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Case Report

Sialoblastoma of the Sublingual Gland: A Case Report

Ndongo Pilor[1]*, Ciré Ndiaye[1], Mame Sanou DIOUF[2], Houra Ahmed[1], and Issa Cheikh NDIAYE[1]

[1]Clinique d'ORL et de Chirurgie cervico-faciale, CHU de Fann, Dakar – Sénégal
[2]Clinique d'ORL et de chirurgie cervico-faciale, Centre Hospitalier Universitaire Idrissa Pouye de Grand Yoff, Dakar - Sénégal

***Corresponding author**
Ndongo Pilor, Clinique d'ORL et de Chirurgie cervico-faciale, CHU de Fann, Dakar, Sénégal, Tel : 00221775635500; Email: ndongopilor@gmail.com

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Abstract

Introduction : Sialoblastoma is a rare, congenital malignant epithelial tumor of the salivary glands. It is located mainly in the parotid and maxillary glands. Its location in the sublingual gland is exceptional.

Summary of the clinical case : We report the case of a 10-month-old infant, with no pathological history, who consulted for a congenital mass under left mandibular, gradually increasing in volume.

The examination found a left submandibular mass of about 10 centimeters long axis, firm, mobile compared to the 2 planes, painless with a healthy skin.

The cervical CT showed a left mandibular mass, tissue, with regular contours; without lymphadenopathy.

The patient had a complete excision of the mass under general anesthesia. Intraoperatively, we found a mass of cerebral aspect extending forward and inward of the left maxillary gland which was normal in appearance. The postoperative course was simple.

Anatomopathological examination of the operative specimen concluded with sialoblastoma.

Conclusion : Topographical diagnosis of the sialoblastoma of sublingual gland is possible in peroperatory. There is no consensus on the treatment which is essentially surgical by complete exeresis.

INTRODUCTION

Sialoblastoma, also known as embryoma, congenital basal cell, basal cell adenoma or basaloid adenocarcinoma, is an epithelial tumor of the basal cells of salivary glands with predominantly local malignancy [1-3].

It is a rare tumor, described for the first time in 1966 by VAWTER and TEFFT [2]. It is TAYLOR, which gives it the name of 'sialoblastoma' in 1988 [2].

It accounts for 2-5% of salivary gland tumors in children with a worldwide incidence of 0.8 per million [4].

We found 63 cases in the English literature. The parotid and maxillary glands were the preferred site [2,4].

The aim of this work is to report a case of sialoblastoma of the sublingual gland and to discuss the epidemiological, clinical, paraclinical, therapeutic and evolutionary aspects.

OBSERVATION

It was a 10-month-old infant, with no particular pathological history, who consulted for a congenital left mandibular mass, gradually increasing in volume, painless, without signs of compression.

The examination found a left mid-maxillary mass of about 10 centimeters long axis, firm, mobile relative to the surface and deep, painless, covered with a healthy skin. Cervical ganglionic areas were free.

The cervical CT scan revealed a left submandibular mass, hypodense, with regular contours; without lymphadenopathy.

A complete excision of the mass was done under general anesthesia, first by Sebileau CARREGA. The mass was cerebriform, yellowish, multi-lobulated, easily cleavable extending into sub mental (Figure 1). It was developed at the expense of the

left sublingual gland. The left maxillary gland was in place and normal in appearance (Figure 2).

Histopathological examination of the operative specimen showed tumor proliferation of differentiated epithelial cells. They formed small clusters with in the center, a light roughly reminiscent of acini. The border cells described a palisade structure and were basaloid. The capsule was intact with some mitoses (Figure 3). With a follow-up of 02 years, we don't have recurrence.

DISCUSSION

The incidence of salivary gland tumors before the age of 10 is 0.25% [1]. The majority of these tumors are non-epithelial and dominated by hemangioma. A review of the literature found 63 cases over 50 years (1966 to 2016) with 44 cases of the order of unity [2,4].

It is a tumor that most often affects the subject of male, less than 10 months old with a sex ratio of 2.1. [2-4].

The sialoblastoma is most often located in the parotid gland or the maxillary gland. Octopus and jugal ectopic locations have been noted [1-4]. The localization of the sublingual glands has not been reported to our knowledge. Clinically, at an advanced stage, the gland is difficult to identify for sub maxillary or sublingual localization. However, intraoperatively, the topographic diagnosis is easy, as in our patient.

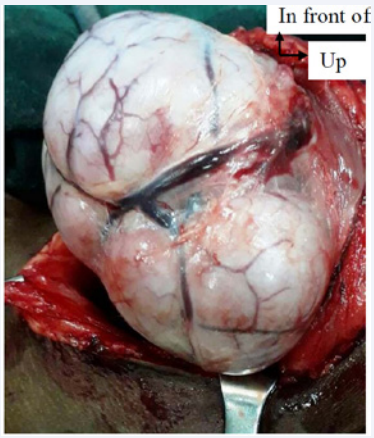

Figure 1 Intra-operative view: aspect of mass.

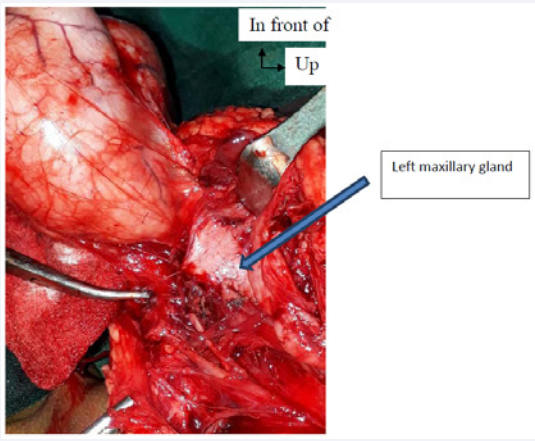

Figure 2 Intraoperative view: masse extending under the mind with a normal-looking, maxillary gland in place.

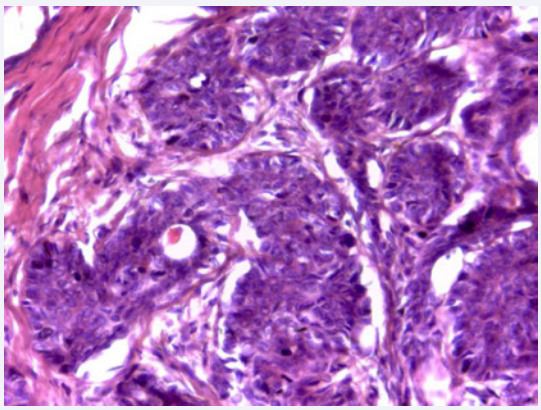

Figure 3 Histological aspect of sialoblastoma (Gx100, HE coloration).

Its size varies between 1.5 - 15 cm long axis [2,3]. The imaging is not specific. MRI shows a hypodense tissue mass in relation to the brain, and isodense compared to muscles with T1 hypointense and T2 hypersignal [2,4]. A cervical scanner is required in case of non-availability of the MRI and for the extension assessment.

Histologically, sialoblastoma is characterized by basal palisade epithelial cells with mitoses, separated by bands of fibrous tissue, mixed with ductular proliferative structures in double layers around muscle cells [1-4].

Some histological criteria would be poor prognosis such as anaplasia, neurovascular invasion or necrosis [4].

The treatment of sialoblastoma, whatever its location, consists of complete resection surgery with margins of healthy resections [2,4,5]. In case of irreversible lesion or metastasis, neo-adjuvant chemotherapy is indicated based on 5-fluorouracil, cisplatin and vincristine or radiotherapy [1-5]. Authors have reported cases of local recurrence with lymph node and lung metastases [2-4]

We found 10 cases of recurrence at about 24 months and 3 cases of death by sepsis and respiratory failure, hence the importance of regular monitoring with imaging [2,4,5].

CONCLUSION

Sialoblastoma is a rare malignant congenital tumor of the salivary glands. Its location at the level of the sub-lingual glands is exceptional. Topographic diagnosis is possible intraoperatively. The imaging is not specific and its diagnosis is anatomo-pathological.

There is no consensus on treatment, which is essentially surgical by complete excision. Rigorous, long-term surveillance is necessary because there are loco-regional and remote recurrences.

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