

A Case of Management of Benign Lymphoepithelial Cysts of the Parotid Gland in Patient Infected with Human Immunodeficiency Virus

Ji Hoon Kim

Department of Otorhinolaryngology-Head and Neck Surgery, Seoul Medical Center, Seoul, Korea

인간면역결핍 바이러스 감염자에서 발생한 이하선 양성 림프상피낭종의 치료 1예

김 지 훈

서울의료원 이비인후과

Received June 3, 2014

Revised August 20, 2014

Accepted August 20, 2014

Address for correspondence

Ji Hoon Kim, MD

Department of Otorhinolaryngology-

Head and Neck Surgery,

Seoul Medical Center,

156 Sinnae-ro, Jungnang-gu,

Seoul 131-795, Korea

Tel +82-2-2276-8635

Fax +82-2-2276-7870

E-mail travet@hanmail.net

Benign lymphoepithelial cysts (BLEC) are the most common cause of the parotid gland swelling in human immunodeficiency virus (HIV)-positive patients. The diagnosis of these HIV-associated BLEC is made based on history, physical examination, computerized tomography or magnetic resonance image, and fine-needle aspiration biopsy. The treatment is not established. We report the first case in Korea of a 43-year-old HIV-positive male with a 1-year history of enlargement in both parotid glands.

Korean J Otorhinolaryngol-Head Neck Surg 2015;58(6):417-20

Key Words Benign lymphoepithelial cysts · Human immunodeficiency virus · Parotid · Parotidectomy.

Introduction

Human immunodeficiency virus (HIV)-positive patients rarely develop benign lymphoepithelial cysts (BLEC) in the parotid gland. BLEC is characterized by painless and commonly bilateral enlargements of the parotid gland, CD8 lymphocytosis, and cervical lymphadenopathy. The management of these disorders includes repeated simple aspiration, antiretroviral medication, parotidectomy, radiotherapy, and sclerotherapy.¹⁾ Parotidectomy is known to have the best cosmetic result and the lowest recurrence rate.²⁾ We present the first case of BLEC from a HIV-positive patient in Korea.

Case

A 43-year-old, HIV-positive and bisexual man was referred to the otolaryngology department for bilateral en-

largements of the parotid glands (Fig. 1A). There were no other symptoms. He had these parotid swellings for 1 year. They were cosmetically unsightly. He had been known to have HIV infection for 9 years. The submandibular and sublingual glands were unremarkable. Current CD4 lymphocytes were 155/mL (normal 480–1580/mL), which was 9% of the total amount of lymphocytes (normal 29–57%). CD8 lymphocytes were 81% (normal 11–38%). He had been taking 300 mg tenofovir disoproxil fumarate and 600 mg efavirenz once a day for 7 weeks. 5 weeks before the operation, He was prescribed 400 mg sulfamethoxazole mixed with 80 mg trimethoprim once per day. White blood cell (WBC) count was 7400/ μ L 2 weeks before the operation. A computed tomography showed multiple, thin-walled, low attenuated and cystic masses within both parotid glands, and multiple cervical, homogenous, noncavitated, and sharply outlined lymphadenopathy through the supraclavicular region (Fig. 1B). A Fine-

needle aspiration biopsy (FNAB) done under direct vision showed squamous epithelial cells, mature lymphocytes without any sign of malignancy, and a few giant cells with many

macrophages. The probable diagnosis of BLEC associated with HIV infection was made. It was explained that there were many variable treatments. At that time he had hidden

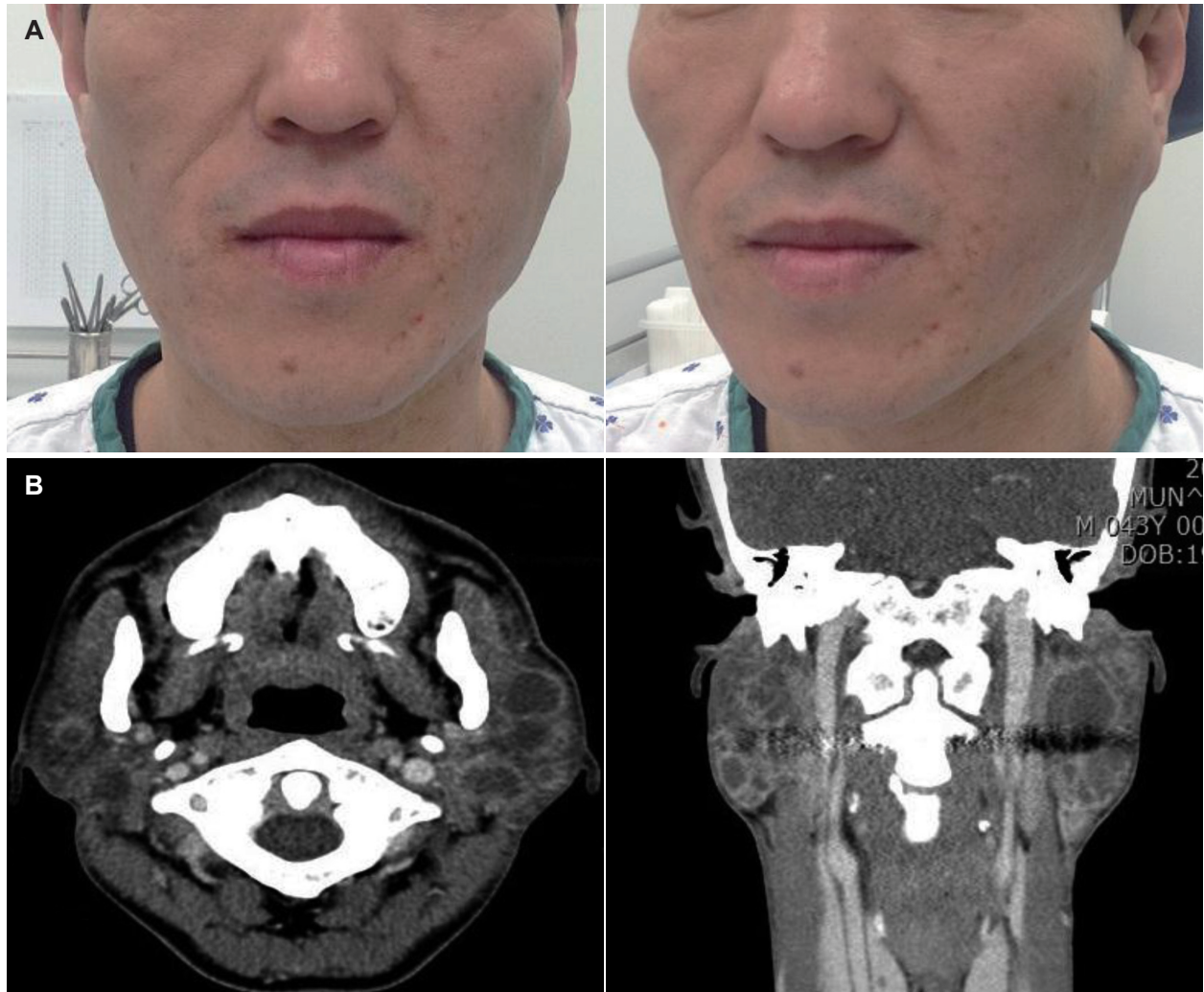


Fig. 1. HIV-positive man with bilateral benign lymphoepithelial cysts (A). Computed tomography scan of HIV-positive patient with multi-loculated thin-walled cysts within both parotid glands. This is the typical radiographic presentation of benign lymphoepithelial cysts (B). HIV: human immunodeficiency virus.

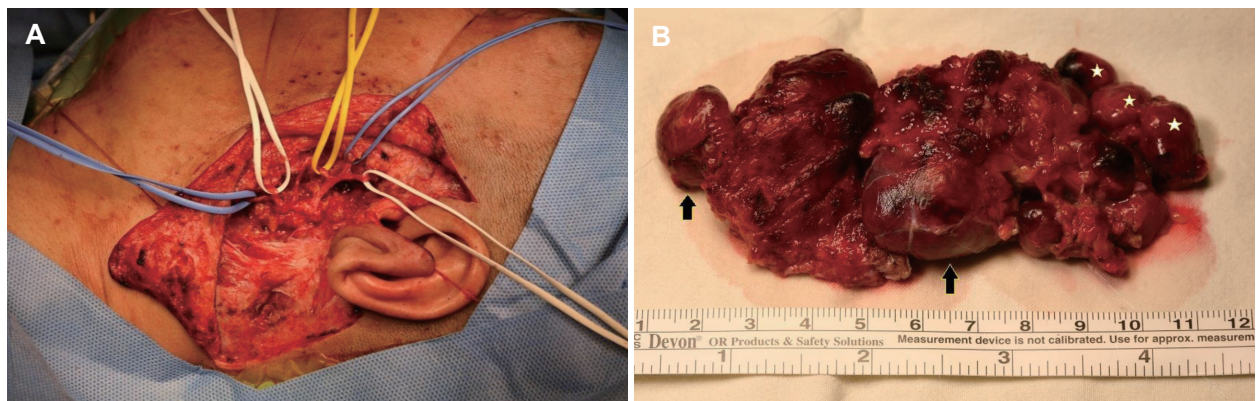


Fig. 2. Wound bed after total parotidectomy showing all branches of the facial nerve (A). 10 cm sized specimen showing multicystic masses (arrows) within the parotid gland and cystic lymph nodes (stars) (B).

the fact of his HIV infection from his family. He wanted to be treated for his disfiguration as soon as possible and return to his daily life like being treated for the common neck mass. So, the operative management was chosen for him. The left parotid mass was bigger than the right mass. Also, he was burdened about both side operations. So first, the surgical excision for the left parotid mass and the second operation after observation with medication for the right parotid were planned. A total parotidectomy was performed for 10×9.5 cm sized BLECs on the left (Fig. 2A). The lesions were removed and all branches of facial nerve were saved. Gross examination of a specimen showed intra-parotid cysts with enlarged lymph nodes

(Fig. 2B). He got augmentation for parotidectomy defect with a sternocleidomastoid muscle flap. Histologic findings revealed that cysts contained keratinous material and cyst wall was lined with stratified squamous epithelium and surrounded by lymphoid tissue with germinal center (Fig. 3). Postoperative WBC count was 7000/μL and antibiotic was taken for only 1 week postoperatively. The same medication has been prescribed for HIV continuously after the operation. He was pleased with the cosmetic outcome. He was very satisfied with the fact that unbeknown to his family, he could continue the treatment for AIDS and do his work with his colleagues naturally. We did follow-up for 15 months, and no recurrence

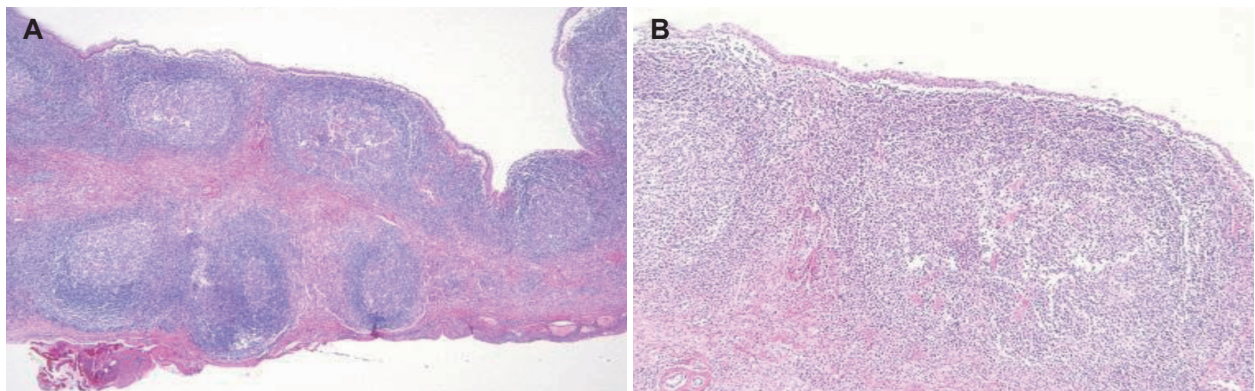


Fig. 3. Histologic findings of the mass. Cysts contain keratinous material (H&E stain, ×40) (A). Cyst wall is lined with stratified squamous epithelium and surrounded by lymphoid tissue with germinal center (H&E stain, ×100) (B).



Fig. 4. This patient still remained recurrence free 15 months after parotidectomy, at which time he was working.

was found (Fig. 4).

Discussion

BLEC of the parotid gland in HIV-positive patients were first described as of 1987.³⁾ Originally, cystic lesions of salivary glands were quite rare. Recently the incidence rate of these lesions has been increased with increasing frequency of HIV infection since then. BLEC in HIV-positive patients are found not infrequently in the Western society now.^{1,3)}

Diseases of the head and neck develop in over 50% of HIV-positive population, and mostly involve the parotid gland.⁴⁾ When neck mass is found in these patients, we should consider not only BLEC but infection or malignancies in the differential diagnosis. However, BLEC which is quite rare in general, is found most commonly in HIV-positive parotid glands. So, once a patient without any information of his status is diagnosed with BLEC, an HIV test should be performed. We can suspect the possibility of BLEC in HIV-positivity when there are two characteristics as follows. First, BLEC in HIV-positivity usually are found bilaterally in up to 80% and secondly, these are multi-cystic in up to 90%.^{5,6)} The multi-cystic presentation is pathognomic and easily recognizable on radiologic studies. And the diagnosis of BLEC is made based on FNAB, which typically shows fluid with clumps of epithelial cells in a background of scattered lymphocytes. Additionally histopathology can be helpful on diagnosis, which reveals squamous or cuboidal epithelial lined cysts containing a pale and homogeneous material populated by numerous foamy macrophages and lymphocytes.

The management is not established. To date, various managements have been introduced, including close observation, antiretroviral medication, repeated aspiration, low-dose radiotherapy, parotidectomy, and injection sclerotherapy. Parotidectomy has been advocated by some investigators.^{7,8)} The main cause for treatment is cosmetic. So, when the treatment can provide the best cosmetic outcome, it will be the gold standard treatment. To date, the only treatment to show a complete response consistently without recurrence is parotidectomy.²⁾ However, operative morbidity and mortality, and the exposure of hospital personnel to HIV-infected body fluids are weakness of surgery. Recent studies have shown that the rate of postoperative infection in HIV-positivity is similar to that in the normal population.⁹⁾ BLEC can become quite large, and

as this case, some patient desires surgical excision despite the benign natural course. Also, when the diagnosis is uncertain, surgery is mandatory to obtain material for definite histopathological diagnosis.⁸⁾ Steehler, et al.²⁾ said that parotidectomy offered the best cosmetic result for the patients after long-term follow-up, and concluded that surgery was the treatment of choice for BLEC. As result, there is no definite indication for parotidectomy. But we can choose the surgical management when doctor and patient all set importance on the cosmetic result most of all or when we don't know the definite preoperative histology even if FNAB is done right.

Because the early treatment for HIV infection is difficult due to the social atmosphere even though the increasing incidence of HIV infection in the oriental society, we can expect to meet more cases of BLEC in future. Therefore, it is said that this report is of great significance to the otolaryngologists who will be faced with the challenge of managing these patients diagnosed with BLEC from a more diagnostic and treatment perspective.

Acknowledgments

We are greatly indebted to Radiology department, Seoul Medical Center (Dong Hun Lee, MD., Ph.D.) for interpretation of image.

REFERENCES

- 1) Huang RD, Pearlman S, Friedman WH, Loree T. Benign cystic vs. solid lesions of the parotid gland in HIV patients. *Head Neck* 1991;13(6):522-7.
- 2) Steehler MK, Steehler MW, Davison SP. Benign lymphoepithelial cysts of the parotid: long-term surgical results. *HIV AIDS (Auckl)* 2012; 4:81-6.
- 3) Morris MR, Moore DW, Shearer GL. Bilateral multiple benign lymphoepithelial cysts of the parotid gland. *Otolaryngol Head Neck Surg* 1987;97(1):87-90.
- 4) Yengopal V, Naidoo S. Do oral lesions associated with HIV affect quality of life? *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;106(1):66-73.
- 5) Dave SP, Pernas FG, Roy S. The benign lymphoepithelial cyst and a classification system for lymphocytic parotid gland enlargement in the pediatric HIV population. *Laryngoscope* 2007;117(1):106-13.
- 6) Kooper DP, Leemans CR, Hulshof MC, Claessen FA, Snow GB. Management of benign lymphoepithelial lesions of the parotid gland in human immunodeficiency virus-positive patients. *Eur Arch Otorhinolaryngol* 1998;255(8):427-9.
- 7) Shaha AR, DiMaio T, Webber C, Thelmo W, Jaffe BM. Benign lymphoepithelial lesions of the parotid. *Am J Surg* 1993;166(4):403-6.
- 8) de Vries EJ, Kapadia SB, Johnson JT, Bontempo FA. Salivary gland lymphoproliferative disease in acquired immune disease. *Otolaryngol Head Neck Surg* 1988;99(1):59-62.
- 9) Cavasin H, Dola T, Uribe O, Biswas M, Do M, Bhuiyan A, et al. Postoperative infectious morbidities of cesarean delivery in human immunodeficiency virus-infected women. *Infect Dis Obstet Gynecol* 2009;2009:827405.