



Case report

Regression of both oral mucocele and parotid swellings, following antiretroviral therapy

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ABSTRACT

HIV-salivary gland associated disease is a well accepted concept in the HIV-related literature. Parotid swellings, especially in its cystic benign lymphoepithelial form, have been largely reported. Oral mucoceles (ranulas) were also associated with HIV in some publications. The exact nature of this link between mucoceles and HIV is still to be clarified. The mainstream treatment of most of parotid pathologies and oral mucoceles remains surgical approach. Strong evidences do, however, exist about lymphoepithelial lesions of parotid glands that have been successfully treated with antiretroviral drugs. We present a case of intraoral mucocele, coexisting with bilateral parotid gland lymphoepithelial lesions, on a 2-year-old HIV-positive patient. Both parotid gland swellings and the sublingual mucocele have completely regressed following antiretroviral therapy. No surgical intervention was required. Conversely to benign lymphoepithelial lesions of parotid glands, the regression of oral mucocele on HIV-positive patient, following antiretroviral drugs therapy appears to be a rare phenomenon.

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1. Introduction

HIV-salivary gland associated disease (HIV-SGD) is a well accepted concept in HIV literature [1,2]. Benign lymphoepithelial lesions (BLEL) of parotid salivary glands are among the most commonly described BLEL [2–4]. These lesions appear more often in the early stage of HIV infection, and may be its first manifestation. There is no universally accepted treatment for BLEL of parotid glands. Different modalities of treatment for BLEL have been suggested in the literature. Superficial parotidectomy and low-dose radiotherapy were advocated for in the early days [5,6]. Simple observation, fine needle aspiration as well as sclerotherapy have also been suggested, with variable results [3,7]. Finally, BLEL of parotid glands have been reported to respond dramatically well to antiretroviral therapy (ART). Benign lymphoepithelial lesion of parotid glands have, under ART, regressed to almost their original size in some cases [8–10].

The association of oral mucoceles (ranulas) with HIV infection has already been reported in the literature [11–13]. The exact pathogenesis of this relationship between these two different pathologies remains, however, unclear.

Several modalities of treatment for oral mucoceles have been also described: laser excision [14], cryosurgery, sclerotherapy [15], and marsupialization. Radical surgery of the cystic lesion and / or the attached salivary gland is by far the most accepted standard mode of management [16,17]. Little is, however, known about the effect of ART on oral mucoceles. This report presents a case of an intraoral (floor of the mouth) mucocele, in a HIV-positive young patient that, with the associated bilateral cystic benign lymphoepithelial lesions of the parotid glands, has completely regressed, following antiretroviral drugs therapy.

2. Case report

A 2-year-old girl was brought to the out patients clinic of the Maxillofacial Surgery department with the complaint of a slow growing, bilateral parotid gland swelling, as well as the presence of a small growth in the floor of the mouth, that was interfering with the normal process of feeding and speech. The medical history revealed that she was a known HIV-positive patient, and she just completed her treatment against pulmonary tuberculosis (PTB). Physical examination noted the presence of large bilateral parotid gland swellings (Fig. 1A and B), which were slightly tender on palpation. The intraoral examination revealed the presence of a cystic lesion in the right floor of the mouth, which characteristics corresponded clinically to that of an oral

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Fig. 1. Massive swelling of parotid gland in a pediatric HIV-positive patient. (A) Right side and (B) bilateral view.

mucocele. There was no history of any other congenital growth. A fine needle aspiration (FNA) was performed on the intraoral lesion, and yielded approximately 2.5 ml of amber and mucus-like fluid, consistent with a salivary gland origin of the intraoral lesion. The fluid from the FNA was sent for cytology study. The cytological report did not show the presence of any abnormal cells, and suggested strongly a possible benign lymphoepithelial lesion of parotid glands. A computerized tomography scanner (CT Scan) examination demonstrated multilocular images in line with cystic benign lymphoepithelial lesions of parotid glands (Fig. 2A and B). The treatment with antiretroviral drugs was initiated, based on the percentage of CD4 T-cell (<20%). The parotid gland swellings, as well as the cystic lesion in the floor of the mouth completely regressed three months after the beginning of ART (Fig. 3A and B). CT Scan examination performed at six months follow up, confirmed the physical reduction of parotid glands (Fig. 4A and B). The patient, now 4-year-old, has since been clinically free of any salivary gland lesion (Fig. 5).

3. Discussion

Oral mucoceles are common lesions of the oral cavity [18,19]. The etiology is traditionally believed to be linked to previous trauma in minor salivary gland bearing areas [18,19]. We usually prefer the term of “mucocele” in absence of any histological report

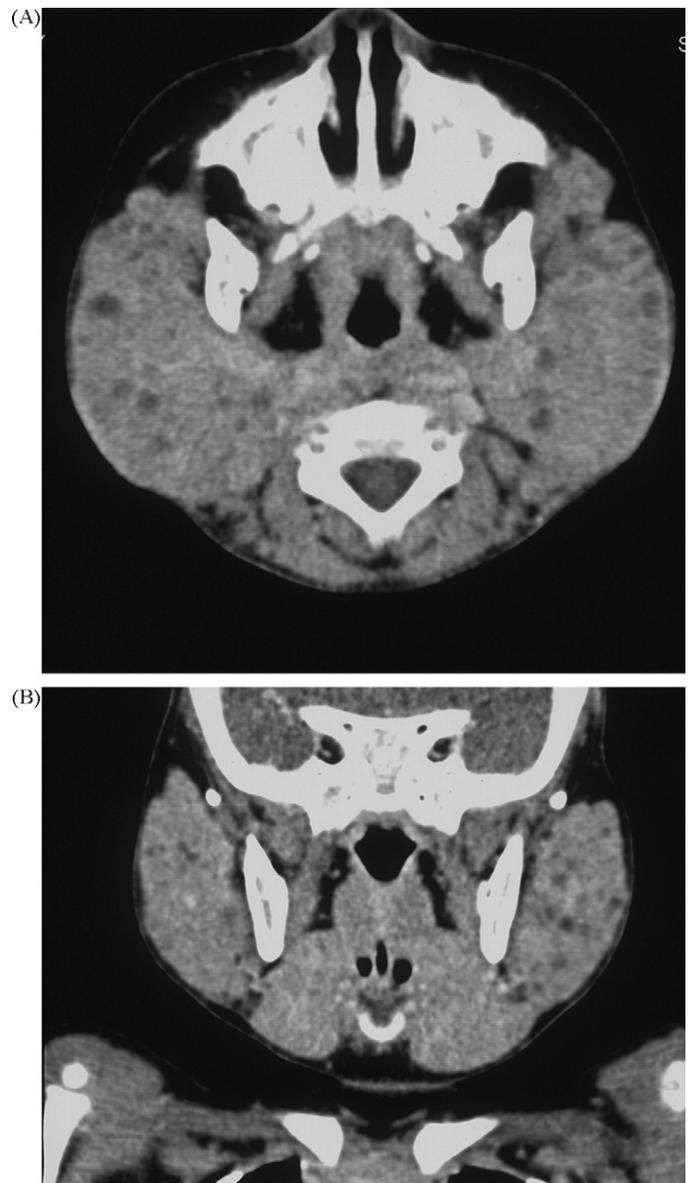


Fig. 2. Radiological expression of BLEL on CT Scan, (A) axial and (B) coronal, of parotid glands of the patient in Fig. 1.

confirming either the extravasation phenomenon, or the retention cyst.

The association of mucoceles (ranulas) and HIV-infection has been mentioned in the literature [11–13]. Chidzonga [12] has reported 88.5% of HIV-infected patients from a study group of 38 cases of ranula. He further raised the question of considering mucoceles as a HIV/AIDS associated oral lesion [11]. A positive answer to this later question would imply a sort of paradigm shift on the treatment planning for mucoceles on HIV-positive patients, taking in consideration for example, the very positive and spectacular response of BLEL to antiretroviral therapy.

The treatment of mucoceles is currently based on a surgical approach, irrespective of the histological type. Different authors have proposed variety of modified surgical procedures, but there is a generally accepted consensus on the excision of associated salivary gland with or not the excision of the cystic lesion [16–19]. Sclerotherapy [15] and the use of botulism toxin [20] are also presented as alternative to surgery for the treatment of mucoceles



Fig. 3. Clinical appearance of the very same patient shown in Fig. 1, three months after the start of antiretroviral therapy. (A) right side) and (B) left side.

with promising results. The regression of an oral mucocele following ART, as reported in this paper suggests the possibility of a medical treatment for mucoceles on HIV-positive patients. Indeed there is already a high level of expectation generated by the effect of ART on BLEL of parotid glands on HIV-positive patients. Cases of substantial, if not complete regression of parotid gland swellings following ART have been well documented in the literature [8–10]. It would be of great benefit for HIV-positive patients to be spared of any elective and invasive surgical procedures such as superficial parotidectomy or excision of the ipsilateral sublingual or submandibular salivary glands. These complex surgical procedures are not without complications. Special caution needs to be exercised during these operations to avoid an unsightly facial scar and to preserve the facial and the lingual nerves as well as the salivary gland duct (Wharton) [13,16,18,21,22]. The young age of some patients is also an important factor to be considered. If the result we have observed in this case is effectively related to the ART, then the link between HIV infection and mucoceles, including ranulas, may be consolidated, and the possibility of causative relationship may be hypothesized.

We need to bring to the attention of the readers the fact that the complete resolution of the intraoral mucocele in this case may also not be attributed exclusively to the effect of ART. Some mucoceles are short-lived lesions that rupture and heal by themselves [18,19]. Zhi et al. have reported in their publication two cases of ranula that did not recur following fine needle aspiration and observation for six months [23]. One possible explanation for such results is that, after spontaneous rupture or fine needle aspiration, usually done

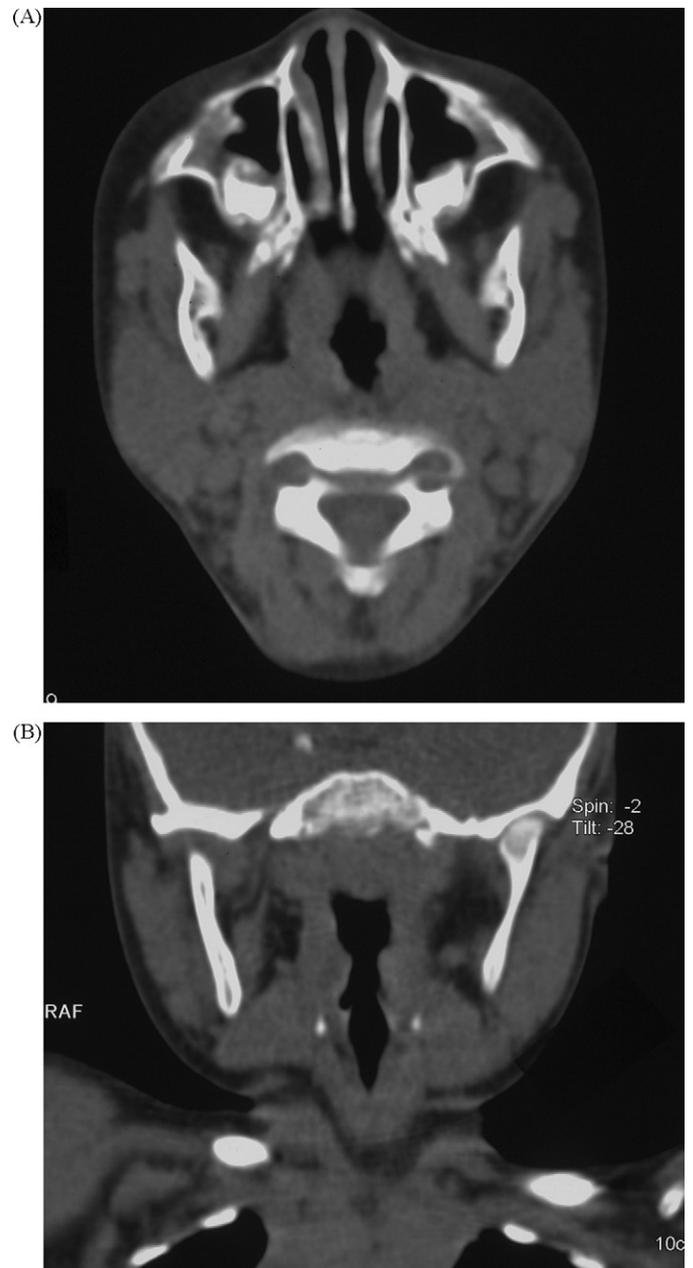


Fig. 4. Radiological confirmation, six months after the ART of the spectacular reduction of parotid salivary glands. (CT scan: A, axial view; and B, coronal view).

with large needle in the case of thick mucus, a permanent fistula may be created and provide thus a new exit orifice for the accumulated saliva. Such a scenario may be considered in case of small and superficial mucoceles that can, time and again, spontaneously rupture and heal or recur [18,19]. The difference with the case we have observed is based on the size of the lesion, the location in the floor of the mouth, and most importantly on the overall pattern of the involution of the oral mucocele, concomitantly with the parotid gland swellings. No further recurrence has been observed at two-year follow up. The rate of recurrence for ranulas is reportedly very high following simple drainage or marsupialisation [18]. It would not be, therefore, an over statement to consider that the regression of the oral mucocele as presented in this paper, may be included in the specific context of the interaction between ART and HIV-associated salivary gland diseases in HIV-infected patients.



Fig. 5. Two years follow up under ART, patient presents no sign of any salivary gland disease.

4. Conclusion

It is generally accepted that benign lymphoepithelial lesions of parotid glands on HIV-positive patients are part of HIV-SGD. It is also well documented that ART has a real therapeutic effect on the parotid gland swellings on HIV-positive patients. It is not, however, completely clear at this stage that mucocèles are also part of HIV-SGD. Further more the response of oral mucocèles to ART is not known. Further studies on the interaction between oral mucocèles, including ranulas, with HIV infection are, therefore, needed, not only to clarify the nature of relationship between HIV and mucocèles, but also to ascertain the possible effect of ART on these lesions.

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