Bilateral Facial Nerve Palsy and Appendicitis Occurring during Acute Retroviral Syndrome

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Abstract

We herein report the case of a 41-year-old homosexual man who presented to our hospital with typical acute retroviral syndrome. Complications of bilateral facial nerve palsy and appendicitis appeared eight days after admission. The bilateral facial nerve palsy spontaneously recovered one month later; however, the appendicitis required surgical intervention. To our knowledge, this is the first reported case of appendicitis related to acute retroviral syndrome.

Key words: acute retroviral syndrome, acute HIV infection, appendicitis, bilateral facial nerve palsy

Introduction

Primary HIV infection is known to cause acute retroviral syndrome (ARS) in 40% to 90% of recently infected individuals (1). The clinical features of ARS are nonspecific and variable. Fever, myalgia, headache, lymphadenitis and elevated transaminase levels are commonly observed in affected individuals. Aseptic meningitis, encephalopathy and unilateral facial nerve palsy have also been reported to develop in patients with ARS, although less frequently (1, 2). Bilateral facial nerve palsy (BFNP) is a very rare neurologic complication of ARS (2, 3). Appendicitis commonly occurs in HIV-infected patients (4); however, no reports of appendicitis associated with ARS have been published.

In this paper, we report a unique case of ARS complicated with both BFNP and appendicitis.

Case Report

A 41-year-old homosexual man was referred to our hospital with symptoms of fever, headache, sore throat and myalgia, all of which had persisted for approximately 10 days. He had no past medical history of note. His vital signs were as follows: blood pressure, 132/84 mmHg; pulse rate, 90 beats/min; respiratory rate, 16 breaths/min; temperature, 36.0 °C. A physical examination demonstrated positive jolt accentuation, multiple swollen lymph nodes and splenomegaly. The laboratory data showed elevated liver function parameters: total bilirubin (T-Bil)=0.4 mg/dL, asparate aminotransferase (AST)=141 IU/L, alanine aminotransferase (ALT)=164 IU/L, alkaline phosphatase (ALP)=829 IU/L. The results of a cerebrospinal fluid (CSF) analysis were as follows: a leukocyte count of 135/μL with 98% lymphocytes, a protein level of 83.6 mg/dL, a glucose level of 60 mg/dL (vs. 99 mg/dL glucose in the blood). A CT scan demonstrated multiple lymphadenopathy in the neck, underarms, mediastinum and intraperitoneal and inguinal regions. CT also showed splenomegaly; however, no evidence of hepatobiliary or intestinal abnormalities was observed. ARS was diagnosed based on a positive third-generation enzyme-linked immunosorbent assay (ELISA) HIV antibody test and negative Western blot analysis. The CD4 count and HIV-RNA level were 612/µL and 1.6×10⁶ copies/mL, respectively.

The patient’s headache and elevated liver function parameters both improved. Eight days after admission, he began to experience difficulties in closing his right eye and holding water in his mouth. Grade III right facial nerve palsy according to the House-Brackman Scale was diagnosed. Left facial nerve palsy appeared the next day, although no rash was observed. The patient developed bilat-
Intraoperatively, the appendix was found to have previously been divided, and the distal portion adhered stubbornly to the retroperitoneum. The size of the appendix was 40 mm ×15 mm. Pathological findings of infiltration of lymphocytes, proliferation of lymphoid follicles and fibrosis of the subserosal layer were observed (Fig. 3).

The patient’s postoperative course went well, and he was discharged five days after the surgery. The Western blot analysis was found to be positive. The BFNP spontaneously recovered and was no longer observed one month after the onset of symptoms (Fig. 1b, 4).

Discussion

BFNP is much less common than unilateral facial nerve palsy. One paper reported that only 0.3 to 2% of all facial nerve palsy cases were bilateral (5). The differential diagnosis of BFNP includes Bell’s palsy, Lyme disease, Guillain-Barré syndrome, trauma, sarcoidosis, neurosyphilis and HIV infection (3).

HIV-BFNP is classified into ARS- or AIDS-related BFNP types (2, 6). One cohort study reported an 0.08% prevalence of BFNP in ARS patients. ARS-BFNP is estimated to occur a median of 15 days after the onset of ARS, and all reported cases of ARS-BFNP have been complicated with aseptic meningitis. The etiology of ARS-BFNP remains unclear, although one group has proposed the involvement of either direct nerve lesions caused by HIV-1 or an immunologically-mediated inflammatory polyneuropathy (2). HSV and VZV are both known to cause peripheral neuropathy (7, 8); however, PCR assays for these viruses were negative in our pa-

![Figure 1.](image1.png)

Figure 1. a: The patient with bilateral facial palsy on the 12th hospital day. b: The patient’s bilateral facial nerve palsy fully recovered one month later.

![Figure 2.](image2.png)

Figure 2. A contrast-enhanced CT scan of the abdomen and pelvis showing the following: A: the orifice of the appendix, B: appendiceal wall enhancement and obstruction of the appendix, C: a swollen appendix with a diameter of 2 cm and slightly increased attenuation of mesentric fat.

![Figure 3.](image3.png)

Figure 3. Hematoxylin and Eosin staining. Lymphoid hyperplasia with numerous lymphocytes and subserous fibrosis. No obvious neutrophil infiltration is noted.
Our patient, similar to that observed in other previously reported cases, developed ARS-BFNP with aseptic meningitis (on the 18th day of ARS). To our knowledge, ARS-BFNP has not been previously reported to develop in cases of improvement of meningitis. The involvement of a direct nerve lesion was unlikely in our case because the meningitis itself improved. The etiology of ARS-BFNP is thought to involve an immunologically induced inflammatory polyneuropathy associated with HIV.

There have been no controlled studies of corticosteroids or antiretroviral therapy for ARS-BFNP. Most patients with ARS-BFNP recover fully without any treatment. Serrano et al. recommended the administration of a short course of corticosteroids, especially in cases of BFNP, because the risks of management are outweighed by the possible development of ophthalmologic and dental complications of neuropathy (2). Our patient received 30 mg of prednisolone for only one day, as we were forced to withdraw the drug due to the occurrence of appendicitis. Yet even without further prednisolone treatment, the BFNP fully recovered one month later. This outcome supports the findings of a prior report showing that ARS-BFNP is self-limiting, similar to other ARS symptoms (2). The effectiveness of corticosteroids and antiretroviral therapy (ART) for ARS-BFNP is unclear; therefore, the accumulation of further evidence is needed.

Viral infection is thought to be one cause of appendicitis. Lymphoid hyperplasia causes appendicitis due to swelling, ultimately obstructing the appendix (15). Systemic viral infections, such as adenovirus, VZV and measles, can cause lymphoid hyperplasia of the appendix (15, 16). Hyperplasia of lymphoid follicles was detected as a pathological finding in our patient. We know that the appendicitis developed after admission, as the abdominal CT scan obtained on admission depicted a normal appendix. Lymphoid hyperplasia due to ARS was the most likely cause of the appendicitis in our patient.

ARS is caused by an immune response to the HIV infection. Dendritic cells, monocytes, macrophages, natural killer cells and T-cells all produce cytokines that can cause ARS (17). The symptoms of ARS are highly individual; thus, the diagnosis of ARS is often missed in general practice. Clinicians should carefully watch the clinical course of ARS patients, even after their condition seems to improve, because ARS may cause unpredictable complications, including BFNP and appendicitis. ART is known to play an important role in treating symptoms and preventing HIV transmission (18, 19). In addition, ART may also prevent the
further development of complications of ARS, such as BFNP and appendicitis. The administration of ART should be considered in all patients with ARS symptoms, particularly prolonged or central nervous symptoms.

The authors state that they have no Conflict of Interest (COI).

References