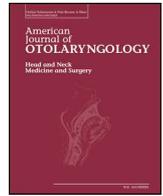




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Fungus ball of the maxillary sinus: Retrospective study of 48 patients and review of the literature

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ABSTRACT

Background: Maxillary fungus ball (FB) is the most frequent paranasal localization.

Objective: To review clinical presentation, surgery and results of treatment in our series of patients with maxillary FB. To review the literature concerning treatment of maxillary FB.

Patients and methods: 48 patients with a diagnosis of maxillary FB were treated with endoscopic sinus surgery (ESS) alone or in association with external approaches. Before surgery all patients received computed tomography (CT), nasal endoscopy and dental examination. All the patients were followed for 1 year after surgery. Studies concerning surgical treatment of maxillary FB from 2006 were reviewed.

Results: The mean age of patients was 53.6 ± 11.9 years. 20 patients (41.6%) did not present any symptom, 19 patients (39.7%) had nasal symptoms, 3 patients (6.2%) had facial pain, 6 patients (12.5%) had a combination of both. Endoscopic examination was positive in 31 patients (64.6%), 17 patients (35.4%) showed negative findings. Logit regression model demonstrated that clinical symptoms contribute to the prediction of a positive endoscopic examination. 25 patients (52.1%) presented odontogenic factors. Complete clinical and radiological resolution of FB was observed in 46 patients (95.8%).

Conclusions: Comparing our sample to the studies reviewed we may concluded that odontogenic factors were frequently reported and should be treated at the same time of maxillary FB. ESS alone or in association with external approaches is an effective treatment for patients with maxillary FB.

1. Introduction

In 1998, DeShazo proposed a classification of sinonasal fungal infections separating non invasive from invasive forms [1]. The non-invasive form is the most prevalent and FB is the most frequent type of non-invasive sinonasal fungal infections. FB generally affects immunocompetent and no atopic subjects and is localized in the maxillary sinus in more than 80% of patients [2]. It is mostly encountered in older individuals with a female predominance [3]. FB is characterized by a mass of fungal debris and mucus growing into the sinus cavity, without involvement of the underlying mucosa.

Although FB has been occasionally reported in combination with other pathology these associations must be considered incidental. Predisposing local factors such as anatomical variation of the

osteomeatal complex have been advocated but not clearly demonstrated [4,5]. A history of previous dental care and especially of dental filling was associated with maxillary with FB and seems to be an important predisposing factor [6–8].

For these reason patients with maxillary FB can be easily referred to maxillofacial departments. ESS is considered the gold standard in treating fungus FB [3,9,10]. The goal of surgery is to ensure complete removal of FB. For this reason there are different opinions regarding the size of the middle meatal antrostomy, the need to associate an inferior antrostomy or to associate other technique to ensure complete removal of the FB [11–13]. The aim of this study is to review the literature concerning treatment of maxillary FB in the last 10 years and to review clinical presentation, surgery and results of treatment in patients with maxillary FB in our department.

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2. Patients and methods

From 2001 to 2017, 536 patients underwent ESS in our department. Of those, we analyzed retrospectively the medical records of 48 patients with a diagnosis of maxillary sinus FB. The local Institutional Review Board reviewed and approved this study.

Before surgery all patients received CT scans of paranasal sinuses, nasal endoscopy and dental examination. Nasal endoscopy was reviewed in order to evaluate clinical signs of maxillary sinusitis such as mucopurulent discharge from the osteomeatal unit and presence of polypoid degeneration. All the patients were treated under general anesthesia. ESS was performed in all patients. ESS was the only surgical procedure in 11 patients. ESS with canine fossa access was performed in 13 patients. ESS with a bone window in the anterolateral wall of the maxillary sinus was performed in 24 patients. The bone window was always performed with piezoelectric instrumentations and repositioned. The canine fossa access and the bone window in the anterolateral wall of the maxillary sinus were performed to ensure complete removal of the FB and in cases where a contemporary oral surgical approach was necessary to treat an odontogenic source of infection (i.e.: dental or implant removal, closure of oroantral communication, removal of odontogenic cysts, dental apicectomy). Maxillary sinus mucosa has never been removed and abundant irrigation with normal saline was performed during surgery to make sure not to leave any fungal debris. Nasal packing was applied for 2 days. Oral antibiotics were prescribed for 1 week to prevent postoperative infections. Systemic or topical antifungal agents were not prescribed.

All the patients were followed clinically and with endoscopic examination 1 week, two weeks, 4 months and 1 year after discharge from the hospital. Four months after surgery CT scans were performed to evaluate radiological healing of the maxillary sinus. We analyzed the patients' clinical data including sex, age, medical record and dental focus.

Studies concerning surgical treatment of maxillary FB from 2006 were reviewed.

3. Results

Presentations of demographic, clinical, and therapeutic data of the patients examined are summarized in Table 1. The mean age of patients was 53.6 ± 11.9 years (range, 29 to 79). A female predominance was seen in our series with 33 female patients (69%) and 15 males (31%). The disease was predominantly unilateral in 46 patients (95.8%, 26 left, 20

Table 1
Presentation of demographic, clinical, and therapeutic data of the patients examined.

Variable	Descriptive statistics
Total number of patients	N = 48
Age	53.6 (SD 11.9) years
Sex	
Male	15 (31%)
Female	33 (69%)
Site	
Unilateral maxillary FB	46 (95.8%)
Bilateral maxillary FB	2 (4.2%)
Symptoms at presentation	
No symptoms	20 (41.6%)
Nasal symptoms	19 (39.7%)
Facial pain	3 (6.2%)
Combination of nasal symptoms and facial pain	6 (12.5%)
Nasal endoscopy	
Mucopurulent discharge	22 (45.8%)
Polypoid mucosa and mucopurulent discharge	9 (18.8%)
Negative	17 (35.4%)
Postoperative healing	
Uneventful	46 (95.8%)
Partial opacification of the maxillary sinus	2 (4.2%)

Table 2
Presence and typology of odontogenic factors in the sample.

Variable	Descriptive statistics
Total number of patients	N = 48
Presence of odontogenic factors	25 (52.1%)
Absence of odontogenic factors	23 (47.9%)
Detailed odontogenic factors	N = 25
Previous dental extraction or implant removal	5 (20%)
Dental overfilling	14 (56%)
Apical periodontitis	5 (20%)
Foreign body (luxated root)	1 (4%)

right) and bilateral in 2 cases (4.2%). Regarding clinical presentation 20 patients (41.6%) did not present any symptom, 19 patients (39.7%) had nasal symptoms similar to those of chronic rhinosinusitis (post nasal drip, nasal obstruction and rhinorrhea), 3 patients (6.2%) had facial pain, 6 patients (12.5%) had both nasal symptoms and facial pain. On the pre-operative endoscopic examination, mucopurulent discharge was observed in 22 patients (45.8%), 9 patients (18.8%) had polypoid mucosa and mucopurulent discharge, 17 patients (35.4%) showed negative findings. Regarding potential correlation 25 patients (52.1%) presented odontogenic factors in the affected side while 23 patients (47.9%) did not have any dental source of infection. Odontogenic factors are reported in Table 2. Histological examination confirms the presence of aspergillus ifae in all patients while microbiological examination was always negative for fungal growth. Complete clinical and radiological resolution of FB was observed in 46 patients (95.8%). 2 patients (4.2%) showed residual radiological mucosal thickening with increased density suggestive for incomplete FB removal. Both patients received only ESS.

In order to evaluate the relationship between clinical symptoms and objective endoscopic findings, a Logit regression model was employed. The Logit regression model was based on assuming that the residuals were distributed according to the standard binomial family of distributions, whose dispersion parameter was 1. The *p*-value of the Logit regression model (probability of obtaining the observations if the clinical symptoms had no effect on the endoscopic signs) was less than 0.001. Being the *p*-value for the model fit statistic less than 0.05, then there is evidence that the clinical symptoms contribute to the prediction of a positive endoscopic examination.

Studies reviewed concerning surgical treatment of maxillary FB from 2006 were reported in Tables 3 and 4.

4. Discussion

We found eight studies concerning treatment of maxillary FB from 2006. Four of these studies [9,10,14,15] reported a series of patients with paranasal sinus, not only maxillary, FB. This must be emphasized as clinical and treatment results are often reported in a manner from which is not possible to analyze patients with maxillary FB only. Mean age of patients (51.8 years) and a higher female versus male (66.4% vs. 33.6%) incidence are not different from our series.

Regarding symptoms two studies [11,12] did not report any evaluation. Nasal symptoms and facial pain are reported respectively in 58% and 46% of the patients in the remaining six studies. Only 5.4% of the patients are reported without symptoms.

In our series of patients we observed nasal symptoms in 39.7% of the patients, facial pain in 6.2% of the patients and a combination of both in 12.5% of the patients. In our sample there is also evidence that the clinical symptoms contribute to the prediction of a positive endoscopic examination and to our knowledge this is the first study that demonstrates such a correlation.

We observed a higher percentage of asymptomatic patients (41.6%). In our opinion this may be explained by the difference between patients referred to our department and patients referred to department of otorhinolaryngology such as all the patients of the studies reviewed. In

Table 3
Presentation of demographic, clinical, and therapeutic data of the studies reviewed (2006–2017).

Authors and institute	Year	No. patients	Age (years)	Sex	Symptoms			Nasal endoscopy			Odontogenic factor
					Female %	Male %	Nasal symptoms %	Facial pain%	None %	Positive %	
Dufour et al. [14] Department of Otorhinolaryngology Head & Neck Surgery, Centre Hospitalo-Universitaire, Poitiers, France.	2006	143*	47	60.1	40.9	50.8	37	10.4	86.1	13.9	10.8% dental overfilling
Ting-Kuang Chao and Chia-Ming Liu [12]. Department of Otolaryngology, Far Eastern Memorial Hospital, Taipei, Taiwan	2006	15	43.7	60	40	NR	NR	NR	NR	NR	NR
Pagella et al. [15] Department of Otorhinolaryngology, IRCCS Policlinico San Matteo, University of Pavia, Pavia, Italy	2007	50*	49.4	66.6	33.4	42	52	0	100	0	81% previous endodontic treatment
Pagella et al. [16] Department of Otorhinolaryngology, University of Pavia, Foundation IRCCS Policlinico S. Matteo, Pavia, Italy	2009	65	48.8	66.1	33.9	49.2	60	NR	100	0	81.5% previous endodontic treatments
Nicolai et al. [10] Department of Otorhinolaryngology, Department of Odontostomatology and the Department of Radiology, University of Brescia, Brescia, Italy.	2009	135*	52.7	73.7	26.3	64.4	61.5	8.9	48.2	51.8	86.7% previous endodontic treatments
Sawatsubashi et al. [11] Department of Otorhinolaryngology, Kyushu University, Fukuoka, Japan	2015	28	60.0	82.1	17.9	NR	NR	NR	NR	NR	NR
Garofalo et al. [17] ENT Department, University of Piemonte Orientale, Novara, Italy	2016	25	55.0	56	44	84	44	0	NR	NR	NR
Young Hoon Yoon et al. [9] Department of Otorhinolaryngology–Head and Neck Surgery, Chungnam National University, School of Medicine, Daejeon, Korea.	2017	477*	58.3	67.5	32.5	61.8	22	7.8	90.1	9.9	NR
Mean			51.8	66.4	33.6	58.7	46	5.4	84.9	15.1	

Legend *: Number of patients with maxillary FB from a larger series of paranasal sinus FB; NR: not reported.

Table 4
Surgical treatment of maxillary FB (2006–2017).

Authors and institute	Year	No. patients	Surgical technique	Recurrence	Comments
Dufour et al. [14] Department of Otorhinolaryngology Head & Neck Surgery, Centre Hospitalo-Universitaire, Poitiers, France.	2006	143	ESS	1 case, not specified if maxillary FB	CT scan findings of calcifications or metallic densities within an opacified sinus cavity are highly suggestive of fungus balls. Further studies are needed to get a clearer understanding of the true incidence, without forgetting the risk factors as well as the prevention of this particular pathology in the healthy population. In this study, we proposed a new and timesaving technique to assist the complete removal of fungus balls of the maxillary sinus with usual endoscopic instruments. Because of the excellent results, we strongly recommend this easy technique.
Ting-Kuang Chao and Chia-Ming Liu [12]. Department of Otolaryngology, Far Eastern Memorial Hospital, Taipei, Taiwan	2006	15	ESS with gauze-assisted technique	None	Actually functional endoscopic sinus surgery is the gold standard for treatment of this pathology, and antifungal therapy is unnecessary.
Pagella et al. [15] Department of Otorhinolaryngology, IRCCS Policlinico San Matteo, University of Pavia, Pavia, Italy	2007	50	ESS with sinusoscopy in 33/50 cases (66%)	2 patients	Transoral sinusoscopy can be avoided. With the assistance of lateral-view and flexible endoscopes, angled surgical equipment and maxillary saline solution irrigations, complete removal of the diseased material and sinus clearance can be achieved by a sole middle meatotomy.
Pagella et al. [16] Department of Otorhinolaryngology, University of Pavia, Foundation IRCCS Policlinico S. Matteo, Pavia, Italy	2009	65	A) 33 patients ESS and transoral approach B) 32 patients ESS	3 patients: 2 group A and 1 group B	Endoscopic surgery is a safe and effective treatment for paranasal sinuses FB.
Nicolai et al. [10] Department of Otorhinolaryngology, Department of Odontostomatology and the Department of Radiology, University of Brescia, Brescia, Italy.	2009	135	ESS	2 patients, not specified if maxillary FB	
Sawatsubashi et al. [11] Department of Otorhinolaryngology, Kyushu University, Fukuoka, Japan	2015	28	14 patients ESS with middle and inferior meatal osteotomy; 14 patients ESS with middle meatal osteotomy	2 patients in the middle meatal osteotomy group	FESS with a combination of middle and inferior meatal anastomies proved more effective for treating fungal maxillary sinusitis.
Garofalo et al. [17] ENT Department, University of Piemonte Orientale, Novara, Italy	2016	25	19 ESS with gauze technique; 6 ESS without gauze technique	1 patient in the gauze technique group	The data obtained in this study demonstrated that the "gauze technique" is a safe, simple, and quick technique, able to reduce surgery procedure time whilst providing excellent functional outcomes and patient satisfaction.
Young Hoon Yoon et al. [9] Department of Otorhinolaryngology-Head and Neck Surgery, Chungnam National University, School of Medicine, Daejeon, Korea.	2017	477	ESS	6 patients not specified if maxillary FB	Endoscopic surgery is the treatment of choice, with a low morbidity and recurrence rate.

a maxillofacial surgery department it is not unusual to see asymptomatic patients referred by dentists for occasional finding of maxillary sinus opacification during rehabilitative dental care.

In a department of otorhinolaryngology it is more likely to evaluate a symptomatic patient and this would explain the increased incidence of positive nasal endoscopy (84.9%) compared to our series (64.6%).

Evaluation of a possible correlation of maxillary FB with odontogenic factors was reported in four studies [10,14–16]. Three studies [10,15,16] reported a high incidence of previous endodontic treatment. Two of these studies [15,16] did not discuss this data. Nicolai et al. [10] reported an increased risk of maxillary FB in patients who received endodontic treatment even if this does not explain the occurrence in other sinuses or in the maxillary sinus of previously untreated patients. One study [14] reported dental overfilling in 10.8% of the patients and concluded that despite previous reports odontogenic factors had no influence in the pathogenesis of maxillary FB.

Recently Gomes et al. [7] concluded that root canals near the maxillary sinus with endodontic treatment and apical periodontitis may exhibit positive cultures for filamentous fungi. They suggested that interested professionals should be alert, because these microorganisms have pathogenic characteristics that can cause disease of odontogenic origin in the maxillary sinus.

It is of interest that even if a potential dental correlation was suspected none of the studies reviewed planned oral surgical treatment of the source of infection and all the patients were treated only with ESS.

In our sample odontogenic factors were present in 52.1% of the patients. In particular dental overfilling was present in 14 patients (56%) but also previous dental extraction or implant removal, apical periodontitis and foreign body were present as possible odontogenic factors.

The goal of surgery is to ensure complete removal of FB. Some studies reported ESS without external approaches as effective for the treatment of maxillary FB [10,16].

Complete removal of FB through the middle meatotomy was obtained with the assistance of lateral view and flexible endoscopes, angled surgical equipment and maxillary saline solution irrigations [16]. There are different opinions regarding the size of the middle meatotomy. In general FB in the maxillary sinus can be removed by middle meatotomy alone if a wide opening enables sufficient access to the maxillary sinus. However, in the case involving a large maxillary sinus completely filled with FB, canine fossa puncture, inferior meatal antrostomy and even Caldwell-Luc approach would be required. Several authors reported that observing the maxillary sinus from a middle meatus window only, it is sometimes difficult to see the anterior inferior or medial inferior wall of the maxillary sinus even when a 70° endoscope is used [11]. For this reason a combined middle and inferior meatal antrostomies has been proposed [2,11]. The canine fossa puncture has been proposed for two reasons: it is much easier to perform, it is more effective than middle meatal antrostomy for reducing the size of the large FB using microdebrider [13]. Finally the “gauze technique” was also proposed in order to avoid removal difficulties of maxillary FB in the anterior ad inferior recesses [17]. Therefore it can be concluded that ESS is the treatment of choice for maxillary FB but other external surgical approaches should be considered to ensure complete removal in selected cases. In our sample of patients we performed ESS alone in 11 patients, ESS with canine fossa access in 13 patients and ESS with a bone window in the anterolateral wall of the maxillary sinus in 24 patients. The canine fossa access and the bone window in the anterolateral wall of the maxillary sinus were performed for two reasons: to ensure complete removal of the FB and in cases were a contemporary oral surgical approach was necessary to treat an odontogenic source of infection. Patients with odontogenic factors need contemporary surgical oral approach to treat the odontogenic source of infection. Dental overfilling or apical periodontitis may be treated with apicectomy or dental extraction. Therefore, since an oral surgical approach was planned, it was useful to associate a canine fossa puncture or a bone window in the anterolateral

wall to ensure complete removal of the FB. Results of treatment in our sample of patients were similar to that reported in the studies reviewed. In particular we observed complete clinical and radiological resolution of FB in 46 patients (95.8%). Only 2 patients (4.2%) treated with ESS only showed residual radiological mucosal thickening with increased density suggestive for incomplete FB removal.

5. Conclusions

Comparing our sample to the studies reviewed we may concluded that maxillary FB can be easily referred to a maxillofacial department. A higher female versus male incidence was confirmed. Nasal symptoms, facial pain or a combination of both are common clinical presentation. About half of the patients may be asymptomatic. Clinical symptoms contribute to the prediction of a positive endoscopic examination. Odontogenic factors were frequently reported and should be treated at the same time of maxillary FB. ESS is the treatment of choice for maxillary FB but other external surgical approaches should be considered to ensure complete removal. External surgical approach through the oral cavity is useful particularly when contemporary treatment of odontogenic source of infection is planned. In our experience ESS alone or in association with canine fossa puncture or a bone window in the anterolateral wall of the maxillary sinus is an effective treatment for patients with maxillary FB.

Declaration of Competing Interest

None.

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