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Obstructive Intra-Parotid Cystic Lymphangioma in Child: About Two Malagasy Cases

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Abstract

Objective: To report the clinical manifestations and the methods of taking charge of this pathology from two cases observed at the Mahajanga University Hospital.

Observation: The first case was a 3-year-old child referred for a right parotid swelling progressing from birth. This benign, non-inflammatory tumor was associated with sleep apnea episodes. Echography investigation confirmed the diagnosis of a parotid gland cyst, a surgical excision was carried out with simple follow-ups.

The second case was a 1 month old infant referred for suspected right parotitis. It was a non-inflammatory parotid swelling, gradually increasing in volume, with no other symptoms. The cervical scanner confirmed an intra-parotid cystic mass. After two months of monitoring, the tumor increased in size and episodes of sleep apnea appeared. Surgical excision was performed.

Conclusion: The diagnosis of intra-parotid cystic lymphangiomas is suspected by clinic examination. The natural evolution of the tumor can cause airway obstruction. Its parotid location complicates its management because this gland is transfixed by the facial nerve, thus increasing the risk of nerve damage during surgery. However, faced with the vital risk of obstruction of the airways, the surgery remains life-saving.

Keywords: *Surgery; Facial; Cystic Lymphangioma; Parotid*

Abbreviation

ENT: Ear Noise and Throat

Introduction

Cystic lymphangiomas are rare birth defects that cause tumor syndrome due to angiolymphatic proliferation [1]. Their cervico-facial locations are the most frequent, but their parotid seat is rare [2]. Untreated, it can lead to obstruction of the upper airways, which is life-threatening by asphyxiation [3]. Its severity is also due to its difficulty in excision due to the proximity of the parotid gland to the facial nerve. His delay in treatment may be linked to an unknowing of this child's pathology, wrongly evoking parotitis. We report two Malagasy cases of intra-parotid localization of this pathology in which we report the clinical aspects and the methods of management.

Observation

Case 1

A 3-year-old boy was referred to the ENT department of Mahajanga University Hospital for a right pre-auricular swelling (Figure 1). This mass has evolved since birth by gradually increasing in volume, without any care. The mother reported localized pain in this swelling one month earlier, as well as parts of sleep apnea. No family history was found, the pregnancy and childbirth were uneventful, the child's vaccines were up to date. The clinical examination showed a localized swelling in the right parotid region, covered with normal, relentless, painless, non-blowing skin. The oropharynx was asymmetrical at the expense of the right tonsillary space. No cervical lymphadenopathy was palpated. The cervical echography found a multi-located cystic mass in the parotid gland (Figure 2). Given this context suggestive of a cystic lymphangioma with sign of airway compression, surgical treatment by the preauricular access with under facial tumor excision and total parotidectomy associated with dissection of the facial nerve was performed. The cystic tumor was located in the deep lobe of the parotid gland (Figure 3).

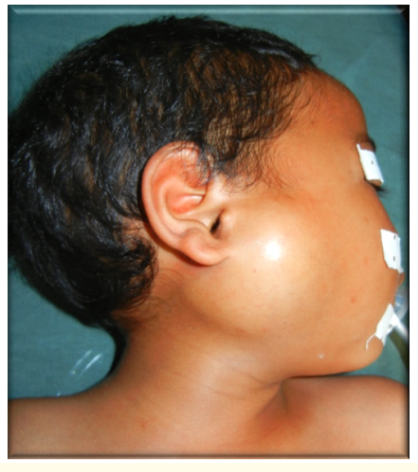

Figure 1: *Right non-inflammatory parotid swelling in a 3-year-old boy (Case 1).*

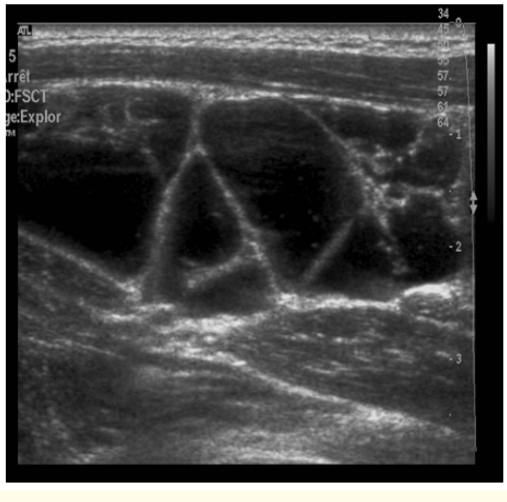

Figure 2: *Ultrasound image showing a thin-walled polycystic mass in favor of a cystic lymphangioma.*

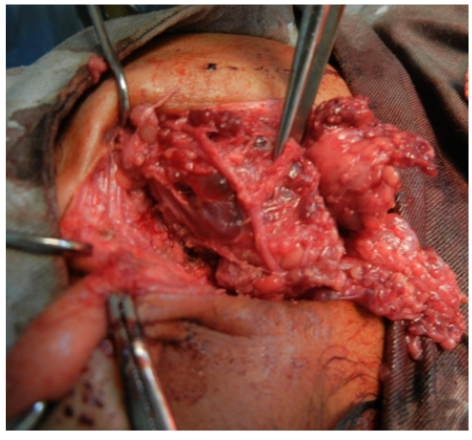

Figure 3: *Intraoperative view of a cystic lymphangioma under the facial nerve.*

The postoperative operations were uneventful with disappearance of sleep apnea, an anatomopathological examination of the operating room confirmed a cystic lymphangioma.

Case 2

A 1-month-old male infant was referred to the ENT department for a right parotid swelling discovered since birth. This child in good general condition had no particular history, the pregnancy had proceeded normally. There was no swallowing or respiratory disorder. On clinical examination, there was a right parotid swelling peeling off the ear lobule, covered with normal, transluminal skin, of a soft and painless consistency (Figure 4). The rest of the examination was without particularities, the face was symmetrical. The cervical scanner found a multi-lobulated parotid mass without compression of the upper aerodigestive tract (Figure 5). The exploratory puncture was non-contributory. After a surveillance period of 04 months, an increase in tumor volume with episodes of apnea was noted. Surgical treatment by total parotidectomy at the age of 6 months was decided. A meticulous dissection of the facial nerve on a tumor that completely invaded the parotid gland was performed (Figure 6). A poly cystic tumor (Figure 7) was referred for anatomic pathology, which confirmed a cystic lymphangioma without signs of malignancies A transient peripheral facial paralysis was noted (Figure 8) which recovered completely in 6 months.. No tumor recurrence was noted at 3 years post-operative

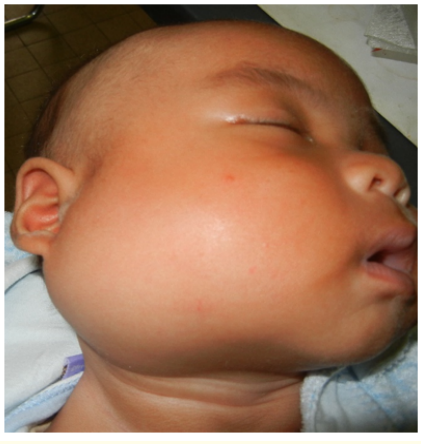

Figure 4: *Right non-inflammatory parotid swelling in a 6-month-old boy (Case 2).*

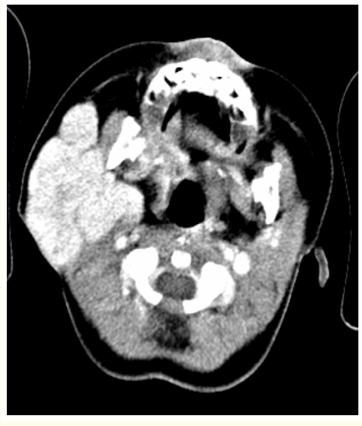

Figure 5: *Cervical CT showing a parotid mass, infiltrating the parapharyngeal region.*

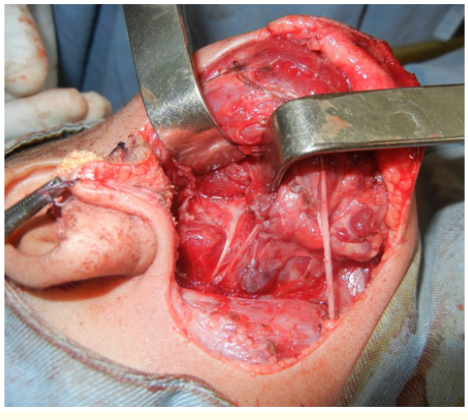

Figure 6: *Exo and sub facial cystic lymphangioma, Dissection of the facial nerve.*

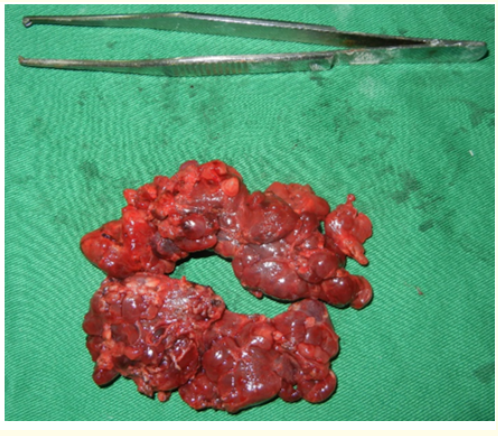

Figure 7: *Macroscopic post-operative aspect of the microcystic lymphangioma.*

Figure 8: *Facial paresis at 10 days Postoperative of case 2.*

Discussion

Cystic lymphangioma is a tumor caused by a malformation of the lymphatic system [2]. The formation of the cyst follows a defect in connection of one of the primitive lymphatic bags with the rest of the lymphatic system from the sixth week of embryogenesis [4]. This benign tumor would be rare, its incidence has been estimated between one in 6,000 and one in 16,000 births without racial or sexual predominance [5].

Cystic lymphangiomas can be localized in different parts of the body but the cervico-facial localization is the most encountered in 90% of cases [6]. Its primary location in the parotid gland is rarely reported in the literature [2]. They are frequently discovered in the first two years of life, but adult cystic lymphangiomas have been reported [7].

Clinically, cervico-facial cystic lymphangiomas evolve from birth and present as an often latero cervical, painless and transilluminable swelling, well limited but gradually increasing in volume over time. These clinical features already make it possible to eliminate inflammatory swellings of the parotid gland, except in cases of superinfection in which the swellings are inflammatory, wrongly evoking parotitis.

Ultrasound will typically show a thin-walled, multi-walled, hypoechoic lesion and will also allow ante natal diagnosis in almost 90% of cases [8]. Computed tomography will show the involvement of the deep lobe of the parotid gland and the condition of the airways. However, magnetic resonance imaging is the examination of choice in the diagnosis of cystic lymphangiomas. It confirms the presence of a thin-walled, intra-parotid multi-cystic lesion, hypointense in T1-weighted images and hyperintense in T2-weighted images [9] and makes it possible to rule out the main differential diagnosis, which is hemangioma.

Cystic lymphangiomas can progress for a long time without any symptoms. This situation is responsible for a delay in treatment. However, complications of the type of infection, of intracystic hemorrhage can occur, causing compression of the facial nerve and obstruction of the upper airways [4] due to the intimate relationship of the parotid gland with the lateral walls of pharynx.

Conservative treatment such as intra-lesional injection of sclerosing agent such as OK 432, bleomycin and Ethibloc or evacuation puncture may be offered in asymptomatic patients [4]. These treatments are particularly interesting in the parotid localizations of Cystic Lymphangiomas considering the surgical risk of the facial nerve. However, tumor recurrences in these conservative treatments are not negligible [10]. Surgical treatment is indicated before the onset of signs of severity such as airway obstruction. Despite the risk of surgical damage to the facial nerve due to its close relationship with the tumor which naturally invades the glandular and surrounding tissues [4], surgery for intra-parotid cystic lymphangioma in children remains saving, removing the tumor obstruction airways. It consists of a preauricular approach with total parotidectomy and meticulous dissection of the facial nerve allowing the obstruction to be removed.

Aesthetic sequelae with a type of peripheral facial paralysis may be only partial and of good prognosis to be discussed in the face of the urgency of the situation.

Conclusion

Intraparotid cystic lymphangioma is a rare congenital tumor. The diagnosis is strongly suspected before the evolutionary mode and the clinical characteristics. Ultrasound is a non-invasive method to support the diagnosis, but the CT and MRI allow to specify its nature and its para-pharyngeal extension. The appearance of signs of obstruction of the upper airways is a crucial element in the surgical decision, despite the difficulties and aesthetic risks of this parotidectomy in children.

Conflict of Interest

We declared that there was no conflict of interest in this study.

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