Odontogenic Sinusitis Caused by an Inflammation of a Dentigerous Cyst and Subsequent Finding of a Fibrous Dysplasia. A Case Report

Carmen López-Carriches1,*, Inmaculada López-Carriches2 and Rafael Baca-Perez Bryan3

1School of Dentistry, Universidad Complutense de Madrid, Spain
2Hospital Nuestra Señora del Prado, Talavera de la Reina, Spain
3Facultad de Odontología, Universidad Complutense de Madrid, Spain

Received: March 10, 2016 Revised: October 09, 2016 Accepted: October 26, 2016

Abstract: We report the case of a 38-year old male patient with sinusitis caused by an infected follicular cyst due to an ectopic impacted third molar in the right maxillary sinus. A 10-day antibiotherapy regimen was administered; subsequently, the cyst and the third molar were removed achieving complete recovery. Fibrous dysplasia was diagnosed at follow-up examination (occupation of the maxillary sinus by bone tissue was observed in a radiographic examination) and confirmed by biopsy. In cases of odontogenic sinusitis, thorough examination is crucial, as evidenced by the case reported in this study. A Literature review was performed in order to identify the diagnostic methods currently available and the clinical features, complications and treatment for both, odontogenic maxillary sinusitis and fibrous dysplasia.

Keywords: Dentigerous cyst, Fibrous dysplasia, Odontogenic maxillary sinusitis, Third molar.

INTRODUCTION

Odontogenic sinusitis is a unilateral infection of the maxillary sinus caused by a dental infection [1]. It can be found mixed with aerobic and anaerobic oropharyngeal flora such as streptococci, bacteroides, veillonella, fusobacterium, etc. [2].

A periapical infection can occasionally reach the maxillary sinus causing odontogenic sinusitis due to proximity of the upper posterior teeth. Other causes of odontogenic sinusitis include periodontitis, perforation of the Schneiderian membrane of the sinus during extraction or surgery, or the presence of root tips or other foreign bodies such as endodontic obturation materials in the sinus [2, 3]. The palatal root of the first molar is the part most frequently associated with odontogenic sinusitis [4].

An ectopic tooth in the maxillary sinus can cause sinusitis [5] as well as dentigerous cysts, keratocystic odontogenic tumor, odontomas and bone tumors like ossifying fibromas [2, 6].

There is evidence that maxillary sinusitis of odontogenic origin represents 10% of all maxillary sinusitis [3, 7]. According to Maillet [4], the incidence can raise to 51.8%.

In terms of clinical symptoms, many patients with odontogenic sinusitis do not refer upper teeth pain [1]. The most common symptoms are nasal obstruction, facial pain, headache, snoring, acute sinus perforation and swelling [8].

* Address correspondence to this author at the School of Dentistry, Universidad Complutense de Madrid, Rey Francisco, 11-bajo izda, 28008 Madrid, Spain; Tel: 0034915429435; Fax: 0034915422507; E-mail: maclopez@pdi.ucm.es
odontogenic sinusitis can manifest in the form of nasal congestion or discharge, facial pressure, anosmia and cacosmia [1, 9]. The patient may have a dental infection or have undergone previous oral surgery [10].

Diagnosis is based on a combination of clinical symptoms and radiographic findings. When sinusitis is unilateral and non-responsive to treatment, then odontogenic sinusitis should be considered [2, 9].

Panoramic radiographies have an important drawback: there are anatomical superimpositions, and visualizing the topographical relationship between the upper molars and the maxillary sinus may be difficult [7]. Nevertheless, an ectopic molar in the maxillary sinus can be easily diagnosed with a panoramic radiography [5].

Water’s view provides a good visualization of the sinus, but it is useless for detecting an ectopic molar in the maxillary sinus [8].

Three-dimensional imaging systems display multiple sections of the maxillary sinus (bone and soft tissue), whereas computed tomography eliminates the superimposition of structures [1, 4].

Cone Beam computed tomography (CBCT scanning) - a more effective imaging system- shows apical periodontitis and mucosal thickening, as well as dental pathologies related to the maxillary sinus [7, 11, 12]. CBCT scanning is a three-dimensional radiography that produces high-resolution images with low doses of radiation, as compared to conventional computed tomography scanning [3]. Although resolution and radiation dosage are very heterogeneous among different CBCT devices. Whereas, the mucosa in a healthy sinus is less than 2 mm thick, it increases to an average of 7.4 mm in chronic sinusitis [4]. Another characteristic of sinusitis is radiographic opacity due to accumulation of fluid.

A panoramic radiograph can be enough for follow-up examination [8]. Although magnetic resonance imaging provides a better visualization of soft tissue, it is not useful for odontogenic sinusitis.

Odontogenic sinusitis can be resolved by just treating the dental cause (endodontic treatment, cystectomy, etc.) [9]. However, when symptoms are acute, antibiotic treatment with amoxicillin-clavulanic acid or clindamycin is necessary. Other useful antibiotics are: trimethoprim-sulfamethoxazole or doxycycline, cefuroxime, ceftriaxone, cephalaxin, cefoxitin, ceftriaxone and azitromycin [1, 2]. Saline nasal sprays and decongestants may be necessary [2].

Surgery can be carried out endoscopically better than the Cadwell-Luc approach [2].

The infection can progress and cause serious complications such as cerebral abscess [13]. Akhaddar reported an orbital abscess associated with an odontogenic sinusitis [14].

Fibrous dysplasia (FD) is a benign tumor of the bone characterized by replacement of normal bone by fibro-osseous tissue. It is caused by somatic activating mutations in the subunit of the stimulatory G protein [15]. It begins in childhood and it usually stops at puberty, although about a third of patients have progression in adulthood [16].

FD may involve a single bone (monostotic) or multiple bones (polyostotic), and it affects long bones, ribs and cranio-facial bones more frequently. As many as 70% of lesions are monostotic and asymptomatic. Diagnosis is usually incidental finding. The extent of the lesion can be determined by computed tomography imaging, but a presumptive diagnoses must be confirmed by a histopathological study. Radiographic findings include radiolucency, ground-glass appearance, and a smoking cloudy and even radiopaque fingerprint [17, 18].

Between 10% and 25% of FD lesions involve the cranio-facial bones, being the maxilla the most frequently affected bone. Finally, it also can cause obliteration of the paranasal sinuses [16, 19].

Treatment for FD is conservative [19]. Bisphosphonate therapy is indicated for selected patients with pain, fracture risk and dysfunction. Surgery is indicated for biopsy, correction of deformity and painful lesions [20].

FD must be differentially diagnosed from ossifying fibroma, Paget’s disease and giant cell tumor [18, 21]. Regarding the prognosis, fibrous dysplasia commonly has a slow growth. Malignant change to osteosarcoma is very rare [22].

**CASE REPORT**

A 38 year-old man presented with a swollen right cheek, pain and rhinorrhea of 15 days of evolution. He had not got any systemic diseases.

The patient reports to have been examined by an otolaryngologist, who prescribed him a treatment with antibiotics. Since no remission was evident, the patient was referred to our clinic to rule out an odontogenic cause.
The patient provided an orthopantomography that revealed that the upper third molar was displaced to the maxillary sinus by the pressure of a cyst that went unnoticed by the otolaryngologist (Fig. 1).

![Initial orthopantomography](image1)

**Fig. (1).** Initial orthopantomography.

A computed tomography (CT) scan was obtained to improve diagnostic imaging, showing a cystic lesion with involvement of the bone (Fig. 2).

![Initial CT scan](image2)

**Fig. (2).** Initial CT scan.

The cyst and the ectopic third molar were dissected under local anesthesia (Articain 40/0,005 mg/ml) (Figs. 3-6). Amoxicillin 875mg/clavulanic acid 125mg, for seven days and ibuprofen 600mg, three times daily for pain control, if needed, were administered postoperatively.
Fig. (3). Right third molar surgery.

Fig. (4). Suture.
The cystic capsule was sent for histopathological examination; the diagnosis was a dentigerous cyst with signs of inflammation.

Postoperative complications were not observed and the patient remained asymptomatic over a follow-up period of seven months.

Follow-up orthopantomography imaging was performed at seven months following surgery (Fig. 7). The maxillary sinus was almost completely occupied by bone, so we decided to refer the patient for another scan. The new scan revealed that the maxillary sinus was largely and abnormally occupied by bone, what made us think of a possible fibrous dysplasia (Fig. 8). This was confirmed by a biopsy carried out by a maxillofacial surgeon and by histopathological examination. Conservative treatment was administered with regular follow-up examination.
DISCUSSION

The presence of ectopic teeth in the maxillary sinus is very rare. In this case, an ectopic third molar caused pain, inflammation and rhinorrhea. Displacement to the maxillary sinus had been caused by the pressure exerted by an odontogenic cyst.

It is very important to diagnose the odontogenic origin of maxillary sinusitis, since only the management of the odontogenic cause can lead to the resolution of the infection and avoid recurrence [7].

Most authors use 3D imaging to find the aethiological cause of maxillary sinusitis [2, 7, 23, 24]. In a survey conducted among otolaryngologists, CT scanning was the preferred imaging technique for the diagnosis of odontogenic maxillary sinusitis [6]. Before any kind of sinus surgery it is very important the sistematic study of radiographic anatomy of maxillary sinus [25]. In this sense, the patient’s CT-scan already showed a bony coating around the cystic lesion, so we should have suspected a bone lesion but it went unnoticed.
The pressure exerted by a dentigerous cyst can displace molars into ectopic positions like as in this case- the maxillary sinus [8, 26]. Other sites of ectopic third molars are the mandibular condyle, the coronoid process and the mandibular angle [27].

Dentigerous cysts are the second most common type of odontogenic cyst after radicular cyst and they can occur in the maxillary very close to the maxillary sinus [2, 28]. Differential diagnosis must be done with radicular cyst, keratocystic odontogenic tumor, ameloblastoma, even extramedullary plasmacytoma of the maxilla [29].

A dentigerous cyst may remain asymptomatic for several years [19, 23, 30]. Treatment includes the enucleation of the cyst and impacted upper third molar [2]. Histopathological examination is essential. A multidisciplinary therapy administered by a dentist and an otolaryngologist is occasionally recommended; in fact, 54% of patients have been reported to resort to a dentist to treat their maxillary sinusitis [9]. Although the symptoms may initially improve with the administration of antibiotics, if the dental pathology remains untreated, the therapy will eventually fail and the symptoms will return [6, 31].

As regards fibrous dysplasia, surgery is controversial. If the lesion is stable with no growth, annual evaluation should be enough [15]. Remodeling resection should be performed to improve the patient’s quality of life and minimize symptoms like exophthalmia, nasal obstruction, etc. However, remodeling should not be performed until the growth phase is completed, and still there is a risk of recurrence (25%) [32].

Lee [33] treats fibrous dysplasia of the maxillary sinus by decortication through the anterior wall of the maxillary sinus by removing all fibrous tissue until the sinus cavity was clear. According to Lee, internal decortication of fibrous dysplasia is effective in minimizing external spread. On the other hand, Yang [34] proposes subtotal or total resection of the lesions and reconstruction with bone grafts. Remodeling resection can cause complications like recurrent painful ophthalmoparesis due to the involvement of the superior orbital fissure [35].

Sontakke [18] has reported two cases of complete obliteration of the maxillary sinus as a result of a fibrous dysplasia. When it affects the maxillary sinus, FD can displace the orbital floor and then fractures can occur in the bones involved [36]. Also, cases of trigeminal nerve compression have been reported [37].

The dental community is wary of treating patients with fibrous dysplasia but Petrocelli in 2014 performed implant rehabilitation of the mandible in a patient with fibrous dysplasia [38]. According to Bajwa [39] dental implants may be considered if the lesions are quiescent. There aren’t many cases published, maybe there is a higher risk of implant failure even osteomyelitis development. There aren’t any complications after dental restorations, tooth extraction or orthodontic therapy [15].

CONCLUSION

In cases of odontogenic sinusitis, thorough examination is crucial as evidenced by the case reported in this study, where a sinusitis caused by a third ectopic molar associated with a dentigerous cyst was accompanied by a fibrous dysplasia that went initially unnoticed and was eventually diagnosed in follow-up examination some months later.

CONFLICT OF INTEREST

The authors confirm that this article content has no conflict of interest.

ACKNOWLEDGEMENTS

Declared none.

REFERENCES

[1] Mandal R, Patel N, Ferguson BJ. Role of antibiotics in sinusitis. Curr Opin Infect Dis 2012; 25(2): 183-92. [http://dx.doi.org/10.1097/QCO.0b013e328350f728] [PMID: 22327465]
[2] Mehra P, Jeong D. Maxillary sinusitis of odontogenic origin. Curr Infect Dis Rep 2008; 10(3): 205-10. [http://dx.doi.org/10.1007/s11908-008-0034-7] [PMID: 18510882]
[3] Cymerman JJ, Cymerman DH, ODwyer RS. Evaluation of odontogenic maxillary sinusitis using cone-beam computed tomography: three case reports. J Endod 2011; 37(10): 1465-9. [http://dx.doi.org/10.1016/j.joen.2011.06.015] [PMID: 21924204]
[4] Maillet M, Bowles WR, McClanahan SL, John MT, Ahmad M. Cone-beam computed tomography evaluation of maxillary sinusitis. J Endod 2011; 37(6): 753-7.
Kaygusuz I, Karlidag T, Akyigit A, Yalcin S. Mucocele associated with an ectopic tooth in the maxillary sinus: A case report. Int J Pediatr Otorhinolaryngol 2011; 6(4): 401-2.

[5] Sotobori M, Marukawa K, Higuchi M, et al. An Odontoma found in the Wake of Maxillary Sinusitis Onset. Case Rep Dent 2013. 2013: 834715

[6] Shahbazian M, Jacobs R. Diagnostic value of 2D and 3D imaging in odontogenic maxillary sinusitis: a review of literature. J Oral Rehabil 2012; 39(4): 294-300.

[7] Baykul T, Doğru H, Yasan H, Cina Aksoy M. Clinical impact of ectopic teeth in the maxillary sinus. Auris Nasus Larynx 2006; 33(3): 277-81.

[8] Hoskison E, Daniel M, Rowson JE, Jones NS. Evidence of an increase in the incidence of odontogenic sinusitis over the last decade in the UK. J Laryngol Otol 2012; 126(1): 43-6.

[9] Mehra P, Jeong D. Maxillary sinusitis of odontogenic origin. Curr Allergy Asthma Rep 2009; 9(3): 238-43.

[10] Laino L, Troiano G, Giannatempo G, et al. Sinus lift augmentation by using calcium sulphate. A retrospective 12 months radiographic evaluation over 25 treated Italian patients. Open Dent J 2015; 9(1): S1-10.

[11] Akhaddar A, Elasri F, Elouennass M, et al. Orbito-axial abcess associated with sinusitis from odontogenic origin. Intern Med 2010; 49(5): 523-4.

[12] Lee JS, FitzGibbon EJ, Chen YR, et al. Clinical guidelines for the management of craniofacial fibrous dysplasia. Orphanet J Rare Dis 2012; 7(Suppl. 1): S2.

[13] Reis C, Genden EM, Bederson JB, Som PM. A rare spontaneous osteosarcoma of the calvarium in a patient with long-standing fibrous dysplasia: CT and MR findings. Br J Radiol 2008; 81(962): e31-4.

[14] Carini F, Saggese V, Porcaro G, Baldoni M. Treatment of craniofacial fibrous dysplasia: a case report. Minerva Stomatol 2014; 63(1-2): 43-9.

[15] Nair UP, Nair MK. Maxillary sinusitis of odontogenic origin: cone-beam volumetric computerized tomography-aided diagnosis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010; 110(6): e53-7.

[16] Rancitelli D, Borgonovo AE, Ciccù M, et al. Maxillary sinus septa and anatomic correlation with the Schneiderian membrane. J Craniofac Surg 2015; 26(4): 1394-8.
Buyukkurt MC, Omezli MM, Miloglu O. Dentigerous cyst associated with an ectopic tooth in the maxillary sinus: a report of 3 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010; 109(1): 67-71.

López-Quales Martínez J, López Carriches C, Baca Pérez-Bryan R. Tercer molar ectópico en el ángulo de la mandíbula: caso clínico y revisión de la literatura. Cient Dent 2008; 5(2): 27-31.

Statopoulos P, Mezitis M, Kappatos C, Titsinis E, Styliani E. Cysts and tumors associated with impacted third molars: is prophylactic removal justified? J Oral Maxillofac Surg 2011; 69(2): 405-8.

López-Carriches et al. Extraodontal Plasmacytoma of the maxilla simulating a maxillary radicular cyst: quick diagnosis and management. J Craniofac Surg 2016; 27(3): e296-7.

Laino L, Troiano G, Lo Muzio L, Menditti D, Herford AS, Cicciù M. Bone healing in the surgical treatment of dentigerous cysts in critically III patients. J Craniofac Surg 2015; 26(6): 2030-1.

Longhi AB, Branstetter BF, Ferguson BJ. Otalaryngologists perceptions of odontogenic maxillary sinusitis. Laryngoscope 2012; 122(9): 1910-4.

Béquignon E, Cardinne C, Lachiver X, Wagner I, Chabolle F, Baujat B. Craniofacial fibrous dysplasia surgery: a functional approach. Eur Ann Otorhinolaryngol Head Neck Dis 2013; 130(4): 215-20.

Lee YH, Hwang K. Decortication of fibrous dysplasia of the maxillary sinus. J Craniofac Surg 2011; 22(1): 139-41.

Yang SJ, Choi JW, Chung YS, et al. Midfacial degloving approach for resectioning and reconstruction of extensive maxillary fibrous dysplasia. J Craniofac Surg 2012; 23(6): 1658-61.

Navarro-Muñoz S, Rueda-Medina I, Recio-Bermejo M, et al. Oftalmoplejia dolorosa recurrente secundaria a displasia fibrosa poliostotica del seno maxilar con afectacion de la hendidura esfenoidal. Rev Neurol 2011; 52(2): 90-4.