Maxillary sinus aspergilloma of odontogenic origin: Report of 2 cases with cone-beam computed tomographic findings and review of the literature

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ABSTRACT

Aspergilloma of the maxillary sinus is considered rare in immunocompetent patients, but a considerable increase has recently been seen in the incidence of reported cases. Dental procedures involving the antral region are thought to predispose individuals to this form of aspergillosis. Because aspergilloma shares similar clinical features with other sinus pathologies, its diagnosis may be delayed. Thus, an early diagnosis confirmed by a histopathological examination plays a crucial role in the adequate management of aspergilloma. This article provides a concise review of the reported cases of aspergilloma associated with dental procedures and reports 2 new cases of aspergilloma in middle-aged female patients, with a presentation of their cone-beam computed tomographic findings. (Imaging Sci Dent 2018; 48: 139-45)

KEY WORDS: Fungi; Maxillary Sinus; Aspergillus; Cone-Beam Computed Tomography

Case Reports

Case 1

A 54-year-old woman was referred to our clinic because of a radiopacity in the left maxillary sinus that was noticed on a routine dental examination by a general practitioner. She had undergone endoscopic sinus surgery due to this...
radiopacity 3 years ago. The patient’s general health was unremarkable. She had a history of a nasal obstruction that occurred after root canal treatment of the left maxillary first molar tooth several years previously. Upon clinical examination, no pathology was observed. Panoramic radiography showed a small radiopacity that resembled a foreign body in the maxillary antrum (Fig. 1). Cone-beam computed tomography (CBCT) revealed an iron-like opacity in the central area of the left maxillary sinus (Fig. 2). No evidence of bone destruction was seen on the sinus walls. Under local anesthesia and sedation, extraction of the left maxillary first molar and the Caldwell-Luc procedure were planned. During the operation, a thick gray-brown, paste-like material that resembled root-canal sealer was observed in the sinus. The palatal root of the left maxillary first molar seemed to have perforated the floor of the sinus. Complete curettage and irrigation were performed. An antibiotic was prescribed after the operation to prevent bacterial superinfection. The specimen was sent for a histological examination, which revealed matted fungal hyphae that were evident on hematoxylin and eosin staining. The acute-branched septate hyphae were similar to *Aspergillus* and showed no tissue invasion. The histopathologic diagnosis was aspergillosis (Fig. 3).

The patient had an uneventful postoperative recovery. One year after surgery, she had no clinical and radiographic evidence of disease (Fig. 4).
Case 2

A 41-year-old woman was referred to our clinic with a complaint of occasional left-sided pain in her upper face. Her medical history included Raynaud syndrome and asthma. Upon clinical examination, no pathology was observed. A panoramic radiographic examination showed a radiopaque area at the level of the left maxillary sinus (Fig. 5). Her left first molar tooth had been extracted 13 years ago and the left first premolar and second molar teeth had undergone endodontic treatment. CBCT showed a piece of the root of the left first molar tooth in the sinus cavity and mucosal thickening in the sinus walls (Fig. 6). A Caldwell-Luc procedure was performed on the left maxillary sinus under local anesthesia and sedation. A
full-thickness mucoperiosteal flap was raised. A bony window was made using a trephine bur and the root of the left first molar tooth with granulation tissue was removed. The sinus mucosa seemed healthy. The bony window was replaced and sutured with 4-0 Vicryl stitches to the bone. Closure of the flap was performed with 3-0 silk stitches. An antibiotic and analgesic were prescribed after the operation. Curetted granulation tissue around the piece of the root was submitted for a histological examination, the results of which were compatible with aspergilloma (Fig. 7). The patient was followed periodically for 1 year. At 1-year check-up, she was completely asymptomatic (Fig. 8).

Discussion

Fungal infection of the maxillary sinus is relatively rare in healthy individuals, but because of the globally uncontrolled consumption of chemotherapeutics that cause patients to be vulnerable to fungal infections and as a result of improvements in diagnostic imaging techniques, the detection of this infection among healthy subjects seems to be increasing.

It has been reported that more than 10% of patients who had chronic sinusitis were found to have aspergilloma, predominantly in the maxillary sinus. The main aspects of the pathophysiology of aspergilloma of the maxillary sinus are still debated, although contamination of the maxillary sinus with Aspergillus has been suggested to occur through various pathways. Because Aspergillus species do not have the ability to penetrate intact mucus membranes, Aspergillus is usually considered to cause infections in maxillary sinus as a result of the inhalation of the airborne spores of Aspergillus that are ubiquitous in the environment. According to the aerogenic theory, the accumulation of fungal spores in the maxillary sinus may become pathogenic under relatively anaerobic conditions. However, unlike the other paranasal sinuses, Aspergillus spores may also be transmitted to the maxillary sinus through an iatrogenic pathway associated with dental procedures.

Recently, an increasing number of researchers have suggested that because of the close relationship between the antral teeth and sinus floor, dentogenic factors increase the risk of aspergilloma of the maxillary sinus. Tomazic et al. suggested that if a dentogenic factor is present, the risk of developing an aspergilloma is 2.7-fold higher than in unaffected sinuses. Dental procedures are thought to be able to cause massive fungal inoculation of the maxillary antrum as a result of perforation of the sinus membrane. Additionally, dental materials that contain heavy metals (e.g., zinc), such as root canal sealer, gutta-percha, silver cones, and amalgam, may penetrate into the sinus during dental procedures and provide favorable conditions for the growth of Aspergillus species. Although eugenol in dental mate-

Fig. 7. Histopathologic finding shows the characteristic septate hyphae (Grocott-Gomori methenamine silver stain, × 400).

Fig. 8. Postoperative panoramic radiograph of the sinus at 1-year follow-up.
Table 1. Published cases of aspergilloma of odontogenic origin in the literature

| Author Year | Age-Sex | Systemic status | Site | Symptoms          | Etiology           | FBS | DOS | Imaging modality | Surgical approach | Antifungal therapy | Follow-up |
|-------------|---------|-----------------|------|-------------------|--------------------|-----|-----|------------------|-------------------|-------------------|-----------|
| Axelsson et al. 1978 | 36-F | NC | L | P, NO, R | Extraction-NA | – | 6Y | NA | CL | – | 3Y |
| Kawana et al. 1987 | 47-M | NC | L | S, P, F, ND | Extraction-26 | Root | 22Y | W | CL | – | NA |
| De Foer et al. 1990 | 31-F | NA | L | İmp. surgery-25 | İmplant | 6M | NA | CL | – | 9Y |
| De Foer et al. 1990 | 31-F | NA | R | P | Extraction-16 | – | 3M | NA | CL | – | 6Y |
| De Foer et al. 1990 | 30-M | B | L | – | RCT-26 | Sealer | 10M | NA | CL | – | 4Y |
| De Foer et al. 1990 | 35-F | NA | L | S, P | RCT-25,27 | Sealer | NA | NA | CL | – | 1Y |
| De Foer et al. 1990 | 35-F | NA | R | – | RCT-26 | Sealer | 12M | NA | CL | – | 7Y |
| De Foer et al. 1990 | 56-M | NC | R | NO, R | Extraction-13 | Root | 1M | NA | CL | – | 5M |
| Kobayashi 1995 | 22-F | NA | R | – | RCT-15,16 | Gatta | 6M | OM, PAN | CL | – | 8Y |
| Falworth et al. 1996 | 41-F | NA | L | P | RCT-24 | Sealer | 6M | OM, PAN | CL | – | 1Y |
| Ogata et al. 1997 | 58-M | NA | L | PD | Extraction-NA | Antrolith | NA | CT | CL | – | 6M |
| Khongkhunthian et al. 2001 | 25-F | NC | R | – | RCT-16 | Sealer | NA | PAN, W | A | – | NA |
| Khongkhunthian et al. 2001 | 25-F | NA | L | P, S | RCT-14 | Sealer | 2Y | PAN, W | A | – | NA |
| Horre et al. 2002 | 28-F | NC | R | P | RCT-16 | ZOE | 10Y | PAN, W | NA | Antifungal | NA |
| Martin et al. 2004 | 30-F | NC | L | S, NO, PD | RCT-27 | Sealer | NA | W | CL | Itraconazole | 12M |
| Matjaz et al. 2004 | 22-NA | NA | L | S, P | RCT-26 | Sealer | 1Y | PAN | ESS | – | 4M |
| Giardino et al. 2005 | 60-M | NC | R | NA | RCT-15 | Sealer | 2Y | PAN, CT | CL | – | 1Y |
| Burnham et al. 2009 | 46-M | NA | R | P, NO, ND | Extraction-17 | Amalgam | 2Y | CT, OM | FESS | – | 6M |
| Sano et al. 2009 | 48-M | NC | R | – | Bone grafting | Graft | 6M | CT, PAN | CL | – | 15M |
| Bosi et al. 2010 | 78-F | NA | L | P | RCT-26 | Sealer | 6M | CT | ESS | – | NA |
| Sato et al. 2010 | 50-M | NC | L | S | İmp surgery-Zygomatic | İmplant | 12M | CT | SE | – | 12M |
| Fanucci et al. 2013 | 54-F | NA | L | Sin, P, R | RCT-26 | Sealer | 2Y | PAN, CT | CL | – | NA |
| Guivarc'h et al. 2015 | 64-NA | PR, D | R | Sin | RCT-16 | Sealer | NA | CT | CL | – | 6M |
| Urs et al. 2015 | 35-F | NC | L | P, S | Extraction-25 | Root | 1M | CECT | NA | Itraconazole | 6M |
| Vinciguerra et al. 2016 | 34-M | NC | B | – | RCT-15,16,25,26 | Sealer | – | PAN, CT | ESS | – | 12M |
| Harada et al. 2017 | 59-F | NC | L | NO, ND, P, S | İmp. surgery-NA | İmplant | 4Y | PAN, CT | CL | – | 12M |
| Cansiz et al. 2017 | 32-F | NC | L | NO, ND | SARPE | – | 2W | CT | FESS | – | 3M |
| Present case 1 2017 | 54-F | NC | L | NO | RCT-26 | – | 6Y | PAN, CBCT | CL | – | 12M |
| Present case 2 2017 | 41-F | A, RF | L | P | Extraction-26 | Root | 13Y | PAN, CBCT | CL | – | 12M |

NA: not available M: male F: female NC: non-contributory, B: Behçet, D: diabetes, PR: psoriatic rheumatism, A: asthma, RF: Raynaud phenomena, R: right, L: left, S: swelling, P: pain, F: fewer, ND: nasal discharge, NO: nasal obstruction, PD: purulent discharge, R: rhinorrhea, NB: numbness, Sin: sinusitis, RCT: root canal treatment, FBS: foreign body in maxillary sinus, M: month, Y: year, DOS: duration of symptoms occur, CL: Caldwell-Luc, LR: lateral rhinoscopy, A: antroscopy, ESS: endoscopic sinus surgery, FESS: functional endoscopic sinus surgery, SE: sinusectomy, SARPE: surgically assisted rapid palatal expansion
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Aspergillus cells, and in some cases may result from accumulation of the heavy metals that were pushed into the sinus with the dental materials. A more precise examination with computed tomography (CT) may be necessary to exclude other sinus diseases, such as antrolith, osteoma, mucocele, B cell lymphoma, squamous cell carcinoma, adenoid cystic carcinoma, and inflammatory myofibroblastic tumors, from the differential diagnosis.

The extent of the lesion, bone involvement, and erosion can also be evaluated using CBCT, which requires a lower radiation dose, is cost-effective, and is not time-consuming. Magnetic resonance imaging (MRI) can also be helpful, as decreased signal intensity on T2-weighted MRI has been described as characteristic of aspergilloma, and MRI can also help clinicians to differentiate aspergilloma from inflammatory or neoplastic changes.

The treatment of aspergilloma primarily consists of surgical removal of the lesion. Both the Caldwell-Luc and endoscopic techniques can be used. In most of the reported cases, the Caldwell-Luc procedure was used successfully for the management of aspergilloma. Systemic antifungal therapy is not generally required. However, if symptoms persist for a long time after surgery, an oral antifungal drug may be required as an additional therapy. Nonetheless, clinicians should be careful about using these drugs because of severe adverse effects, such as nephrotoxicity. Since bacterial superinfection can cause acute sinusitis attacks, an appropriate antibiotic therapy is recommended in order to avoid bacterial coinfections.

Different types of dental procedures that involve the maxillary sinus may facilitate the occurrence of fungal sinusitis, which shares similar features with other infections of the sinus. Clinicians should be aware of the possibility of fungal etiology, especially in cases resistant to treatment, and should follow the patient periodically if sinus perforation occurs during a procedure to minimize toxicity, costs, and other complications because of an inappropriate treatment strategy. Although the management of aspergilloma is much simpler than the management of the invasive form of aspergillosis, delays can occur in management because the likelihood of fungal origin may be underestimated.

Thus, diagnostic tools, especially imaging modalities, play a crucial role in detecting aspergilloma, which is usually an incidental finding. CBCT can provide useful information to clinicians about the location and the extent of the lesion.

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