

Lymphoepithelial Cyst of the Parotid Gland Revealing an HIV Infection: A Pediatric Case Report

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Abstract

Case Report

Benign lymphoepithelial cyst is an uncommon lesion that usually occurs in the parotid gland or lateral cervical area, including the lymph nodes. In adults, It often reveals an HIV infection. Very few cases in children are reported in the literature. We report the case of a girl, aged 7 years, who consulted for bilateral parotid swelling. Parotid ultrasound and MRI revealed multilocular cystic lesions of both parotids suggestive of lymphoepithelial cysts, who had objectified multiloculate cystic lesions in both parotid. Confirmed by anatomopathological study of the superficial parotidectomy part. The search for anti-HIV-1 and anti-HIV-2 antibodies requested postoperatively came back positive. The patient was referred for care and follow-up in a specialized service.

Keywords: lymphoepithelial cyst- HIV- child.

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INTRODUCTION

Benign lymphoepithelial cyst is a benign tumor related to dysembryoplasia involving the lymphatic system; it is often associated with HIV infection, with a predominance in adults [1]. We report a case of lymphoepithelial cysts of the parotid glands revealing retroviral infection in a 7-year-old child.

OBSERVATION

A 7-year-old girl, with no known personal or family pathological history, presented with an isolated bilateral parotid swelling that had been evolving for 1 year. The patient had no history of pain or purulent discharge. Physical examination revealed a bilateral,

well-limited, homogeneous, depressible, soft, mobile, painless parotid swelling covered by healthy skin. Parotid ultrasound revealed cystic parotid lesions, multiloculated; of fine echogenic content (Fig. 1). Parotid MRI concluded to a multiloculated cystic formation, with a liquid signal and enhanced walls after gadolinium injection (Figure 2). A superficial parotidectomy was performed. Pathological examination confirmed the diagnosis of lymphoepithelial cyst of the parotid gland. Anti-HIV-1 and anti-HIV-2 antibodies were found to be positive. The patient was referred for management and follow-up in a specialized service.

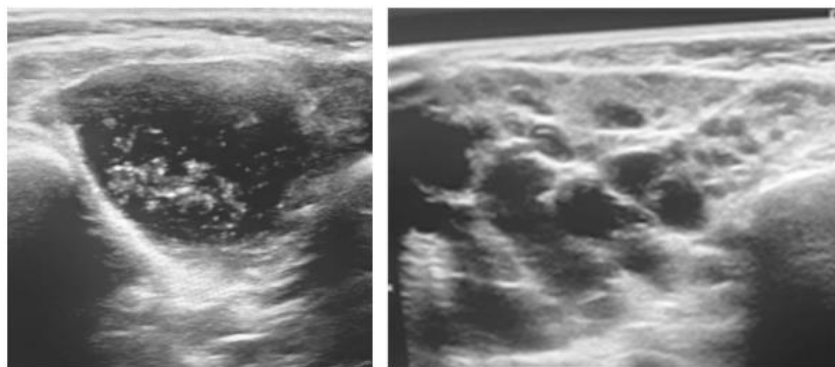


Fig-1: Parotid ultrasound showing multiloculated cystic lesions with fine echogenic content

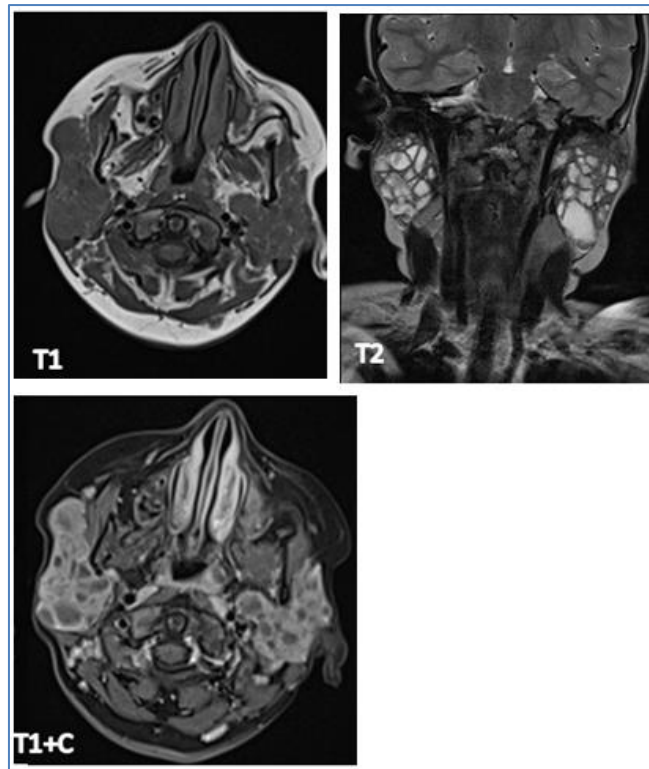


Fig-2: MRI of the parotid glands in T2 coronal, T1 axial sequences before and after injection of gadolinium; showing multiloculated cystic lesions, in T2 hypersignal, T1 hyposignal, whose partitions are enhanced after injection of gadolinium (arrow)

DISCUSSION

Benign lymphoepithelial cyst is a rare condition that affects the salivary glands, primarily the parotid gland. It is associated with HIV infection with an incidence between 3 to 6%. The cases reported in the literature are between 16 and 69 years old with a female predominance. In the majority of cases are unilateral [1]. A reported case from the literature of a 17 year old adolescent girl with negative HIV status[2].

The origin of lymphoepithelial cysts is unclear. We sometimes suggest a hyperplastic activity of intraparotid lymphocytes capable of generating a cystic lesion, or, we would rather speak of glandular epithelial inclusions even inside the intraparotid lymph nodes which would lead to clinically significant epithelial proliferation [3].

Regarding HIV + patients, there is a much higher concentration of viral particles in the cysts compared to the bloodstream [4]. We also observe the presence in the pericystic parotid tissue of a proliferation mainly of CD8 lymphocytes within the framework of diffuse infiltrative CD8 lymphocytosis [5]. There is therefore probably a close relationship between HIV acting as an activating factor in intraparotid lymphoid tissue and the appearance of these cyst.

Radiological diagnosis is based on MRI because of its performance in analyzing the internal

structures of lesions and their contrast in soft tissues. In typical cases, these cysts generally present an appearance of hypointense T1 and homogeneous hypersignal in T2. The wall of the cyst is barely detectable in the two sequences, but it can become thick, visible and irregular due to the inflammatory changes.

In atypical cases, we can find slight heterogeneous hyperintensities in T1 and hypointensities in T2 and on the sequences with saturation of fat signal (STIR).

The signal differences are caused by inflammation which results in the formation of other materials (blood, proteins, etc.) [6].

The differential diagnosis arises with Warthin's tumor, benign intramuscular hemangioma, branchial cyst and lymphoma. MRI is the imaging modality of choice and allows the lesion to be characterized. Benign intramuscular hemangioma is multiloculated and produces a "bunch of grapes" appearance. It is T1 isointense and T2 hyperintense and presents a heterogeneous peripheral enhancement after administration of contrast product. [7] Warthin's tumors present T1 hypointense and moderate T2 hypersignal, and do not enhance after injection of contrast PDC [8].

Although the definitive treatment is surgical excision, other treatment modalities include abstention,

cyst aspiration, sclerotherapy, radiotherapy, and antiretroviral therapy in immunocompromised patients. A superficial parotidectomy was performed in our patient and follow-up at 6 months revealed no sign of local recurrence. Close monitoring is necessary in immunocompromised patients because of the high risk of developing lymphomas [9].

CONCLUSIONS

Benign parotid lymphoepithelial cyst is a rare condition and often associated with HIV infection. Imaging allows to evoke the diagnosis and to eliminate the differential diagnoses

Conflicts of interest

The authors declare no conflict of interest

Contributions of the authors

All authors contributed to the conduct of this research work. All authors have read and approved the final version of the manuscript.

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