



Recurrent Lymphoepithelial Parotid Cyst in HIV Negative Treated with Mixture of Bleomycin and Doxycycline Solution: A Case Report

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Abstract

Introduction and importance: Lymphoepithelial parotid cysts are rare in HIV-negative patients and recurrence rate is very low when they occur. As of our knowledge, there is no similar reported case in the English literature.

Case presentation: We present a case of a 42-year-old female Human retroviral infection (HIV) negative female patient who presented with recurrent parotid swelling. She was diagnosed with recurrent Lymphoepithelial cysts of the parotid treated with a mixture of bleomycin and doxycycline solution. The patient is having a smooth post-operative course at 1- year follow-up.

Clinical discussion: Parotid gland cysts are uncommonly occurring in HIV-positive patients and are very few reported cases. The diagnosis can be made by ultrasound, CT scan, and MRI and FNAC. Its treatment can be frequent fine needle aspiration, simple cyst excision, superficial parotidectomy, radiotherapy, or sclerotherapy. Sclerotherapy is the preferred mode of treatment with low complication and recurrence rate.

Conclusion: Recurrent Lymphoepithelial cysts of the parotid in HIV-negative is extremely rare. It can be managed with minimally invasive procedures by sclerotherapy with better results in a short period.

Introduction

Lymphoepithelial cysts of the parotid gland are essentially salivary duct cysts associated with a dense lymphoid stroma [1-3]. Its incidence in HIV-negative patients is rare and the recurrence rate is shallow [4-6]. There are different management options for Lymphoepithelial cysts of the parotid gland including surgical excision and sclerotherapy. Previously, most surgeons preferred to do superficial parotidectomy which is an aggressive

procedure, potentially associated with different complications including facial nerve injury [7-9]. Currently, Sclerosant therapy is preferred with a low complication rate and better results [10]. We are reporting HIV-negative patients with recurrent Lymphoepithelial cysts of the parotid gland after being treated with mixed bleomycin and doxycycline which will be the first case to be reported in English literature.



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Case presentation

A 42-year-old female patient presented with left parotid area swelling of 2 years. The swelling increases in size gradually and reaches the current size. Previously, 6 years back after the swelling appeared for 3 years she went to one of the peripheral hospitals, and a simple excision of the cyst was done. Currently, she presented with painless swelling over the same area for a 2-year duration. The swelling increased in size gradually to attain the current size on physical examination there is an old scar over the area. There is a 4cm*3 cm soft, non-tender mass over the left parotid area with no skin color change. Fine needle aspiration cytology was done and showed Lymphoepithelial cysts. The patient was imaged with MRI and the finding was a T1 hypo and T2 hyperintense oval lesion in the inferior pole of the parotid gland (Figure 1 A,1B).

Serology for retroviral infection was done and it turned negative. She was appointed for a minimally invasive procedure, sclerosant therapy (mixing bleomycin and doxycycline) as an outpatient management. First, a doxycycline solution was prepared by dissolving 100mg of doxycycline in 10 ml of normal saline. Also, Bleomycin hydrochloride solution was prepared by dissolving 15 units by 5ml of normal saline. Bleomycin solution was injected at a dose of 0.5 unit/kg body weight. The patient was 62kg. So the maximum dose of bleomycin solution was 31 units nearly 10ml and doxycycline solution was 10ml per day. The solutions were mixed. Under ultrasound guidance about 8ml of clear fluid was aspirated from the cyst. An equal amount (8ml) of the mixed solution was injected into the cyst cavity. The patient was observed for about 2 hours in the hospital and she was discharged home. There was no complication (no sign of allergic reaction). At 2-month post procedure follow up the cyst was resolved (Figure 2). And seen white under Ultrasound evaluation (Figure 3).

Discussion

Parotid gland cysts are uncommon and there are very few reported cases [1,5,6]. It forms when the cysts replace parotid tissue due to lymphocytic infiltration predominantly by CD8 cells [4,5]. This cyst usually occurs in HIV-positive patients, unlike our cases which are HIV-negative [1-8]. It may also be associated with Sjögren syndrome. They are usually bilateral and present as a slow-growing multilocular or unilocular soft painless swelling [3,6,10,]. The disease shows female predominance (60-80%). The diagnosis can be made by radiologic imaging (like ultrasound, CT scan, and MRI) and FNAC [1,9]. Previously the lymphoepithelial cyst was treated by frequent fine needle aspiration and decompression, simple excision of the cyst, superficial parotidectomy (commonly), radiotherapy, and sclerotherapy [1,3,7,9]. Fine needle aspiration and simple excision are mostly associated with recurrence.

Superficial parotidectomy with facial nerve sparing is an aggressive procedure with possible complications like facial palsy and scarring [1,3,4,8]. Radiotherapy also may cause xerostomia and secondary malignancies in the irradiated areas [3,4,9].

Sclerotherapy has many advantages over other procedures. As it is a short procedure (can be done as an outpatient), no exposure to radiation or general anesthesia, very little recurrence rate, and there will be no scar. Moreover, the disadvantage of sclerotherapy is minimal, limited to small surgical site swelling which will subside by itself. However, patients should be carefully evaluated for drug allergy, especially for bleomycin [1,3,9,10].

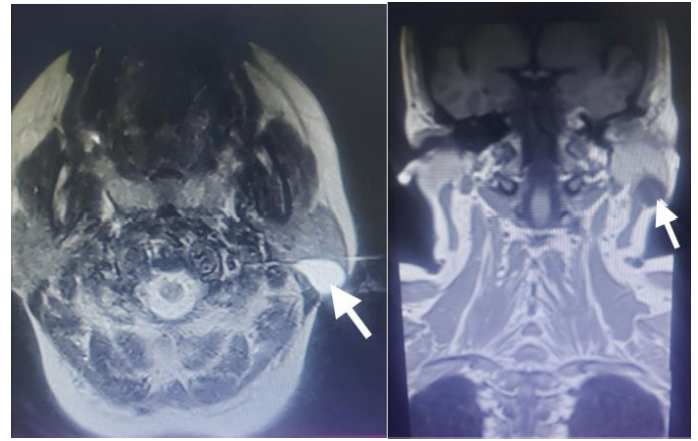


Figure 1: Preoperative image of MRI: The arrow shows the lesion area.



Figure 2: Post-procedure image of the patient with a previous scar.

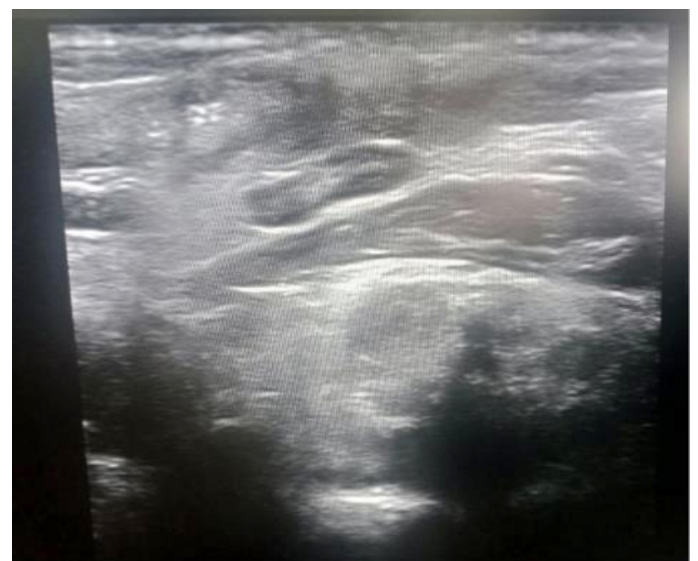


Figure 3: Control ultrasound after 2 months of sclerosant injection.

Using the synergistic effect of bleomycin and doxycycline (mixing them) as sclerotherapy is shown to be very effective and associated with minimal side effects [4,9,10].

Conclusion

Recurrent Lymphoepithelial cysts of the parotid gland in HIV-negative is extremely rare. It can be managed with minimally invasive procedures by mixing bleomycin and doxycycline, and better results will be seen within a short period.

Declarations

Ethics approval and consent to participate: Both ethical clearance and consent to participate were approved by the institutional review board for this case report.

Consent for publication: Ethical approval is held to be unnecessary by St. Paul's Hospital Millennium Medical College Institutional Review Board as this is a single rare case encountered during clinical practice.

Availability of supporting data: All data and materials are available upon request by the Editor-in-Chief.

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Authors' contributions

All authors contributed to different aspects. Abreham Ayele and Habtamu oljira give sclerosant injection to the patient and wrote the final case report and are also following the patient. Berhanu landuber and Tafese Gudissa wrote the case presentation and wrote the draft of the case report. All authors read and approved the final manuscript.

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