CASE REPORT

Unusual case of bilateral maxillary fungus ball

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SUMMARY

An otherwise healthy 34-year-old man was referred to our ear, nose and throat (ENT) clinic for a bilateral maxillary radiologic opacity. This condition was accidentally discovered with a panoramic radiography performed during a follow-up visit after a bilateral endodontic treatment. The patient did not report any specific sinonasal symptom such as purulent nasal discharge, loss of smell and cough, apart from an unspecific sinus pressure. The CT scans showed a bilateral inflammatory process into the maxillary-ethmoidal sinuses and an iron-like density within the maxillary sinuses, while nasal endoscopy showed purulent discharge in the ostiomeatal complex. The patient underwent functional endoscopic sinus surgery under general anaesthesia and the inflammatory material collected was histologically diagnosed as a rare case of bilateral fungus ball. The patient was dismissed the following day with no complications; there were not any sign of recurrence or symptoms during a 4 month follow-up.

BACKGROUND

Odontogenic sinusitis is commonplace in ear, nose and throat (ENT) practice. They are generally regarded as maxillary-only sinusitis arising from dental pathology, featuring unilateral nasal obstruction and/or purulent discharge, pain or pressure on the maxillary area and cacosmia.¹ Patients may also have episodes of gravel-like discharge from their nose.²

More modern and broader views on the subject showed that odontogenic sinusitis can follow all kind of dental procedures,³ with either unilateral or bilateral disease and mixed aerobic, anaerobe and mycotic infections,⁴⁻⁶ picturing definitely more complex scenarios.

If classic unilateral odontogenic infections usually present no diagnostic or surgical challenge, with approaching bilateral and non-symptomatic then the patient requires additional attention, especially when dealing with conditions such as fungus balls, whose bilateral presentation is almost non-existent. Even when dealing with bilateral sinonasal conditions, the risk of misdiagnosing an unusual presentation of odontogenic pathology should always be taken into account.

CASE PRESENTATION

A 34-year-old man was referred to our ENT department on incidental finding of a bilateral maxillary sinus opacification during a routine follow-up visit by his dentist. The patient underwent endodontic treatment of teeth 1.5, 1.6, 2.5 and 2.6, 1 year before. The endodontic treatment had been

successful and neither the teeth nor the gums showed any signs of infection on patient's presentation; furthermore, his oral hygiene was unremarkable. The patient did not report any specific sinonasal symptom such as purulent nasal discharge, loss of smell and cough; after specific questioning he reported an unspecific sinus pressure localised at the midface.

INVESTIGATIONS

The patient had already undergone an ordinary panoramic radiography during the follow-up dental examination. The X-ray showed a complete opacification of both maxillary sinuses accompanied by a bilateral 'iron-like' signal (figure 1), which is pathognomonic of maxillary fungus balls.⁷ The panoramic X-ray and the dentist confirmed that there were no signs of ongoing dental pathology, which should have been otherwise treated together with the sinonasal condition.

The patient was therefore referred to our clinic for completing the rhinological workup: the ENT physical examination was unremarkable and anterior rhinoscopy did not show any purulent discharge. The endoscopic examination showed a partial occlusion of the ostiomeatal complex (OMC) with a prominent anteromedialisation of both uncinate processes, a specific sign of inflammatory process in the maxillary sinus caused by the pressure of the purulent content of the sinus (figure 2). The middle meatus was occupied by a purulent discharge but no neoplastic signs were seen, so we did not perform any kind of biopsy.

Owing to the founded suspicion of odontogenic sinusitis, maxillofacial CT scans without contrast-enhancement were performed for diagnostic purposes and for potential surgical planning (figures 3 and 4). The CT scans were consistent with the orthopantomography findings: an iron-like

Figure 1 Orthopantomography: endodontic material (arrows) previously used to treat four teeth has disseminated into maxillary sinuses. The microenvironmental changes induced by the endodontic material allowed the progressive development of the bilateral fungus ball.

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Figure 2 Endoscopic view of the right nasal cavity: a purulent discharge can be seen coming from the ostiomeatal complex (lateral nasal wall). Such discharge is a pathognomonic sign of a maxillary infection, in this particular case due to an odontogenic process.

Figure 3 CT coronal image showing bilateral maxillary infection with iron-like material (arrow) inside the left maxillary sinus. The ostiomeatal complex is completely closed bilaterally. There is no sign of tissue invasion or bone erosion.

opacity, usually related to the high metal content of the core of fungus balls, was located within both maxillary sinuses which were filled with inflammatory material; in addition to this, the ethmoidal sinuses were partially involved bilaterally. Moreover, the ostiomeatal complexes (OMCs) appeared completely closed, but there were no traces of bone erosion or tissue invasion.

After the endoscopic sinus surgery (ESS) procedure, the fungus ball specimen was sent for microbiological and histological examination. Culture results on specific media for aerobes, anaerobes and fungi were positive for *Escherichia coli*, *Haemophilus paraphrophilus* and *Staphylococcus epidermidis*, while histology confirmed the presence of fungal hyphae without mucosal invasion.

Figure 4 CT axial scan shows the presence of iron-like density material in the right maxillary paranasal sinus: both sides are involved in an odontogenic disease.

DIFFERENTIAL DIAGNOSIS

In the presented case, images alone were inconclusive and different diagnoses were possible. First of all, it is important to point out the role of CT scan: it can help in a primary diagnosis of the pathological process of the paranasal sinus, but it cannot determine its aetiology. In particular, this kind of bilateral inflammatory processes can be ascribed to purulent rhinosinusitis, fungal rhinosinusitis or nasal polyposis. The lack of bony erosion or soft tissue invasion helped nevertheless to rule out malignancies.

Despite fungus ball occurring usually monolaterally,^{7 8} coupling the iron-like density in CT scans with patient's history (prior bilateral endodontic treatment), the definitive diagnosis could be easily deduced from the data available.

In case of inconclusive diagnosis, a plain head MR could give further confirmation showing a specific signal void in T2-weighted sequences, which is lacking in other odontogenic conditions.⁹ In this case, we had sufficient data to deem the MR unnecessary.

TREATMENT

Fungus balls are not expected to respond to medical therapy;⁷ therefore, the patient was proposed an endoscopic surgical procedure, as proposed by Felisati *et al.*³ This approach was pivotal for healing of inflammatory and infectious process and also for the final anatomical pathology diagnosis.

The patient therefore underwent an ESS procedure under general anaesthesia. During surgery, we created a wide bilateral middle antrostomy and performed a radical ethmoidectomy. Antrostomies allowed removing the fungal material (figure 5) from maxillary sinuses with curved suction tips and performing thorough lavages with oxygen peroxide. Bilateral nasal packs positioning prevented any further bleeding, and the patient was prescribed oral levofloxacin 500 mg once a day for 14 days. After the nasal packs removal, the patient was prescribed to

Figure 5 Intraoperative endoscopic view of the right nasal cavity. After performing right uncinectomy and antrostomy, the fungus ball (arrow) has been removed.

perform nasal douches with saline three times a day and to apply a nasal oil mix (olive, rhyline and vaseline oil) and mupirocine ointment three times a day for 30 days after surgery.

OUTCOME AND FOLLOW-UP

After removing the nasal packs, the patient was discharged the day following the procedure without any complication. The patient underwent ENT evaluation at 1, 4 and 8 weeks without the need of aggressive debridement of the surgical site. There was no recurrence or symptoms relapse during the 12 months follow-up (figure 6).

DISCUSSION

Fungus ball is a common complication of dental pathology, and it usually stems from the improper dental treatment of premolars and molars (more infrequently canines) or dental implant-related procedures.¹⁰ Dental treatments causing fungus balls range from dental reconstruction to tooth extraction and

Figure 6 Right maxillary sinus after 12 months of follow up (FU): there were no sign of recurrence or purulent discharged.

more frequently endodontic treatment. In all these cases, the final pathway is represented by the onset of an odontogenic sinusitis that, first, it is confined to maxillary sinus, than it can spread to other paranasal sinuses, particularly to ethmoidal one and even contralaterally.⁴ It is nevertheless useful to remember that non-odontogenic fungus balls do exist, but their prevalence is unknown and it is not clear how thoroughly the dental history of the patient has been evaluated.

It is widely assumed that ~30% of the maxillary paranasal sinus infections originate from an odontogenic focus.¹¹ Owing to its prevalence, Felisati *et al*³ have proposed a new classification of sinonasal complications of dental disease or treatment in order to permit the foundation of standardised treatment protocols. According to their classification, this case represents a class 3b complication (complication of classic dental treatment without oroantral communication).

The anatomical relation between the apices of premolar and molar teeth increase the likelihood, compared to the other teeth, that a pathological process affecting premolar and molar teeth will affect also the maxillary sinus. In fact, the apices of these teeth are separated from the floor of the maxillary paranasal sinus by a thin layer of bone and, occasionally, may even bulge into the sinus when the sinus is enlarged or the roots are longer than usual.¹²

Particularly, endodontic treatments are considered one of the most important risky procedures for maxillary pathology: in fact, during this procedure, the endodontic material, which contains zinc oxide-eugenol, facilitates the growth of *Aspergillus fumigatus*, paralyzes the mucosa-cilia and induces soft-tissue oedema.^{1 13} Another common consequence of teeth inflammatory process is the formation of odontogenic cyst, whose origin is still unclear:^{2 14 15} this cyst could be another odontogenic focus from which an odontogenic rhinosinusitis may occur.

Many authors have reported that mycotic infections are the most common odontogenic super-infections along with *Aspergillus fumigatus*,^{7 9 16} frequently seen in fungus ball lesion that, thanks to deShazo classification,¹⁷ is considered an extramucosal mycosis. These mycoses usually grow, like in this clinical case, in immunocompetent patients and when cultured, up to 50% have a negative culture result.¹³

Once the mycotic superinfection occurs, it generally leads to the formation of purulent material within the maxillary sinus: this condition causes an improper antero-medialisation of the uncinate process which alters the maxillary drainage pathway and also the ventilation of all the sinuses draining into the OMC. Anatomical modification of this region could therefore be considered the key in spreading the maxillary odontogenic infection to other sinuses: in fact, the ethmoid sinuses are involved in 59.7% of odontogenic disease.^{4 6 18}

Maxillary sinus infection may be seen as one of the most common complications of dental treatments. In the case of endodontic treatments, this type of complication can be prevented: in particular, a proper elimination of organic material, disinfection of the root canal system and a correct three-dimensional root obturation, using appropriate material, are all fundamental to preventing odontogenic sinusitis. In fact, it may be considered that the materials used to fill the treated root canal system (cement and others metal materials) may flow out of the apex and get into the maxillary sinus leading to inflammatory sinus reaction.¹²

Moreover, another possible cause of odontogenic sinusitis related to endodontic treatment is the incorrect anatomical modification of the most apical part of dental root caused by incorrect manoeuvres with manual and/or rotary instruments. In

addition to this, erroneous use of disinfectants for the elimination of bacteria and organic material can lead to inflammatory sinus reaction.¹²

Maxillary fungus ball is commonly seen as a monolateral lesion, and its identification as a bilateral lesion is rare: in fact there has been only one case report of maxillary odontogenic bilateral fungus ball.⁸ Nevertheless, several authors have reported that bilateral fungus ball occurs in 3% of odontogenic sinusitis.^{7 8}

Additionally, the patient's non-specific clinical presentation makes this case extraordinary rare: in fact, usual symptoms of this pathology include nasal obstruction and/or purulent discharge, pain or pressure on the maxillary area and cacosmia; patients may also have episodes of gravel-like discharge from their nose.^{2 17}

However, independently of the clinical presentation, the diagnostic gold standard is represented by head CT scans: in fact, on one hand it can help in visualising sinonasal cavities with high-resolution images; on the other hand, it can provide useful information about the dental disease within the maxillary sinus.¹² Nevertheless, radiological analyses cannot provide a proper aetiological diagnosis and nasal cultures are usually negative.¹⁹

Owing to these findings, the first-line empiric treatment proposed by Felisati *et al* is represented by antibiotic therapy (amoxicillin/clavulanate) associated with local treatment (nasal lavage and topical steroids). This medical therapy is useful for the potential resolution of the odontogenic sinusitis and for the treatment of bacterial super-infection that is virtually present in every odontogenic rhinosinusitis.¹² If the empiric medical therapy does not provide the resolution of odontogenic sinusitis, an endoscopic surgical treatment is required.^{3 13 17 20 21} In patients bearing a concurrent dental condition, a thorough evaluation and the support of an experienced oral or maxillofacial surgeon is pivotal.^{3 22}

Any long-term antimycotic medical therapy is not required after the surgery, because it achieves healing in almost 100% of patients without recurrence.³ A good antibiotic therapy is nevertheless recommended due to the ubiquitous bacterial coinfections.⁶

Learning points

- Despite its rarity, bilateral odontogenic disease (particularly, bilateral fungus ball) may be taken into differential diagnosis when there is a bilateral maxillary infection involvement and a history of dental treatment.
- In the presence of maxillary affection with iron-like density material, always examine dental history, no matter how much symptomatic is the patient and when was performed the dental treatment.
- A long-term postoperative antimycotic medical therapy is not necessary in a case of fungus ball, because surgery achieves healing in almost 100% of patients without recurrence; nevertheless, a good antibiotic therapy is recommended due to the possible bacterial coinfections.

While the management of odontogenic sinusitis remains a simple practice that can be solved with the right approach, the ENT specialist should never undervalue the problem to take this differential diagnosis into consideration when evaluating bilateral maxillary sinusitis.

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