Oculomotor Nerve Palsy as Initial Manifestation of Sinonasal Actinomyces: A Rare Case Report

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Abstract

Objectives: Actinomyces is a rare subacute to chronic infection caused by *Actinomyces*, a gram-positive, filamentous, non-acid, fast, anaerobic to microaerophilic bacteria. Herein, we present an extremely rare case of sinonasal actinomyces with an initial manifestation of oculomotor nerve palsy.

Methods: A 56-year-old man presented to our hospital with a 1-week history of drooping of right upper eyelid, double vision, and right frontal headache. Functional endoscopic sinus surgery was performed bilaterally under endotracheal general anesthesia. The time to surgical intervention following onset of oculomotor nerve palsy was 5 weeks. Antibiotic treatment (ciprofloxacin, 500 mg q12h × 8 weeks) was administered postoperatively for sinonasal actinomyces.

Results: The patient's right oculomotor nerve palsy and right frontal headache were markedly improved in the first week after surgery. The patient had completely recovered from both symptoms 1 month after surgery. Follow-up endoscopy performed at 6 months after surgery revealed no recurrence of sinonasal actinomyces.

Conclusion: This rare case of oculomotor nerve palsy with initial manifestation of sinonasal actinomyces was amenable to surgical intervention combined with antibiotic treatment and carries an excellent prognosis.

Keywords: Actinomyces; *Actinomyces odontolyticus*; Sinusitis; Oculomotor Nerve Palsy; Functional Endoscopic Sinus Surgery

Abbreviations

OD: Right Eye; OS: Left Eye; CT: Computed Tomography.

Introduction

Actinomyces is a rare subacute to chronic infection caused by *Actinomyces*, a gram-positive, filamentous, non-acid, fast, anaerobic to microaerophilic bacteria [1]. It is usually odontogenic. Although the most common clinical forms of actinomyces are cervicofacial, the nose and paranasal sinuses are rarely involved [2]. Subacute to chronic infection of sinonasal actinomyces manifests slowly to a progressive abscess and forms a draining sinus tract

[1]. Most patients with sinonasal actinomyces have nasal symptoms such as runny nose and nasal congestion. Herein, we present an extremely rare case of sinonasal actinomyces with initial manifestation of oculomotor nerve palsy.

Case Report

A 56-year-old man presented to our hospital with a one-week history of drooping of the right upper eyelid, double vision, and right frontal headache. He had type 2 diabetes mellitus. However, he denied any nasal symptoms, a history of facial injury, or dental procedures.

On ophthalmic examination, ptosis and constant exotropia were observed in the right eye (OD), but orthophoria was observed in the left eye (OS). The patient had diplopia caused by misalignment of the visual axes (Figure 1A). His visual acuity with correction was 0.8 (OD) and 1.0 (OS). Intraocular pressure was 18 mmHg (OD) and 19 mmHg (OS), which was within normal limits. Pupil size was symmetric at 3 mm (OD) and 3 mm (OS). Relative afferent pupillary defect was not observed in either eye. Conjunctiva (OD) was mildly injected. The cornea, anterior chamber, iris, and lens were normal in both eyes. Fundoscopy revealed that the optic discs and retinal vessels were normal, and no retinal detachment or break was observed. Optic coherence tomography was within normal limits. In accordance with the serial examinations, oculomotor nerve palsy (OD) was diagnosed.

Figure 1: Extraocular movements in nine cardinal positions of gaze. (A) Ptosis and constant exotropia was observed preoperatively in right eye, and orthophoria was observed in left eye. The patient had diplopia caused by misalignment of the visual axes. (B) The patient had completely recovered from right oculomotor nerve palsy 1 month after surgery.

Imaging studies were subsequently performed to assess the etiology of his oculomotor nerve palsy (OD). Magnetic resonance

imaging of the brain and orbit revealed normal intracranial and intraorbital structures. However, opacification in the bilateral ethmoid sinus and mucosal thickening of the bilateral maxillary sinus were incidentally observed (Figure 2). Computed tomography (CT) of the paranasal sinus revealed opacification in bilateral anterior ethmoid and posterior ethmoid sinuses, as well as mucosal thickening of bilateral maxillary sinus. However, the bilateral frontal and sphenoid sinuses were clear (Figure 3). Imaging studies indicated a diagnosis of sinusitis.

Figure 2: Magnetic resonance imaging of the brain and orbit revealed normal intracranial and intraorbital structures. Nevertheless, opacification in the bilateral ethmoid sinus and mucosal thickening of the bilateral maxillary sinus were incidentally observed.

Figure 3: Computed tomography of the paranasal sinus revealed opacification in bilateral anterior ethmoid (yellow asterisk) and posterior ethmoid sinuses (red asterisk) as well as mucosal thickening of bilateral maxillary sinus (whitish asterisk). However, bilateral frontal (square) and sphenoid (triangle) sinuses were clear.

Because his symptoms (ptosis, diplopia, and headache) were not resolved, the patient subsequently underwent a surgical intervention for sinusitis. Functional endoscopic sinus surgery was performed bilaterally under endotracheal general anesthesia. Intraoperatively, sinonasal polyps were noted (Figure 4A and 4B). The involved tissues were debrided and the sinuses were opened widely for restoration of sinus ventilation (Figure 4C). The sinuses were also irrigated profusely to remove all debris. The time to surgical intervention following onset of oculomotor nerve palsy (OD) was 5 weeks.

Figure 4: Intraoperative sinoscopic examination of right nasal cavity revealed (A) a polypoid tumor (red triangle) medial to right middle turbinate and (B) some polyps (yellow triangle) in right ostiomeatal complex. (C) The involved tissues were debrided and the sinuses were opened widely to restore sinus ventilation (white square: middle turbinate; black square: inferior turbinate).

A pus culture of the sinus revealed growth of *Actinomyces odontolyticus* and *Enterobacter aerogenes*. Antibiotic treatment (ciprofloxacin, 500 mg q12h × 8 weeks) was administered postoperatively for sinonasal actinomycosis.

The patient's postoperative course was uneventful. His oculomotor nerve palsy (OD) and right frontal headache markedly improved in the first week after surgery, and the patient had completely recovered from both symptoms at 1 month after surgery (Figure 1B). Follow-up endoscopy performed 6 months after surgery revealed no recurrence of sinonasal actinomycosis.

Discussion

Actinomycosis was first described by Bollinger in 1877 [3]. Approximately 70% of actinomycosis cases are caused by either *Actinomyces israelii* or *Actinomyces gerencseriae* [1]. In this present case, the infection was caused by a relatively uncommon pathogen, *A. odontolyticus*. *A. odontolyticus* is normally present in human flora and is common in the perimandibular soft tissues [4]. Facial injury or trauma (i.e., dental procedures) can cause the migration of this bacteria to other areas [5]. However, the patient had no history of facial injury or dental procedures.

Actinomycosis is usually a granulomatous and suppurative infection. The chronic form has slowly progressive abscesses and often forms a draining sinus tract that extrude characteristic but not pathognomonic "sulfur granules." Actinomycosis spreads contiguously and very rarely hematogenously [1, 4]. Infections of the oral and cervicofacial areas are most common in actinomycosis; however, any part of the body can be infected. Other areas that are often affected are the thoracic and abdominopelvic areas as well as the central nervous system [6]. Most sinonasal actinomycosis occurs in immunocompetent patients, and the lesions are unilateral [7]. Musculoskeletal, thyroid, and disseminated infections are also rarely observed [6,8]. One fatal case of intracranial and intraorbital actinomycosis of odontogenic origin was reported [9].

Diagnosis of actinomycosis can be difficult because the isolation of the organism requires a prolonged bacterial culture under anaerobic conditions [1]. The infection associated with actinomycosis is usually polymicrobial. Other organisms commonly associated with actinomycosis are *Aggregatibacter actinomycetemcomitans*, *Prevotella*, *Streptococcus*, *Enterobacteriaceae*, *Peptostreptococcus*, and *Staphylococcus* [1]. In this case, an associated organism, *Enterobacter aerogenes*, was isolated. A rare case of simultaneous actinomycosis with aspergillosis in the maxillary sinus was reported [10]. Furthermore, fatal *rhinocerebral mucormycosis* associated with actinomycosis could occur in an immunocompromised individual [11].

The initial diagnostic workup of actinomycosis, predicated upon imaging and tissue biopsies, is frequently nonspecific [2]. On CT scan, the sinuses of patients with sinonasal actinomycosis are opacified with focal calcification densities [7]. However, no focal calcification density was observed in the current patient's CT scan. A definitive diagnosis is usually rendered only after surgical resection and histologic examination of the cervicofacial mass [2]. Clinically, actinomycosis tends to mimic granulomatous diseases and cancers [2]. Rare cases of sinonasal actinomycosis mimicking rhinolith and mucocele have been reported [12,13].

Before the present report, oculomotor nerve palsy had not been recognized as a possible initial manifestation of sinonasal actinomycosis. The oculomotor nerve provides motor innervation for the majority of the extraocular muscles (levator palpebrae superioris, superior rectus, inferior rectus, medial rectus, and inferior oblique), and provides parasympathetic innervation for the sphincter pupillae and the ciliary muscles. In addition, sympathetic fibers run along the oculomotor nerve to innervate the superior tarsal muscle. Therefore, oculomotor nerve palsy leads to a "down and out" position of the eye (i.e., exotropia), diplopia (caused by misalignment of the visual axes), partial or complete ptosis, and dilated pupil. Except for dilated pupil, all of these symptoms were observed in the present case.

The most common etiologies of oculomotor nerve palsy are presumed microvascular constriction (42%), trauma (12%), tumor compression (11%), neurosurgery (10%), and aneurysm compression (6%) [14]. However, cavernous sinus disease, central nervous system infections, and subarachnoid hemorrhage are rare etiologies of oculomotor nerve palsy [15]. In this case, because the oculomotor nerve palsy was relieved after sinus surgery and antibiotic treatment, cavernous sinus infection hematogenously originating from actinomycotic sinusitis was considered to be the cause.

The treatment of actinomycosis may require long-term administration of antibiotics [2,6]. In one case, cervicofacial actinomycosis completely resolved after 4 months of oral penicillin treatment [3]. However, many cases may require complete surgical resection [2]. Sinonasal bacterial infections occur due to the inhibition of host defenses or due to reduced partial pressures of oxygen [1]. Therefore, if the surgical opening of paranasal sinuses involved in actinomycosis could be preserved, a relatively short-term antibiotic treatment of 1 month may be sufficient [10]. In this case, functional

endoscopic sinus surgery combined with antibiotic treatment (ciprofloxacin, 500 mg q12h × 8 weeks) carries an excellent prognosis. Surgical removal of the affected tissues and the restoration of sinus ventilation appear to be important factors for treating the disease [7]. Untreated infections can be life-threatening [2].

Conclusion

This case study demonstrates two pertinent points. First, we present an extremely rare case of oculomotor nerve palsy as an initial manifestation of sinonasal actinomycosis. Second, this condition is amenable to surgical intervention (functional endoscopic sinus surgery) combined with antibiotic treatment (ciprofloxacin, 500 mg q12h × 8 weeks) and carries an excellent prognosis.

Disclosure of Conflict of Interest

None.

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