

Case Report

Primary sinonasal tuberculosis confined to the unilateral maxillary sinus

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Abstract: Extrapulmonary tuberculosis is not rare and occurs mainly in the head and neck region. Cervical tuberculous lymphadenopathy is the most common form of extrapulmonary tuberculosis. Sinonasal tuberculosis is known to occur very rarely due to the protective functions of sinonasal mucosa. Although some signs of sinonasal tuberculosis may be present, such as associated facial abscesses, the symptoms and signs are usually nonspecific. Clinical suspicion is important for timely diagnosis and proper management of sinonasal tuberculosis due to its rarity and nonspecific clinical presentation. We report a case of tuberculosis confined to the unilateral maxillary sinus that was first misdiagnosed as recurrent rhinosinusitis after endoscopic sinus surgery.

Keywords: Tuberculosis, maxillary sinus, endoscopic sinus surgery

Introduction

The proportion of extrapulmonary tuberculosis is increasing along with the rise of human immunodeficiency virus infection [1]. Extrapulmonary tuberculosis can affect any region, including the pleura, the bones and joints, the central nervous system, the gastrointestinal system, the genitourinary system, and the head and neck region. Cervical tuberculous lymphadenopathy is the most common form in the head and neck region [1]. Sinonasal tuberculosis is extremely rare, with only eight cases of nasal tuberculosis reported from a single institution [2]. The symptoms of sinonasal tuberculosis may mimic other rhinosinusitis [3]. If the physician fails to consider the possibility of tuberculosis, the diagnosis of extrapulmonary tuberculosis may be difficult or delayed, resulting in an unfavorable outcome.

Here, we present a case of primary sinonasal tuberculosis, which clinically mimicked recurrent chronic rhinosinusitis after endoscopic sinus surgery, with a review of the literature.

Case report

A 31-year-old woman visited the outpatient clinic with a 12-month history of bilateral yellowish

rhinorrhea, which was more severe on her left side. She had several accompanying symptoms, including bilateral nasal obstruction, posterior nasal drip, hyposmia, and headache. Six months prior to visiting our department, she underwent left endoscopic sinus surgery at another hospital because her sinonasal symptoms had persisted, despite empirical antibiotic therapy for one month. These symptoms persisted even after the sinus surgery. Her family history was not significant.

Anterior rhinoscopy revealed a deviated nasal septum to the left, with purulent discharge in the left nasal cavity. An endoscopic examination showed purulent discharge from the left middle meatus, without a polypoid lesion. The middle meatal antrostomy was almost obstructed. A computed tomography (CT) scan revealed diffuse mucoperiosteal thickening of the left maxillary and ethmoid sinuses and hypertrophy of the left maxillary bone (**Figure 1**). The results of routine laboratory tests were unremarkable. An HIV test was negative. A chest X ray was normal.

Under the impression that a fungus ball or chronic rhinosinusitis had recurred after the previous surgery, left endoscopic sinus surgery, septoplasty, and bilateral submucosal inferior



Figure 1. Preoperative pre-enhanced axial paranasal CT scan. It shows a residual soft tissue lesion with mucoperiosteal thickening in the left maxillary and anterior ethmoid sinuses. Hypertrophy of the left maxillary bone was noted.

turbinoplasty were performed under general anesthesia. There was a purulent discharge from the maxillary and ethmoid sinuses. Part of the inflamed maxillary sinus mucosa was acquired and sent for histological study. The histological investigation revealed chronic granulomatous inflammation and necrosis, and the sample stained positive for acid fast-bacilli (**Figure 2**). Special stains for fungus were negative.

The patient started antituberculous medication (rifampicin, isoniazid, ethambutol, and pyrazinamide). After six months of antituberculous therapy, the patient's posterior nasal drip and rhinorrhea disappeared, and endoscopic examination revealed minimal mucosal thickening of the left maxillary sinus. A follow-up CT scan taken 10 months after the revision surgery showed minimal mucosal thickening of the left maxillary sinus (**Figure 3**). The patient remained symptom free one year after the completion of the antituberculous chemotherapy. We terminated the regular follow up thereafter.

Discussion

Sinonasal tuberculosis is rare disease entity. Moon et al. [4] examined the relative frequency of head and neck lesions in 220 patients with tuberculosis and reported that only two cases involved sinonasal cavities. The largest number

of cases of sinonasal tuberculosis reported by a single institution is eight, only two of which involved both the nasal cavity and the sinus [2]. Primary sinonasal tuberculosis is rare. Its rarity is probably due to the self-protective functions of the nose, such as ciliary movement, bactericidal secretion, and mechanical filtering by vibrissae [5]. Usually, sinonasal tuberculosis occurs secondary to pulmonary tuberculosis by inhalation of infected particles. It is more frequent in females, and common symptoms are nasal obstruction, nasal discharge, crusting, and epistaxis. It commonly involves the nasal septum and the inferior turbinate [5, 6]. Sometimes, septal perforation, cleft of the nasal ala, or facial abscesses may be present [2, 7]. Tuberculosis of the paranasal sinuses can result from the bloodstream or by direct extension. Paranasal sinus tuberculosis without the involvement of the nasal cavity as in the present case is extremely rare.

The diagnosis of primary sinonasal tuberculosis is difficult. As acid-fast bacilli are difficult to detect in surgical specimens, Beltran et al. [8] proposed that the diagnosis of sinonasal tuberculosis should be based on the following criteria: the absence of a clinical response to empirical antibiotics, the presence of caseous granulomatous inflammatory lesions on histopathology, and identification of *Mycobacterium tuberculosis* in the surgical specimen. Nasal secretions and a nasal swab are not used for the diagnosis due to a low yield. Histologically, spherical granulomas with central caseous necrosis imply tuberculosis. Microscopic-positive Ziehl-Neelsen stain is suggestive of tuberculosis, but it does not always indicate confirmation of the diagnosis. Molecular tests allow identification of tuberculosis within one day, with high specificity. However, the high cost is considered a limitation [9]. Often, neither histological nor bacteriological confirmation is possible, and the diagnosis of primary sinonasal tuberculosis is made on the basis of the patient's response to the antituberculous medication [8]. CT or magnetic resonance imaging can be helpful to figure out the extent and pattern of diseases. Although calcification within sinuses on CT scans can be indicative of sinonasal tuberculosis, imaging findings are mostly nonspecific [4]. Several cases of delayed diagnosis and even inappropriate treatment are found in the literature [2]. Extrapulmonary tuberculosis often appears in immunocompro-

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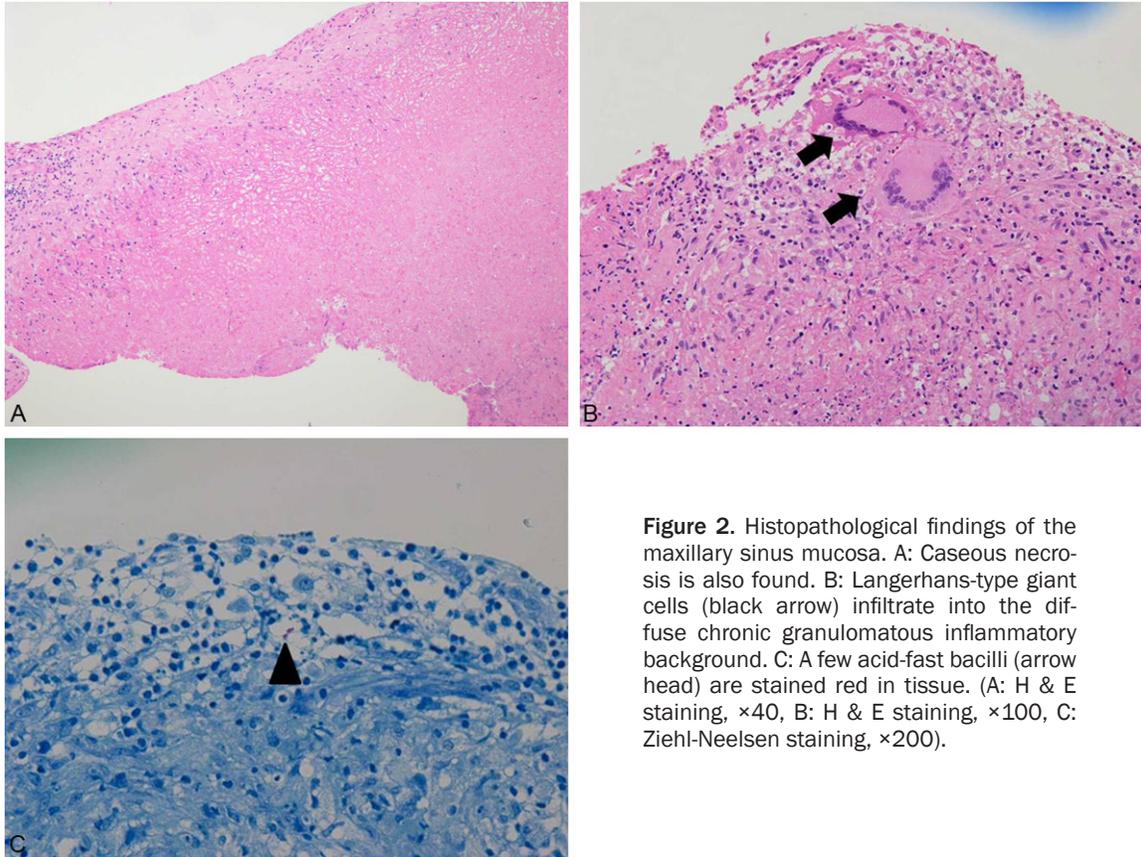


Figure 2. Histopathological findings of the maxillary sinus mucosa. A: Caseous necrosis is also found. B: Langerhans-type giant cells (black arrow) infiltrate into the diffuse chronic granulomatous inflammatory background. C: A few acid-fast bacilli (arrow head) are stained red in tissue. (A: H & E staining, $\times 40$, B: H & E staining, $\times 100$, C: Ziehl-Neelsen staining, $\times 200$).



Figure 3. Follow up pre-enhanced axial paranasal CT scan. It shows minimal mucoperiosteal thickening in the left maxillary sinus.

mised patients. However, Kim et al. [2] reported that most patients were healthy before having nasal tuberculosis. Taken together, the suspicion of sinonasal tuberculosis is a key point in

the diagnosis, regardless of the immune status of the patient.

Tuberculosis involving the nasal cavity can be treated only with antituberculous medication. For tuberculosis involving both the nasal cavity and the paranasal sinus, antituberculous chemotherapy is the mainstay, and sinus surgery can be done additionally. For cases involving only the paranasal sinus, such as the present case, sinus surgery is helpful for sinus drainage and specimen collection. The American Thoracic Society, Centers for Disease Control and Prevention, and Infectious Diseases Society of America suggests that the basic principles of treatment of pulmonary tuberculosis are also suitable for extrapulmonary tuberculosis. Therefore, for patients with extrapulmonary tuberculosis, a six- to nine-month regimen (two months of isoniazid, rifampicin, pyrazinamide, and ethambutol followed by four to seven months of isoniazid and rifampicin) is recommended as initial therapy [10].

We present this case as an additional report of rare primary sinonasal tuberculosis in an immu-

nocompetent patient. The patient suffered persistent symptoms, including rhinorrhea, nasal obstruction, and posterior nasal drip, six months after undergoing previous endoscopic sinus surgery. Empirical antibiotic therapy for two weeks did not relieve any of the symptoms. The patient's CT scans showed mucosal thickening of the unilateral maxillary sinus, with bony hypertrophy. In this clinical setting, sinonasal tuberculosis should be considered in the differential diagnosis.

In conclusion, sinonasal tuberculosis can be primary or secondary, irrespective of the immune status of the host. Clinical suspicion is important when a patient presents with unusual clinical features. Antituberculous medication and/or surgical debridement is the mainstay of the treatment.

Disclosure of conflict of interest

None.

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References

- [1] Al-Serhani AM. Mycobacterial infection of the head and neck: presentation and diagnosis. *Laryngoscope* 2001; 111: 2012-2016.
- [2] Kim YM, Kim AY, Park YH, Kim DH, Rha KS. Eight cases of nasal tuberculosis. *Otolaryngol Head Neck Surg* 2007; 137: 500-504.
- [3] Michael RC, Michael JS. Tuberculosis in otorhinolaryngology: clinical presentation and diagnostic challenges. *Int J Otolaryngol* 2011; 2011: 686894.
- [4] Moon WK, Han MH, Chang KH, Im JG, Kim HJ, Sung KJ, Lee HK. CT and MR imaging of head and neck tuberculosis. *Radiographics* 1997; 17: 391-402.
- [5] Goguen LA, Karmody CS. Nasal tuberculosis. *Otolaryngol Head Neck Surg* 1995; 113: 131-135.
- [6] Butt AA. Nasal tuberculosis in the 20th century. *Am J Med Sci* 1997; 313: 332-335.
- [7] Jha D, Deka RC, Sharma MC. Tuberculosis of the maxillary sinus manifesting as a facial abscess. *Ear Nose Throat J* 2002; 81: 102-104.
- [8] Beltran S, Douadi Y, Lescure FX, Hanau M, Laurans G, Ducroix JP. A case of tuberculous sinusitis without concomitant pulmonary disease. *Eur J Clin Microbiol Infect Dis* 2003; 22: 49-50.
- [9] Masterson L, Srouji I, Kent R, Bath AP. Nasal tuberculosis—an update of current clinical and laboratory investigation. *J Laryngol Otol* 2011; 125: 210-213.
- [10] Blumberg HM, Burman WJ, Chaisson RE, Daley CL, Etkind SC, Friedman LN, Fujiwara P, Grzemska M, Hopewell PC, Iseman MD, Jasmer RM, Koppaka V, Menzies RI, O'Brien RJ, Reves RR, Reichman LB, Simone PM, Starke JR, Vernon AA; American Thoracic Society, Centers for Disease Control and Prevention and the Infectious Diseases Society. American Thoracic Society/ Centers for Disease Control and Prevention/ Infectious Diseases Society of America: treatment of tuberculosis. *Am J Respir Crit Care Med* 2003; 167: 603-662.