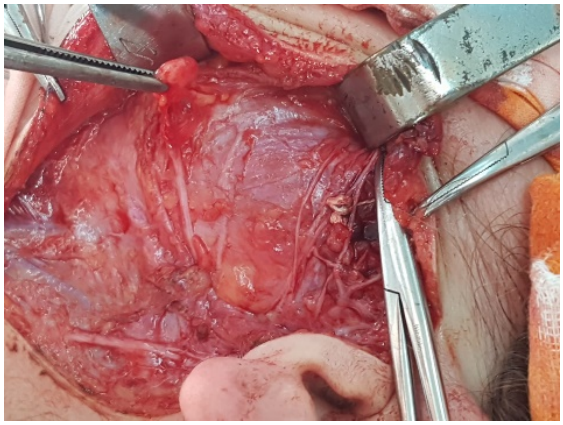

Figure 8. Facial nerve demonstrated.

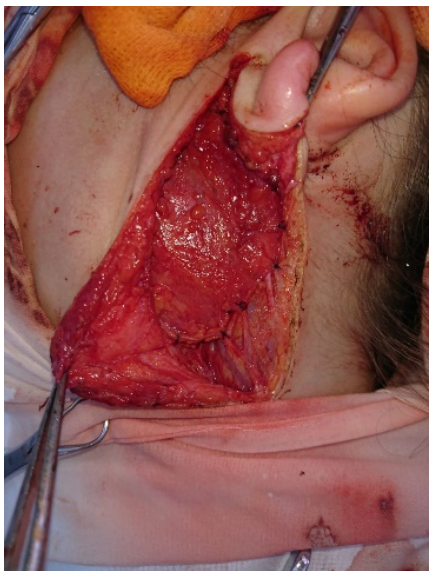

Figure 9. Sutures of anatomical layers.

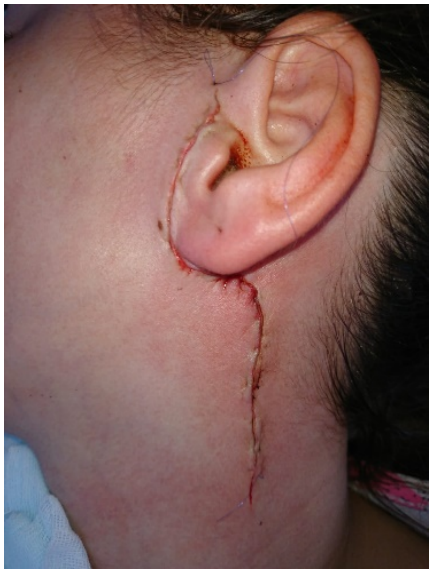

Figure 10. Post-operative image of the patient.

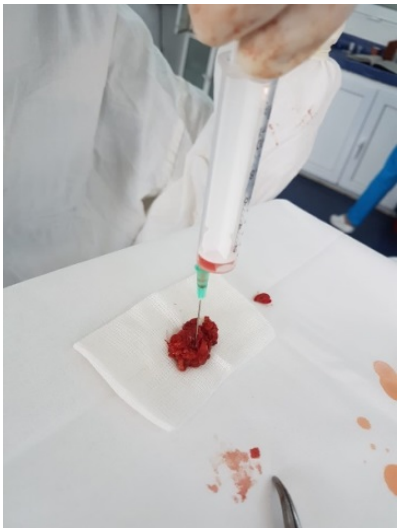

Figure 11. Superficial lobe of the parotid gland with the well-defined sialocyst have been sent for histopathological examination.

There were no post-operative complications, and no recurrence was detected during the follow-up of the patient for the next year (Figures 12-13).

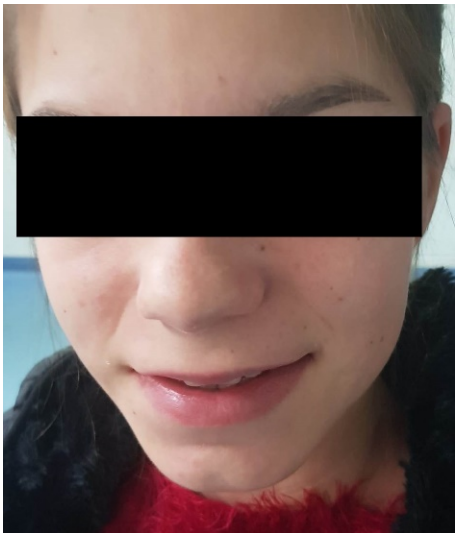

Figure 12. Patient's photos at 6 months post-surgery follow-up (anterior view).

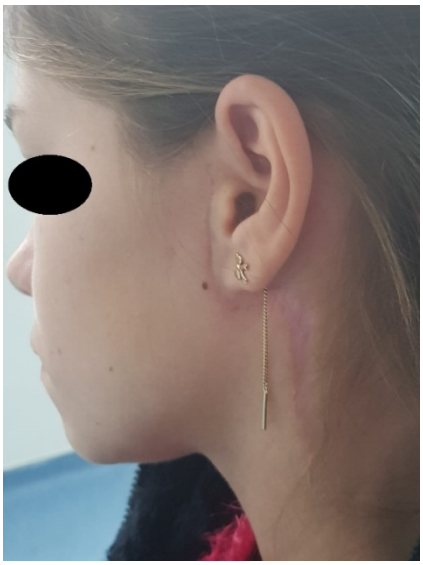

Figure 13. Patient's photos at 6 months post-surgery follow-up (anterior view).

Histopathological investigation has been performed on the excised tissue, in the Pathology Department from the Emergency County Hospital of Craiova.

The specimen was fixated in 10% neutral buffered formalin and it has been processed by classical histopathological technique for paraffin embedding, microtome sectioning and routine haematoxylin-eosin staining.

A written informed consent was obtained from the patient's mother for case publication.

At microscopic examination, an excretory glandular salivary canal of large dimensions has been identified, dilated, with thick, fibrotic walls, and lined by a bistratified flat epithelium. (Figures 14-15).

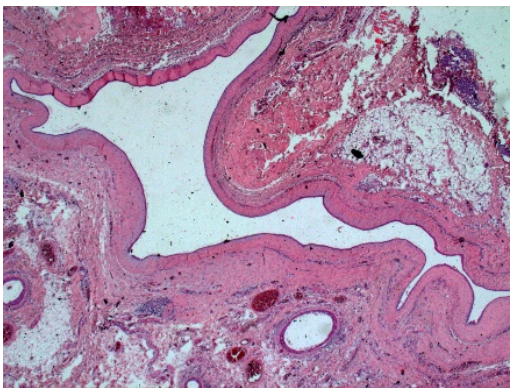

Figure 14. Excretory glandular salivary canal of large dimensions, dilated, with thick walls, fibrotic and lined by a bistratified flat epithelium, Col. HE, X25.

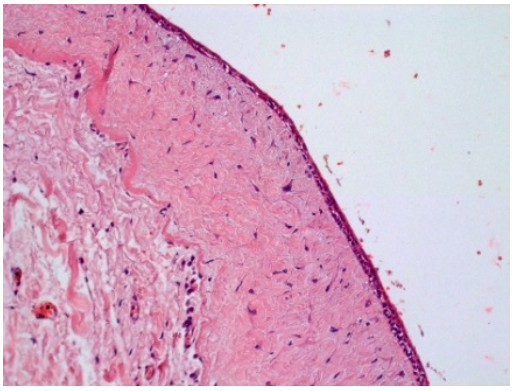

Figure 15. Magnification of the previous picture showing the wall of the excretory glandular salivary canal with a thickened and fibrotic appearance, lined by a bistratified flat epithelium, Col. HE, X200.

The adjacent glandular salivary parenchyma presented lesions of atrophy, lipomatous sclerosis and lymphoid pseudo-nodular infiltrates, with peri canalicular disposition (Figure 16).

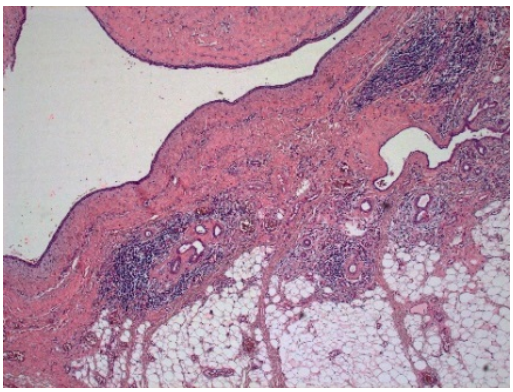

Figure 16. Excretory glandular salivary canal of large dimensions, dilated, with salivary glandular parenchyma and arranged predominantly peri canalicular, Col. HE, X50.

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Further away from the dilated excretory glandular salivary canal, the glandular salivary parenchyma, similar to the parenchyma of the parotid gland, presented lipomatosis lesions, extensive interlobular fibrosis, dilated excretory intra-lobular canals, some of them with sialoliths, surrounded by lymphoid infiltration (Figures 17-18).

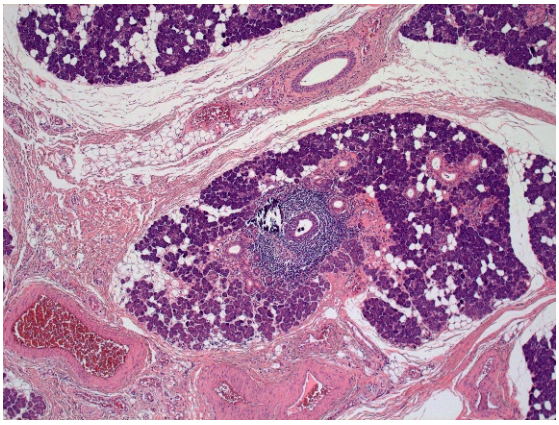

Figure 17. Glandular salivary parenchyma similar to the parenchyma of the parotid gland with interlobular fibrosis, lipomatosis, dilated canals, some of them with sialoliths, surrounded by lymphoid infiltration, Col. HE, X50.

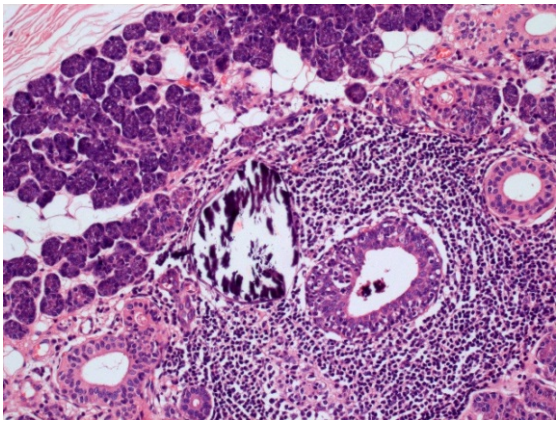

Figure 18. Magnification of the previous photo showing intralobular canaliculi surrounded by lymphoid infiltrations and one intraluminal sialolith. Col. HE, X200.

These histopathological features corresponded to a sialocyst of the parotid gland with secondary lesions of lithiasis caused, most probably, by chronic sialadenitis.

Discussion

Salivary duct cysts occur most commonly following an obstruction in the duct.

Although the exact causing factor leading to obstruction is often unknown, some hypotheses include calculi, mucus plugs, postoperative or inflammatory strictures [2].

Ductal narrowing has also been reported to be associated with frequent mouth wash with hydrogen peroxide and tartar-control toothpastes [1].

In our case report, the etiology is unknown because none of these causing factors has been identified.

Sialocysts are usually observed in the minor salivary glands (on the floor of the mouth, buccal mucosa and lips). They rarely affect the major salivary glands and when they do, they are often present in the superficial lobe of the parotid [2].

Salivary duct cyst of the parotid gland presents mostly as an asymptomatic, unilateral swelling. The facial nerve is not affected and fixation to overlying skin is absent. The cysts range from 0.8 to 10cm, but the majority reach 1-3cm in size [2].

These features are similar with the ones in our case report.

Imaging plays an important role during the investigation of the salivary duct cysts, in determining the borders, extent of involvement and content.

Ultrasonography is the investigation of first intention and in our patient, it has led the diagnosis towards a cyst, although the initial one was of intraparotidian adenitis.

On the computer tomography scan, the lesion appears well defined and hypodense [2].

Fine needle aspiration may not disclose the pathology, since the aspirated cystic content can be devoid of tumor cells, when the needle does not reach the representative site [1].

In our case, the needle aspiration was consistent with the ultrasonography examination.

Histopathological investigation defines the final diagnosis.

Salivary cysts are most frequently unilocular, lined by ductal epithelium which may be flat, cuboidal or columnar and completely or partly lined by squamous epithelium.

Discrete to moderate lymphocytic infiltrations may be present in the cyst's wall. Occasionally, oncocytic metaplasia (often seen in cases following ductal obstruction) is seen [2].

Salivary gland cysts may also be an early manifestation of a salivary gland tumor.

Presence of epithelial alterations, such as metaplasia's and focal papillary proliferations observed are comparable to similar changes seen in odontogenic cysts [2].

Considering the fact that salivary gland cyst could be an early marker of tumor manifestation, long term follow-up has been decided for this case.

Surgical excision is the election curative treatment. Removal of the affected lobe of the gland may also be necessary [2].

Acknowledgment

Maria Cristina Munteanu, Allma-Roxana Pitru and Cristina Babov Opris contributed equally to the manuscript.

Conflict of interests

None to declare.

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