Recurrent Parotid Abscess Causing Facial Paralysis: Tularemia

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Facial nerve paralysis associated with parotid gland mass is usually caused by malignant neoplasms and facial nerve dysfunction due to parotid infection is exceedingly rare. Francisella tularensis is a potential agent of biologic terrorism. Between 11% and 45% of the patients with tularemia have symptoms or signs localized to the head and neck region. We presented a patient with tularemia who developed facial paralysis secondary to recurrent parotid abscess.

Keywords: Tularemia, facial paralysis, parotid abscess.

INTRODUCTION

Facial nerve paralysis due to a parotid mass is most commonly seen in malignant diseases of the parotid gland. However, this paralysis due to parotid gland abscess is exceedingly rare. In this paper, it has been represented; a patient with tularemia, who developed facial paralysis secondary to recurrent parotid abscess, has not been described previously.

Case Report

A 20 year old male patient was admitted to our clinic with a painful swelling on the right side of the jaw. He had been receiving amoxicillin 2x1 gr intravenously for the last five days. The examination revealed a painful firm swelling extending from beneath the right ear lobe through the neck, as well as hyperemia and temperature increase. Oropharynx examination was normal. Otorhinolaryngological examination showed normal results. On admission, laboratory findings were as follows; white blood cell 12.300/mm3, erythrocyte sedimentation rate 65 mm/hour and tuberculin skin test was negative. Chest x-ray showed no abnormality. His medical history was unremarkable, including the absence of diabetes mellitus, HIV infection and malignancy. The patient did not suffer from any immune or endocrine disease and had not taken any other medication in the past. He did not mention any history of diarrhea, vomiting, abdominal pain, joint pain or joint swelling. A CT scan of the parotid region showed an enlarged right parotid gland. A mass with a hypodense, irregular centre and peripheral enhancement was described by CT scan. These findings were consistent with the diagnosis of abscess (Fig. 1). An examination of the patient for facial nerve functions revealed a decrease in motor functions of the right marginal mandibular nerve, as a result of which, the patient was taken into emergency surgery (Figure 2). Surgical drainage was performed under general anesthesia.

After the incision, an elevation parallel to facial nerve trace was performed using a clamp. 15 cc of purulent material was removed. The patient received ceftriaxone 2x1 gr and metronidazole 3x500 mg intravenously for 7 days. Routine culture of the abscess material was negative for pyogenic bacteria, acid-fast organisms and fungi. Pathological examination of drainage material revealed mixed type suppurative inflammation. Blood analysis showed normal results for Anti HIV (-), p ANCA(-), c ANCA(-), C3 and C4. He was discharged on the postoperative 8th day.

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At the time of discharge he could tolerate a regular diet and his swelling and erythema of the parotid region had markedly decreased.

The patient was referred to ear, nose and throat emergency with the same symptoms one month later. He underwent surgery with the diagnosis of recurrent parotid abscess. The multiloculated abscess, extending into the parotid parenchyma, was surgically drained. Samples obtained from the abscess were sent for aerobic and anaerobic cultures and acid fast staining and found to be negative. Tularemia antibody titer was positive in a dilution of 1/320 by a microagglutination test. There was no history...
of contact with animals. The patient was treated with ciprofloxacin 500 mg po every 12 h for 10 days. He was discharged from the hospital 10 days after complete resolution of the infection. He did not have any recurrence of the disease during six months of follow up. Facial nerve paralysis was completely treated.

DISCUSSION

One rare complication of acute suppurative parotitis is a parotid abscess [1]. The most common pathogens associated with acute bacterial parotitis are Staphylococcus aureus and anaerobic bacteria [1]. Streptococcus pneumoniae and Haemophilus influenzae have been reported to be among the most common pathogens in recurrent suppurative parotitis [2]. Parotid abscess in adults is related to poor oral hygiene, long-term debility, and reduction in salivary flow [1]. However, such an abscess can also appear in relatively young and fit adults with no history of oral pathologies [3].

The symptoms of a parotid abscess include marked swelling of the angle of the jaw and pain while eating [4]. Regional lymphadenitis may occur, as well as purulent secretion from Stensen’s duct [4]. Although our patient also had pain and swelling in the jaw, there was no purulent secretion from the stensen’s duct. The treatment includes broad-spectrum IV antibiotics, good oral hygiene, and adequate hydration [4]. If an abscess has formed, surgical drainage is advisable [4]. A parotid abscess is a potentially life-threatening disease, since the inflammation can spread to the head and neck causing fasciitis [5] or deeper head and neck abscesses [6].

Methods include quite radical procedures such as multiple drainage incisions or rising of a full posterior-based flap as for parotidectomy. But it seems that in most cases quite a small procedure is enough [3], especially when modern imaging techniques can assist in the procedure, and broad-spectrum antibiotic therapy is included. During surgical treatment, upper skin flap is elevated. A curved clamp is inserted into the parotid lodge and the abscess pouch is sought for in the direction of the facial nerve branches. The abscess is drained and a Penrose drain is placed. In our case, parotid abscess recurred as early as one month even though a similar surgical procedure was performed.

In the present case, no microorganisms were identified in the culture, which may be caused by performing the drainage five days after antibiotic therapy. In a case report by Orhan et al, no growth was detected in FNAB specimens collected from patients with facial paralysis. The authors attributed this to the antibiotic treatment that the patients were given before FNAB [7].

Facial nerve paralysis associated with a parotid mass suggests the presence of a malignant neoplasm. Eneroth [8] reviewed 2158 cases of parotid gland neoplasms, 46 of which presented in patients with facial nerve paralysis and all of which were malignant parotid tumours. Only a few case reports have documented benign neoplasms and inflammatory processes of the parotid gland as causes of facial nerve palsy and facial nerve dysfunction as a result of parotid infection is exceedingly rare [9]. The severity of facial nerve dysfunction is probably due to the virulence of microorganism, perineuritis, and nerve compression [7].

In a case report by Marioni et al, parotid abscess leading to facial paralysis was present in a 74 year old man with diabetes [10]. Candida albicans grew in culture. After the administration of an antifungal therapy and controlling the diabetes, the patient recovered well. No growth occurred in the cultures when the abscess recurred in our case. Tularemia was suspected for the current case since tularemia is an endemic disease in our region. Tularemia antibody titer was positive in a dilution of 1/320 by microagglutination test. After the second intervention and the administration of appropriate antimicrobialtherapy for tularemia, facial paralysis recovered completely. To the best of our knowledge ours is the first reported case of F. tularensis as a cause of recurrent parotid gland abscess associated with facial nerve palsy.

Tularemia, caused by Francisella tularensis, is a widely distributed zoonosis in the world [11,12]. Francisella tularensis is a Gram-negative, strictly aerobic and facultative intracellular coccobacillus and a potential agent of biologic terrorism [13,14]. It is found in all age groups, with the majority of cases being young [15]. Francisella tularensis may enter into body from skin, respiratory system and mucous membranes such as conjunctiva and oropharynx [16,17,18]. Transmission of F. tularensis to humans occurs predominantly through the bite of a tick or an animal carrying the agent or through contact with contaminated animal products, aerosol droplets, or ingestion of contaminated food or water [11]. However, the most important source of F. Tularemia transmission is the water supply, since the bacteria may remain alive in water for several months [19]. The clinical picture may vary depending on several factors such as the route of transmission, virulence of the microorganism strain and the immune condition of the host [17,18].

In a study by Helvacı et al, lymphadenopathy was found to be the most common clinical finding in patients. 175 of the 205 patients had lymphadenopathy. Seventy of cervical lymphadenopathies (42.6%) became fluctuant and underwent surgical drainage. There were no cases of parotid involvement [15]. Helvacı et al. reported that the majority of cases were young females [15], which may be explained by the increased exposure to water in these groups. In a study by Celebi at al, all patients had oropharyngeal form of the disease [20]. With regard to the clinical findings of the patients, lymphadenitis (97%) and fever (84 %) were found to be the most common findings. 23% of the patients had oropharyngeal ulcer. Between 11% and 45% of the patients with tularemia have symptoms or signs localized to the head and neck region [21]. The clinical course of tularemia may vary according to the site of entry and the virulence of the microorganism. Infected individuals may remain asymptomatic or the disease may progress to clinical forms, such as ulceroglandular, ocular, oropharyngeal, glandular, typhoid, pleuropulmonary, and gastrointestinal forms [22]. Ulceroglandular form is the most common one, and characterized by fever, formation of pustule or ulcer in the inoculation area and lymphadenitis in the regional lymph nodes [23].
However, recent reports from Turkey defined the oropharyngeal type as the predominant form [15,24]. Because of clinical characteristics, we think that our case is a glandular form. Luottonen et al. analyzed 127 tularemia cases in the context of an otolaryngology practice and found that in oropharyngeal tularemia, the primary lesion occurs in the mouth because of ingestion of infected food or contaminated water [25]. Enlarged lymph nodes, especially in the cervical and periauricular area, are seen in approximately 85% of patients and may be the initial or only sign of infection [26]. The gold standard of diagnosis is a positive culture for tularemia [20]. Francisella tularensis is difficult to be isolated from infected materials, so selective cultures have to be used [27,28]. Evans et al. isolated the bacteria from 5.5% of their cases, whereas Bevanger et al. isolated the bacteria from only one of 57 patients [27,29]. We used the microagglutination test for diagnostic confirmation (1/320). Streptomycin or gentamicin are the drugs of choice [27,29]. We used the microagglutination test for diagnostic confirmation (1/320). Streptomycin or gentamicin are the drugs of choice [27,29]. We used the microagglutination test for diagnostic confirmation (1/320). Streptomycin or gentamicin are the drugs of choice [27,29]. We used the microagglutination test for diagnostic confirmation (1/320). Streptomycin or gentamicin are the drugs of choice [27,29]. We used the microagglutination test for diagnostic confirmation (1/320). Streptomycin or gentamicin are the drugs of choice [27,29]. We used the microagglutination test for diagnostic confirmation (1/320).

CONCLUSION

Facial nerve paralysis due to parotid gland abscess is exceedingly rare. The differential diagnosis in these unusual cases occurring in endemic areas should include F. tularensis infection. We presented a patient with tularemia who developed facial paralysis secondary to recurrent parotid abscess.

REFERENCES