CASE REPORT

Sinusitis of actinomycosis infection : a case report

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Abstract : Actinomycosis is an infection caused by anaerobic bacteria, primarily from the genus *Actinomyces*, which normally colonize the several regions including the mouth. Disruption of mucosa may lead to infection of virtually any site, but reports of involvement of the sinuses are rare. We report a case of an actinomycosis infection in the unilateral maxillary sinus. A 47-year-old female visited our hospital with a complaint of mild pain of left buccal region. Computed tomography (CT) revealed that the left maxillary and anterior ethmoidal sinus cavities were opacified along with a calcified fragment located close to the natural ostium. Thus, we provisionally diagnosed as a fungal sinusitis. She underwent trans-nasal endoscopic sinus surgery. The sinuses were opened and the caseous material was removed. The histopathological examination suggested an actinomycosis, but not fungal, infection. The patient's postoperative course was uneventful. No evidence of recurrence has been seen over the 30 months of the postoperative follow-up period. In case of the aggressive actinomycotic sinusitis, extension into the adjacent organs could be occurred. We should be aware that sinusitis of actinomycosis infection could progress in patients with risk factors such as diabetes and immunodeficiency. J. Med. Invest. 68 : 202-204, February, 2021

Keywords **:** *actinomyces, actinomycotic sinusitis, computed tomography, fungal sinusitis, histopathological examination*

INTRODUCTION

Actinomycosis is a progressive infection caused by anaerobic bacteria, primarily from the genus *Actinomyces*, which normally colonize the mouth, bronchial tubes, colon, and vagina. Disruption of mucosa may lead to infection of virtually any site. Actinomycosis occurs in the head and neck region in about half of cases, but involvement of the paranasal sinuses is extremely rare. We present a typical, demonstrative case of an actinomycosis infection in the unilateral maxillary sinus.

CASE PRESENTATION

A 47-year-old Japanese woman had visited a local dental clinic followed by a local ENT clinic, complaining of a mild chronic pain of her left buccal region. She had been not pointed out abnormality on her ipsilateral upper molar in the dental clinic. Then, she was diagnosed as having chronic rhinosinusitis by a previous ENT doctor. The pain temporarily had subsided after antibiotic treatment, but it recurred and persisted. She was referred to our department for further advice. On physical examination, a tender lesion with slight swell was seen on her left buccal region. She was not a diabetes patient, an immunocompromised patient, nor smoker.

Computed tomography (CT) revealed that the left maxillary and partial ethmoidal sinus cavities were opacified along with a calcified fragment located close to the natural ostium, but without any dental structures penetrating through an alveolar bone into the maxillary sinus (Fig. 1A). Hypertrophic maxillary bone was also observed. The results of a blood examination were unremarkable. Based on the comprehensive consideration, we made a diagnosis as a left fungal sinusitis.

The patient underwent trans-nasal endoscopic sinus surgery under general anesthesia. The maxillary natural ostium was obstructed by hemorrhagic granulation tissue (Fig. 1B). As seen in the preoperative CT imaging, the caseous material was found near the natural ostium (Fig. 2Aa), thereby the fragment was gathered as sample of the pathological examination. Sinus mucosa surrounding the cavity was preserved after open the sinuses (Fig. 2Ab). In histopathological findings, multiple colony-forming drusen surrounded by inflammatory fibroid tissue can be observed in Hematoxylin and Eosin staining. Further, the sample tissue consisted of bacterial ooze tissue containing gram-positive *Actinomyces* (Fig. 2B). No evidence of fungal infection was indicated by either PAS- or Grocott-immunohistochemistry (Fig. 2B).

The patient's postoperative course was uneventful. She was discharged 3 days after surgery, and took clarithromycin for 6 months (the low-dose clarithromycin for 5 months following the normal-dose clarithromycin for 1 month). No evidence of recurrence was seen over the 30 months of postoperative follow-up period (Fig. 3A and B).

DISCUSSION

The cases of actinomycotic sinusitis are extremely rare (1-9). It's not easy to definitively diagnose as the actinomycotic sinusitis before obtaining the histopathological results since the actinomycotic sinusitis has similar findings with fungal sinusitis on CT (e.g., the shadow in unilateral sinus with a calcified density, hypertrophic maxillary bone). *Actinomyces* often form bacterial mass with large number of hypha, like a fungus, and make cluster calcification which is composed by $Ca_2H_2(PO_4)_2$ and $CaSO_4$ in that of central necrotic resion. This calcification material is carried to the maxillary sinus natural ostium by

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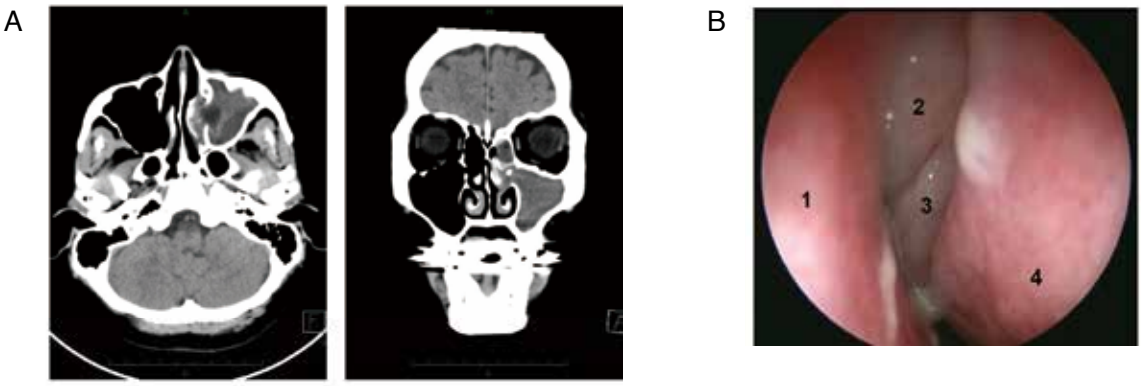

Figure 1.
A) Preoperative findings of CT : Axial (left panel) and coronal (right panel) CT reveal the left maxillary and anterior ethmoidal sinus cavities were opacified along with a calcified fragment located close to the natural ostium. Hypertrophic maxillary bone could be also observed.
B) Preoperative finding of endoscope (left nasal) : 1 ; septum, 2 ; middle nasal turbinate, 3 ; inflamed granulation tissue arising from maxillary sinus, 4 ; inferior nasal turbinate.

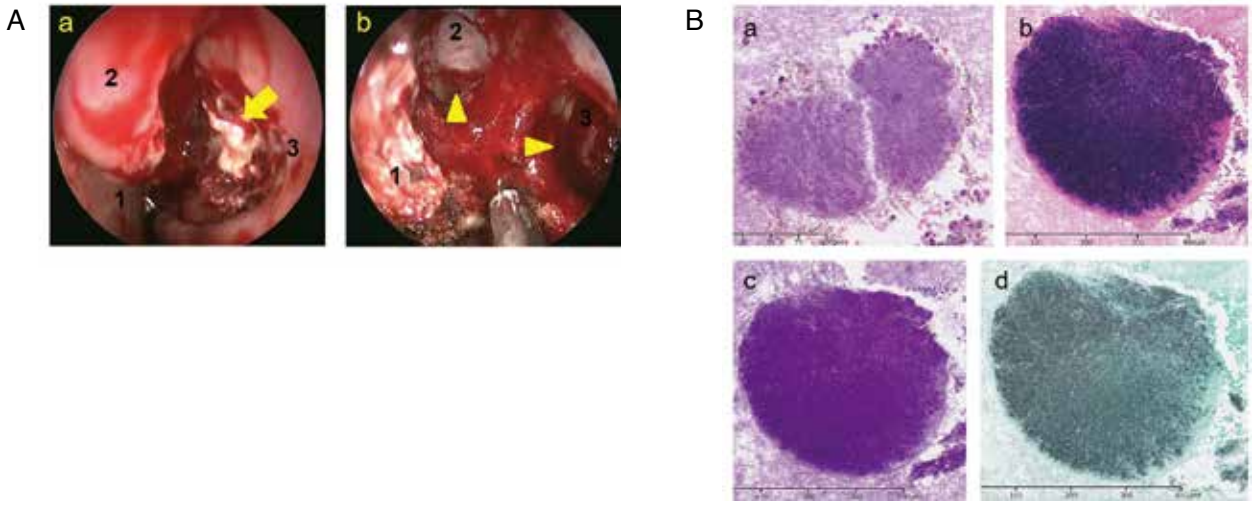

Figure 2.
A) Intraoperative finding : a) Caseous material (arrow) and hemorrhagic granulation mucosa are seen around the natural ostium. 1 ; septum, 2 ; middle nasal turbinate, 3 ; opened maxillary natural ostium and caseous material b) Sinus mucosa surrounding the cavity was preserved (arrow heads). 1 ; middle nasal turbinate, 2 ; opened anterior ethmoidal sinus, 3 ; opened maxillary sinus.
B) Histopathological findings : a) Hematoxylin and Eosin staining ; multiple colony-forming drusen surrounded by inflammatory fibroid tissue b) Gram staining ; bacterial ooze tissue containing granules of gram-positive actinomyces c) PAS staining & d) Grocott staining ; no evidence of fungal infection in these immunohistochemistry.

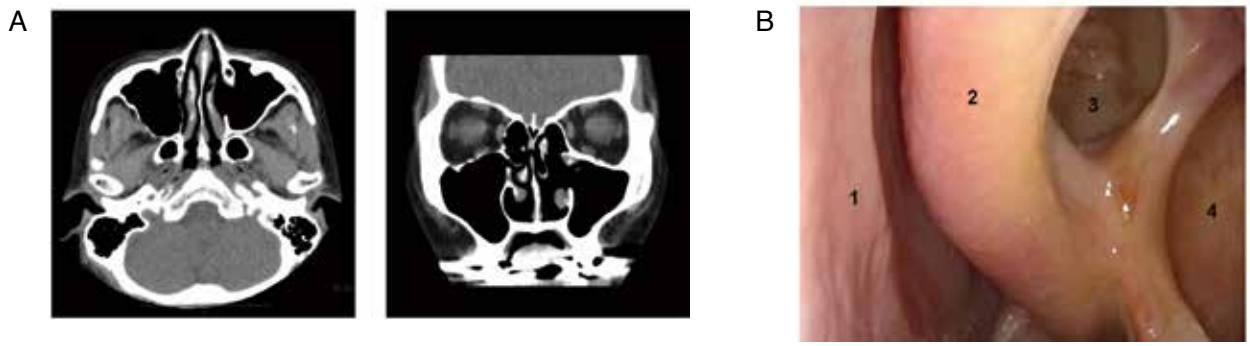

Figure 3.
A) CT findings in 30 months after the surgery : Axial (left panel) and coronal (right panel) CT reveal the left maxillary and anterior ethmoidal sinus cavities get clear.
B) Left-side intranasal finding in 12 months after the surgery: Granulation mucosa disappeared. No evidence of recurrence was seen. 1 ; septum, 2 ; middle nasal turbinate, 3 ; well opened ethmoid sinus, 4 ; well opened maxillary sinus.

ciliary transport mechanism. Finally, the calcification material stagnates near the maxillary sinus natural ostium because of the natural ostium obstruction in most of these cases (10). In addition, a bacterial culture inspection often misses to detect the *Actinomyces* because of their anaerobiosis. It is of interest that actinomycosis is sometimes accompanied with fungal infection in the maxillary sinus (5, 6). In light of these reports, the actinomycotic- and fungal sinusitis might actually coexist more often. The therapeutic methods, especially the preference of the antibiotics after surgery, are different between the two pathogens. Therefore, actinomycotic sinusitis should be diagnosed by minute histopathological examinations, not only Hematoxylin and Eosin (H&E)- and Gram staining but also Periodic acid-Schiff (PAS)- and Grocott staining for ruling out the coexistence of fungal sinusitis. To our knowledge, there are very few previous case reports showing all of the important histopathological findings for making a diagnosis as the actinomycotic sinusitis without coexistence of the fungal infection. Thus, this report is valuable to show typical, demonstrative clinical characteristics of actinomycotic sinusitis.

Surgical removal of the involved tissues and the restoration of sinus ventilation are important factors for treating both of these diseases. In general, long-term antibiotics (e.g., penicillin) therapy following endoscopic sinus surgery is considered to be the gold standard for treatment of the actinomycotic sinusitis (1-9). In the present case, lesion area was limited within the sinus. Her actinomycotic sinusitis were thought to be sufficiently controlled by endoscopic sinus surgery. In addition, it was easy to observe the local finding by using an endoscope. Therefore, we chose the 6 months clarithromycin treatment with expecting that of immunological effects (11-13). Together, we kept observation her sinus condition carefully in order to start the penicillin treatment immediately if sinus condition had gotten worse.

In the case of the aggressive actinomycotic sinusitis, extension into the orbit, cavernous sinus, or intracranial compartments could occur (7-9). However, it is not easy to diagnose as actinomyces infection because the lesion progresses like a carcinoma which makes an extension with bone destruction into the adjacent organs (7-9). In addition, these cases are thought to be extremely rare. Therefore, treatment plan and follow-up period depend on an individual case, whereas surgical removal of the involved tissues and the restoration of sinus ventilation followed by long-term antibiotics therapy is considered to get a broad consensus. Taking these into account, it is the most important that we should be aware of the necessity of several repeat histopathological and bacterial culture examinations with suspecting the possibility of the actinomyces infection in sinus, if the biopsy sample does not reveal a definitive histopathological diagnosis. On the other hand, in case of non-invasive type of actinomycotic sinusitis, we can just keep the patients under observation. However, we should be aware that sinusitis of actinomycosis infection could change to aggressive progress in patients with risk factors such as diabetes and immunodeficiency (7-9).

CONSENT

Informed consent was obtained from the patient for publication of a case report and accompanying images.

CONFLICT OF INTEREST

No competing interests.

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