

Surgical removal of a fungus ball formed in the maxillary sinus secondary to surgically assisted rapid palatal expansion procedure during Le Fort I osteotomy

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Abstract

Surgically assisted rapid palatal expansion (SARPE) is a surgery with a low rate of complications frequently performed in cases of maxillary transverse deficiency. Although fungal infection of the paranasal sinuses is rare in healthy people, this rate rises after the use of antibiotics and steroids, and after invasive interventions of the sinus. In our study, we present surgical removal of aspergillosis formed in the case of an 18-year-old female patient secondary SARPE procedure, with Le Fort I osteotomy. A cone beam computed tomography examination 6 months after the surgical removal of aspergillosis showed no recurrence. In conclusion, we report that aspergillosis may occur secondary to SARPE procedure and can successfully be removed in a single session with Le Fort I osteotomy.

Keywords: Aspergillus; maxillary sinus; osteotomy

INTRODUCTION

Although aspergillosis is rare in non-immunosuppressed people, it is the most common fungal infection of the sinuses and is seen unilaterally in the paranasal sinuses (1-3). The most commonly accepted classification of fungal rhinosinusitis are invasive and non-invasive forms (4). The non-invasive structure commonly caused by *Aspergillus fumigatus*, which is most frequently seen in the maxillary sinus, is called a fungus ball (4). Conditions may cause fungal infections include diabetes, long-term antibiotic and cortisone treatments, radiotherapy, chemotherapy, immunosuppressive therapies, and diseases causing immunodeficiency. With the introduction of tomography into the clinic routine, the diagnosis and treatment of many sinus disorders that have been difficult to diagnose previously has been made possible.

Surgically assisted rapid palatal expansion (SARPE) is a good alternative in treatment of maxillary transverse deficiency in adults. However, in cases where extrusion, intrusion, advancement and setback corrections are planned on the maxilla the viable choice in three-

dimensional space is the Le Fort I osteotomy and rigid fixation.

There is only one case in the literature showing the occurrence of aspergillosis in the maxillary sinus after SARPE procedure. To the best of our knowledge there has been no report regarding the treatment of aspergillosis infection of the sinus with excision during Le Fort I osteotomy. This case report is a presentation of the successful treatment of a fungus ball with excision during Le Fort I osteotomy.

CASE REPORT

An 18-year-old female patient was referred to our clinic for examination of bimaxillary orthognathic surgery. Following clinical and radiographic evaluations a radiopaque mass diagnosed on the right sinus (Figure 1 and Figure 2). The patient has a history of SARPE procedure 17 months before referral. The evaluation of past radiographic data before SARPE procedure revealed that the sinuses were clean (Figure 3). For this reason we inclined to think that the radiopaque mass occurred secondary to SARPE procedure. The pre-diagnosis was

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fungus ball. In patient's anamnesis there was no history of past debilitating diseases such as diabetes, prolonged antibiotic use, cortisone treatment or past fungal infection. We decided to perform excision of fungus ball during bimaxillary orthognathic surgery . The surgical procedure was performed under general anasthesia via nasal entubation, the full thickness mucoperiosteal flap was elevated in between the distal end of the second premolars in the maxilla. Le Fort I osteotomy was performed with a bone saw in a way parallel to the Frankfort horizontal plane, at least 5 mm to root apices. The pterygomaxillary junction was separated by using a curved osteotome. An U-shaped osteotome is used to separate the nasal septum from the maxilla. The maxilla was downfractured, and the lesion within the right maxillary sinus was reached (Figure 4). The lesion was removed, and the sinuses were irrigated with using 10% povidone iodine diluted with 1/10 physiological saline solution. The excised fungus ball was sent for histopathological examination. Then the maxilla was advanced and fixed with a miniplates and screws. Following that, the patient underwent sagittal split ramus osteotomy (SSRO). Orthognathic surgery completed without any complication. In the postoperative period, the patient was awakened without any complication and was admitted to the ward. As postoperative, 1000 mg ampicillin + 500 mg sulbactam and 500 mg paracetamol were prescribed twice a day. The patient was not prescribed any antifungal medicine. She remained in the hospital for 4 days and experienced no postoperative complications. She was discharged healthily. After orthognathic surgery, the patient achieved class I occlusion and had an improved facial appearance. The histopathological examination was aspergillosis. A cone beam computed tomography (CBCT) taken for control 6 months after the operation did not show any lesions in the maxillary sinuses, and ossification of the incision lines was observed (Figure 5).

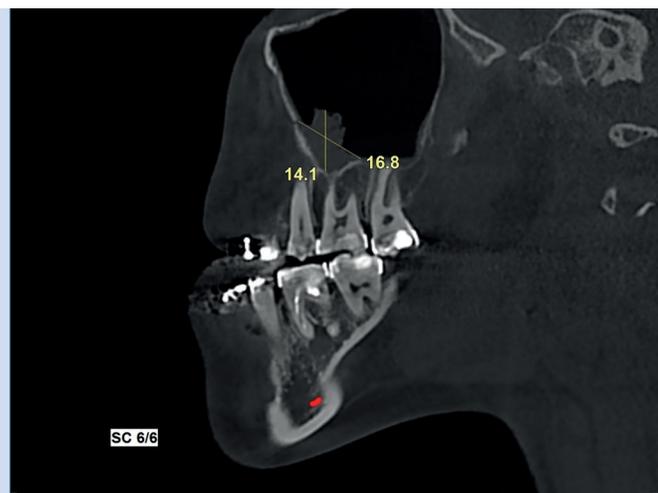


Figure 2. Sagittal CBCT image of the fungus ball in the right maxillary sinus

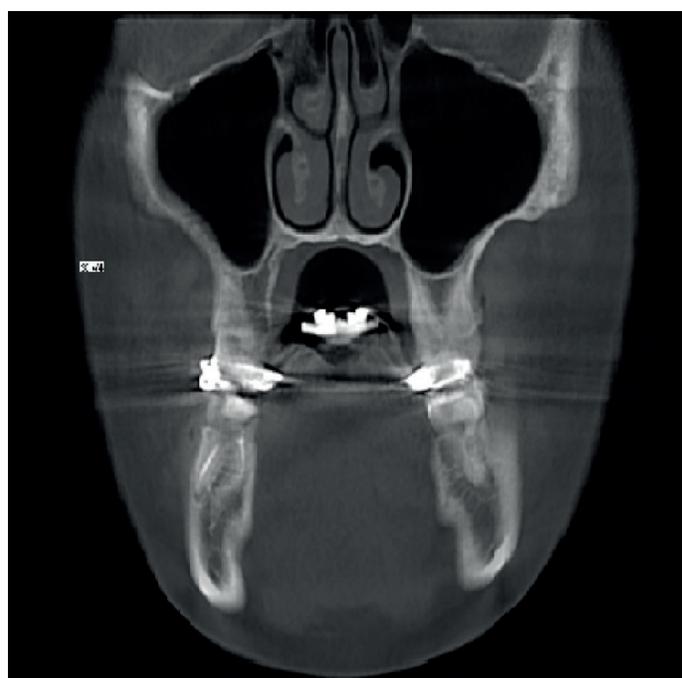


Figure 3. Coronal CBCT image before the SARPE

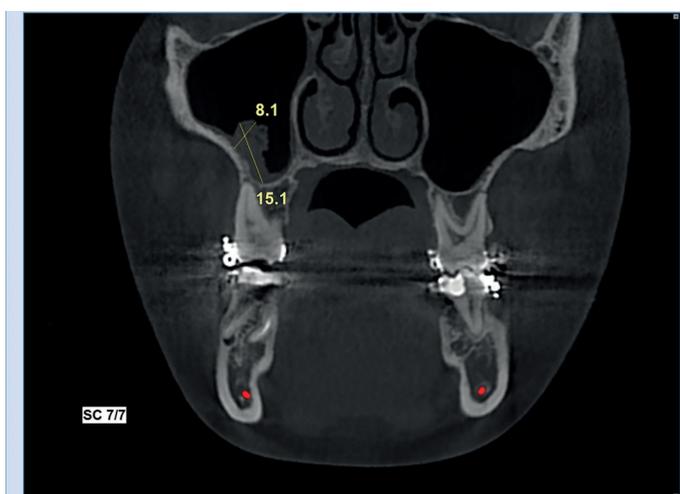


Figure 1. Coronal CBCT image of the fungus ball in the right maxillary sinus

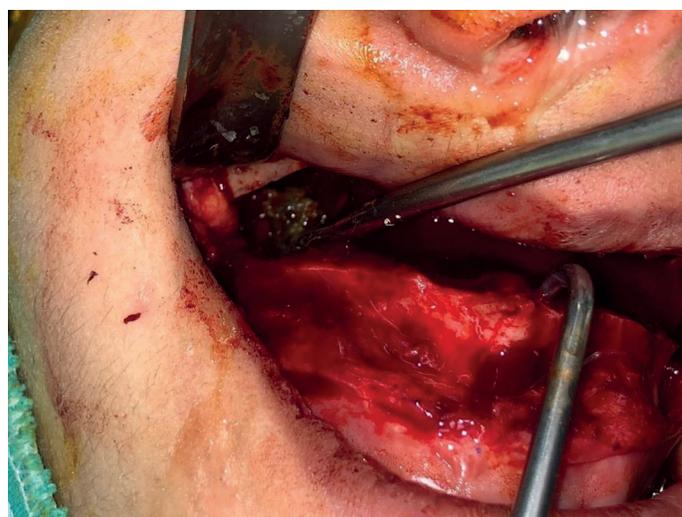


Figure 4 The fungus ball in the right maxillary sinus

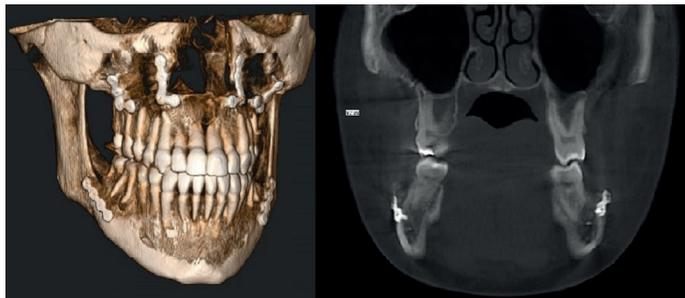


Figure 5. Coronal and 3D images 6 months later than Le Fort I surgery

DISCUSSION

SARPE procedure provides a satisfactory expansion of the maxilla, while also protecting the teeth from side effects caused by orthodontic forces (5). Complications of SARPE are lower than segmental Le Fort I osteotomies that enlarge the maxilla.

Both hereditary and acquired immunodeficiency and chronic pulmonary disease predispose to the development of a variety of pulmonary syndromes in response to *Aspergillus*. For invasive and chronic cavitary pulmonary aspergillosis, and potentially for allergic bronchopulmonary aspergillosis, patients benefit from antifungal therapy, most often with triazole medications (6). Aspergillosis is a rare disease of the paranasal sinuses (3). The fungus ball in the maxillary sinus can cause symptoms such as chronic sinusitis with nasal secretion, pain, swelling in the zygomatic region, as well as asymptomatic. Asymptomatic patients are diagnosed after routine radiographic examinations (7). The patient had any symptoms and fungus ball noticed in radiological examination in our case. Legent et al. (8) found an intense opacity caused by a foreign body within the affected maxillary sinus in 80 of the 85 maxillary sinus *Aspergillus* cases they examined. They associated 72 of these 80 cases with canal filling pastes. In 8 of the cases, there was a dense opacity within the sinus and was not associated with any dental material. Finally, 3 of the cases that did not look like foreign bodies were associated with dental pathologies. In our case, no endodontic treatment and apical pathology were observed in the teeth adjacent to the right maxillary sinus. Clinical presentation of aspergillosis is non-specific, and a diagnosis is usually suspected in imaging studies (4). With the introduction of tomography into the clinic routine, the diagnosis and treatment of many sinus disorders that have been difficult to diagnose previously has been made possible. Superficial sinonasal mycosis, or saprophytic fungal infestation, has been defined as the contamination of the nasal or sinus cavities by fungi (4, 9). In our case, both maxillary sinuses were observed to be clean in the CBCT sections taken before the SARPE procedure, but a fungus ball was visible in the sections taken before the Le Fort I osteotomy (17 months after SARPE). People who have previously undergone sinus surgery may have aspergillosis in the sinus (4). In our

case, the SARPE performed before the Le Fort I osteotomy was a surgery that was invasive to the maxillary sinuses. If the symptoms and radiographic analyses in the treatment correspond to a fungus ball, the treatment is surgical, and Caldwell-Luc surgery and endonasal endoscopic sinus surgery are the primary methods of treatment that considered (10-12). In a review by Costa et al. evaluating 262 patients with fungus ball formation in maxillary sinus, it was concluded that endonasal endoscopic sinus surgery (90 %) is a more preferred method than Caldwell-Luc surgery (10 %) (7). In our case, we cleaned the fungus ball and repositioned the maxilla in 3-dimensional space in a single session instead of two different surgeries.

CONCLUSION

It should be kept in mind that fungal infection may occur after SARPE. Maximum attention should be paid to sterilization in order to avoid such a complication or similar complications. Noninvasive aspergillosis in the sinus can be surgically removed during Le Fort I osteotomy. For orthognathic surgery patients with *Aspergillus* infection Le Fort I osteotomy might be an alternative to Caldwell-Luc and endonasal endoscopic sinus surgeries.

Conflict of interest: The authors declare that they have no competing interest.

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Patient informed consent: The patient approval was received for this case report and she signed informed consent form.

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