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Benign lymphoepithelial cyst of parotid gland without HIV infection: case report and literature review

Liao *et al.* BLEC of parotid gland without HIV

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Abstract

BACKGROUND

Benign lymphoepithelial cyst (BLEC) of the parotid gland is a rare benign embryonic-dysplastic cystic tumor in the anterolateral neck that occurs most commonly in HIV-positive adults and rarely in non-AIDS patients. The main presentation is a slow-growing, painless mass, and secondary infection may cause acute inflammatory symptoms.

CASE SUMMARY

A 44-year-old Chinese male patient presented with a 1-year history of a mass in the left side of the neck. On physical examination, a mass similar in size and shape to a quail egg was found in the left parotid gland. The mass was tough, without tenderness, and easily moveable. The results of HIV tests, including antibody and nucleic acid tests and CD4+ T cell examination, were negative. Imaging examination revealed a left parotid gland mass. The patient underwent surgical treatment, and BLEC was diagnosed based on postoperative pathology. After 2 years of follow-up, the patient survived well without related discomfort.

CONCLUSION

The detailed characteristics of a BLEC in a patient without HIV infection contribute to an improved understanding of this rare disease.

Key Words: Benign lymphoepithelial cyst, Parotid gland, HIV, Case report

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Core Tip: Benign lymphoepithelial cyst (BLEC) of the parotid gland is a rare lesion seldom found in non-AIDS patients. BLECs are benign embryonic dysplastic cystic tumors that typically occur in the anterolateral part of the neck. We present a case of BLEC of the parotid gland in a non-HIV-infected patient in order to improve clinicians' understanding of the disease.

INTRODUCTION

Benign lymphoepithelial cyst (BLEC) of the parotid gland, which is also known as branchial cleft cyst, is a rare benign cystic neoplasm of embryonic dysplasia. It usually occurs in the anterolateral region of the neck but has been reported in the oral cavity or parotid gland in rare cases [1,2]. This disease mainly presents as a slow-growing tumor and is not associated with recurrence or metastasis. These tumors usually occur in patients with HIV infection and are rarely encountered in non-HIV-infected patients [3]. To improve clinicians' understanding of this rare disease, the present report describes the imaging, histopathological, and diagnostic characteristics of a parotid gland BLEC found in a 44-year-old, non-HIV-infected patient. The case description is followed by a review of the relevant literature.

CASE PRESENTATION

Chief complaints

A 44-year-old Chinese man was admitted to the hospital on September 1, 2020 due to a painless mass on the left side of the neck that had been present for 1 year.

History of present illness

One year previously, the patient incidentally noticed a painless mass on the left side of his neck approximately 2.0 cm × 2.0 cm in size. The patient had neck swelling occasionally, and the severity of these symptoms gradually increased over the course of the disease. The patient reported no tenderness, redness, swelling, or heat pain at the

mass; no earache, swelling, or pain around the ear; and no cheek pain, numbness or difficulty when opening the mouth.

5 *History of past illness*

The patient had a history of hypertension for 1 mo and was regularly taking nifedipine sustained release tablets and Irbesartan tablets as antihypertensive treatment.

1 *Personal and family history*

The patient had no personal or family history of benign or malignant tumors.

Physical examination

7 Physical examination on admission revealed a temperature of 36.5°C, resting respiratory rate of 20 breaths/min, heart rate of 80 bpm, and blood pressure of 117/70 mmHg. On physical examination, a mass similar in size and shape to a quail egg was found in the left parotid gland, measuring about 2.0 cm × 2.0 cm in size. The mass felt tough, without tenderness and was easily movable with no local redness, swelling, heat or pain. No abnormalities were observed in the contralateral parotid gland.

Laboratory examinations

1 The laboratory test results showed a white blood cell count of $6.16 \times 10^9/L$, with 74% neutrophils, 135 g/L hemoglobin, **8** and a platelet count of $277 \times 10^9/L$. The erythrocyte sedimentation rate was 2 mm/h, and all results for routine urine, stool, liver and kidney function tests as well as electrolyte and blood biochemical index levels were within normal ranges. The results of HIV tests, including antibody and nucleic acid tests as well as CD4+ T cell examination, were negative. Furthermore, all results of testing for **1** carcinoembryonic antigen, neuron-specific enolase, cytokeratin 19 fragment, squamous cell carcinoma antigen, blood coagulation, and immune indexes were negative.

Imaging examinations

On August 31, 2020, ultrasound of the neck recognized a 2.2 cm × 2.2 cm hypoechoic mass in the left parotid gland, with a clear boundary, coarse calcification foci, and no blood flow signal (Figure 1). On September 2, 2020, neck computed tomography (CT) showed a round cystic lesion in the left parotid gland, with a maximum cross-sectional area of approximately 2.3 cm × 2.2 cm, uneven density, small patches with slightly high-density shadows, and clear boundaries (Figure 2A). On contrast-enhanced CT, the cyst wall was slightly enhanced in the arterial phase, but no obvious enhancement was observed in the cyst (Figure 2B). The enhancement degree in the venous phase was similar to that in the arterial phase (Figure 2C). No abnormality was found in the right parotid gland.

Pathology

In general, the lesion was a pale round nodule with a complete capsule and small volume, accompanied by a small amount of parotid tissue attachment. The size was about 2.2 cm × 2.0 cm × 1.8 cm. The surface of the tumor was smooth and soft, and the surface skin mucosa was not abnormal, and the color was not special. The section was cystic, and the cyst was soybean dreg-like material with a wall thickness of 0.1 cm. Under light microscopy, the cyst wall was laminated squamous epithelium without epithelial nail process and the surface layer was mostly incomplete keratosis. The epithelium was surrounded by a large number of lymphoid stroma with lymphoid follicular formation and a center of occurrence (Figure 3). The histological features of the tumor were consistent with a diagnosis of BLEC. The clinical and pathological data of this case are presented in Table 1.

FINAL DIAGNOSIS

The patient was diagnosed with a non-HIV-infected BLEC of the parotid gland on the basis of history, clinical features, and ancillary examination (imaging and histopathology).

TREATMENT

Because BLECs are benign, most cases of BLEC have a good prognosis. The principles of treatment are early diagnosis, infection control, and complete resection of the lesion without facial nerve injury. Treatment options include observation, repeat aspiration, sclerotherapy, radiotherapy, and surgery.

During the treatment period, the patient in the present case did not stop Nifedipine and Irbesartan for treatment of hypertension. Blood pressure and electrocardiography were normal at admission, and surgical treatment was possible under close monitoring of blood pressure. During surgery, care must be taken to identify the internal and external carotid arteries and vagus nerve, hypoglossal nerve, glossopharyngeal nerve, and superior laryngeal nerve to avoid injury to these structures. Finally, the patient and his family members chose surgical treatment (the specific surgical method was termed left parotid gland tumor + partial superficial lobectomy + facial nerve exploration and protection + fascia flap plasty). The operation was successful, and the patient returned to the ward safely after surgery.

OUTCOME AND FOLLOW-UP

The patient was followed up for 2 years. CT/magnetic resonance imaging (MRI) and color ultrasound were not performed during follow-up, because the patient felt no obvious symptoms.

DISCUSSION

BLEC is a rare cystic neoplasm with benign embryonic dysplasia. The first case of BLEC was reported by Hildebrandt in 1895, but only 21 cases were published by researchers through 1981. With the emergence of the HIV epidemic, the incidence of BLEC in the parotid gland gradually increased, and researchers found that BLECs were closely related to HIV infection in most cases, with BLEC as one of the early clinical manifestations. Around 3%–6% of HIV-positive adults and 1–10% of HIV-positive children experience BLEC symptoms. In sharp contrast, BLECs are rarely found in non-

HIV-infected patients, and the exact prevalence of BLEC in this population has not been reported [1-4]. In the present case, the results of HIV tests, including antibody and nucleic acid tests as well as CD4+ T cell examination, were negative. In previous reports, the disease was also known as branchial cleft cyst and usually observed in either the mandibular angle at the bottom of the outer regions or in the anterior portion of the sternocleidomastoid. It was found to occur less frequently in the mouth or parotid gland. In addition, its distribution in men and women is uniform. Although catheter jam seems to be a cause, the source of the blockage is often not obvious. Furthermore, its pathogenesis remains unclear. Currently, the majority of researchers believe that extraglandular lymphoid infiltration and/or intra-glandular lymphoid hyperplasia with duct obstruction and/or epithelial embedding is the main cause.

BLEC is a slow-growing tumor not associated with recurrence or metastasis [5-8]. The main symptom of this disease is a slowly growing, painless swollen mass, often combined with local symptoms such as dysphagia, dysphagia, dyspnea and stridor, and secondary infection can manifest as acute inflammatory symptoms, such as redness, swelling and fever [3,8]. The diagnosis of BLEC mainly depends on history, clinical manifestations, and preoperative auxiliary examination. Preoperative auxiliary diagnostic procedures include CT, MRI, ultrasound and fine needle aspiration (FNA) [9]. To date, imaging characteristics of this disease have been rarely reported. By reviewing the relevant literature [1,9], we found that the CT manifestations of a typical BLEC are a single, thin-walled round or oval cystic foci in the parotid gland, with clear boundaries and low density in the cyst. On enhanced scanning, only the cyst wall shows mild to moderate enhancement, and no enhancement is seen in the cyst. In the present case, CT showed that the small patches with slightly high-density in the parotid gland were caused by viscous fluid in the capsule or lesions complicated with infection. MRI mainly showed a single thin-walled cystic lesion in the parotid gland, which was round or oval in shape, with a clear boundary, and the cyst wall showed a low signal. Intracystic T1W1 showed low signal, while T2W1 showed high signal. Moreover, the cyst wall was significantly enhanced on enhanced scanning, but no enhancement was

found in the cyst [2]. Ultrasonic features of the lesions included a circular shape with a complete capsule and smooth lining, and cystic wall blood flow signals were not rich. The epithelial cells in a BLEC may contain keratin, and shed inflammatory cells can form a viscous gel-like yellowish-white liquid [10, 11]. The internal echo of the cyst can generally appear as one of three types, including coarse granular hyperecho and cloud weak echo mixed signal, clear cystic echo through sound, and cluster weak echo and cystic echo mixed. In this case, ultrasound showed typical mixed signals including coarse granular hyperechoic signal and cloud weak echo signal. FNA can be used as an important auxiliary method for the clinical diagnosis of lateral cervical lesions. The criteria for FNA cytological diagnosis of BLEC include thick, yellow, pustular fluid; non-nucleated cells; keratinocytes; and squamous epithelial cells of varying maturity [1].

2

Histologically, the walls of BLEC cysts are usually covered with squamous epithelium and/or, in some cases, columnar or cuboidal cells. Lymphoid tissue with or without germinal centers in subepithelial connective tissue is the most prominent morphological feature [12]. Histological examination of the mass case showed that the cyst wall was laminated squamous epithelium without epithelial nail process and the surface layer was mostly incomplete keratosis, surrounded by a large number of lymphoid stroma with lymphoid follicular formation and a center of occurrence. Differential diagnoses include: (1) Warthin tumor [2], which is more common in middle-aged and elderly patients and more likely to occur in the posterior lower pole of the parotid gland, with uneven internal density; (2) intramuscular benign hemangioma [13], which is a fast-flowing intramuscular mass with uneven density and visible fat density in children that requires pathological examination for diagnosis; and (3) lymphoma [14], which is more common in men over 50 years old and appears as irregular soft tissue masses with a large range and uniform density on CT, without obvious calcification, cystic degeneration or necrosis, with diffuse growth to the surrounding area and mostly without adjacent bone destruction, and mild to moderate enhancement on enhanced scanning; (4) thyroglossal duct cysts [15], which present as a painless mass in the front of the neck and are usually dumbbell shaped and movable when the tongue is extended or

swallowed; CT shows a low-density, usually monocular parenchyma lesion during embryonic thyroid migration, mostly located in the midline and associated with the hyoid bone; (5) lymphocele^[16], which typically manifest as a cystic density focus with uniform density, clear boundary, thin cyst wall, no obvious exudation and calcification, and after enhancement, the cyst wall can present slightly uniform enhancement, with no enhancement in the cyst; (6) metastatic lymph nodes^[17], which are accompanied by a history of primary tumor, and on CT exhibit uneven density, calcification, cystic or necrotizing changes, uneven edges, adhesion to surrounding tissues, and obvious annular or peripheral enhancement; and (7) lymphadenitis^[18], which is more common in children and present with local redness, swelling, heat and pain while appearing mostly oval with a thick wall and ring and uniform enhancement without obvious wall nodules and calcification, but with a blurred surrounding fat space. The current treatment principles for this disease include early diagnosis, infection control, and cyst removal, and the treatment options include observation, repeated aspiration, sclerotherapy, radiotherapy, and surgical treatment ^[19]. Observation in asymptomatic patients shows that repeated aspiration therapy is generally ineffective, and cysts will recur within weeks to months. Sclerotherapy is limited in scope and is generally only used for cases in which the cystic fluid can be aspirated, while radiotherapy is mostly used for HIV-infected BLEC ^[3]. For our patient, surgery was the best treatment at present, and attention was paid to identify and avoid injury to adjacent blood vessels and nerves during surgery. In this case, surgical treatment was chosen by the patient, and the specific surgical procedure was termed left parotid gland tumor + partial superficial lobectomy + facial nerve exploration and protection + fascia flap plasty. The potential ² complications of surgical treatment include recurrence, the formation of persistent fistula, and cranial nerve injury, which require close follow-up. In this case, the patient survived well with no related discomfort after 2 years of follow-up.

CONCLUSION

BLEC in the absence of HIV infection is a rare benign cystic tumor of embryonic dysplasia, known as a branchial cleft cyst. The disease progresses slowly. Because the imaging, histopathology, and diagnosis and treatment characteristics of non-HIV-infected BLEC are poorly understood, the present case is reported to increase readers' awareness of the disease.

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