



Paranasal Sinus Mucocelles with Intraorbital Extension

Orbital İnvazyon Gösteren Paranasal Sinüs Mukoselleri

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Summary

Objectives: The aim of this study was to report the clinical features, management, and outcome of patients with paranasal sinus mucoccele with intraorbital extension.

Materials and Methods: Eleven patients who were diagnosed and treated for paranasal sinus mucoccele with intraorbital extension between 2005-2012 were included in this retrospective study. The clinical characteristics, treatment modalities, and complications were recorded and analyzed.

Results: The records of 11 patients (aged 25 to 69, mean 47.6 ± 15.6 years) with orbital mucoccele were included in this study. The most frequent initial symptoms and findings were proptosis in 7 patients (63.6%), diplopia in 6 patients (54.5%), and ocular movement limitation in 6 patients (54.5%). The origin of the orbital mucoccele was frontal sinus in 6 patients (54.5%), ethmoidal sinus in 3 patients (27.3%), and maxillary sinus in 2 patients (18.2%). Eight patients whose mucocelles were approachable with endoscopy (72.7%) were treated with functional endoscopic sinus surgery (ESS) and marsupialization of the sinus to the nasal cavity. Three patients' mucocelles (27.3%) were located in the lateral side of the frontal sinus and were unapproachable with endoscopy. In these patients, the endoscopic approach was combined with the osteoplastic flap technique by external approach. After surgery, all the patients' findings and symptoms improved; only one patient who had recurrent bilateral mucocelles required additional surgery. This recurrence was regressed by endoscopic surgery and no additional complications were observed.

Conclusion: The frontal and ethmoidal sinuses were the most common origin of orbital mucocelles. Proptosis, limitation of eye movements, and diplopia were the most frequently detected signs in patients with orbital mucoccele. Endoscopic sinus surgery produced favorable results in patients with an endoscopically approachable mucoccele, and the osteoplastic flap technique with external approach led to successful results in patients with endoscopically unapproachable mucocelles. (Turk J Ophthalmol 2014; 44: 297-300)

Key Words: Paranasal sinus mucoccele, orbit

Özet

Amaç: Bu çalışmanın amacı orbital uzanım gösteren paranasal sinus mukosellerinin klinik özelliklerinin, yönetiminin ve tedavi sonuçlarının bildirilmesi.

Gereç ve Yöntem: Bu retrospektif çalışmada 2005-2012 yılları arasında orbitaya uzanım gösteren paranasal sinus mukoseli tanısı ile tedavi edilmiş 11 hasta çalışma kapsamına alındı. Hastalara ait klinik özellikler, uygulanan tedavi modaliteleri ve karşılaşılan komplikasyonlar kayıt altına alınarak, analiz edildi.

Bulgular: Orbital mukoseli olan 11 hasta (ortalama yaş $47,6 \pm 15,6$ (25-69) yıl) çalışma kapsamına alındı. Olgularda saptanan en sık başlangıç semptom ve bulguları; 7 hastada (%63,6) proptozis, 6 hastada (%54,5) çift görme ve 6 hastada (%54,5) göz hareketlerinde kısıtlılık idi. Hastaların 6'sında (%54,5) mukoselin kaynağı frontal sinus, 3'ünde (%27,3) etmoidal sinus ve 2'sinde (%18,2) maksiller sinus idi. Mukosele endoskopik olarak ulaşılabilen 8 hastada (%72,7) fonksiyonel endoskopik sinüs cerrahisi ve nazal kaviteye marsupiyalizasyon işlemi uygulandı, ancak endoskopik olarak ulaşmanın mümkün olmadığı frontal sinus laterale yerleşik 3 olguda (%27,3) eksternal yaklaşımla osteoplastik flep tekniği, endoskopik yaklaşımla kombine edilmiştir. Ameliyat sonrası tüm hastaların bulguları gerilerken, bilateral frontal sinus mukoseli olan hasta haricinde nüks veya komplikasyon görülmedi. Bu hastada saptanan nüks tekrarlanan endoskopik girişimle geriledi ve ek komplikasyon izlenmedi.

Tartışma: En sık mukosel kaynağı frontal ve etmoid sinüsler idi. Proptozis, glob hareketlerinde kısıtlılık, çift görme en sık karşılaşılan bulgular idi. Mukosele endoskopik olarak ulaşılabilen olgularda fonksiyonel endoskopik sinüs cerrahisi ile iyi sonuç alınabilirken, endoskopik olarak ulaşmanın mümkün olmadığı olgularda eksternal yaklaşımla osteoplastik flep tekniği ile başarılı sonuçlar alınabilmektedir. (Turk J Ophthalmol 2014; 44: 297-300)

Anahtar Kelimeler: Paranasal sinüs mukoseli, orbita

Introduction

Paranasal sinus mucoceles occur with the collection of mucus in a cystic lesion lined with normal respiratory mucosa, as a result of the obliteration of the sinus ostium caused by chronic inflammation, trauma, iatrogenic injury, and tumors.^{1,2} The term mucocele was first used by Rollet in 1896, and its histological examination was reported by Onodi in 1901.^{3,4} An infected mucocele is called a mucopyocele. Mucoceles are slow growing lesions and may cause the thinning of the surrounding bone and may change its form. Orbital, intracranial, and facial soft tissue extensions can also occur due to expansion towards neighboring tissues. The chance of treatment without complication increases with early diagnosis. Mucoceles with intraorbital and intracranial extension may cause permanent complications. Although there are many reported cases of paranasal sinus mucoceles with intraorbital extension in the literature, studies on this subject are limited in the ophthalmology literature in our country.⁵⁻¹⁰ The objective of this study was to review our experience in the management of paranasal sinus mucoceles with intraorbital extension.

Materials and Methods

Patients who were diagnosed with and had treatment for paranasal sinus mucocele with orbital extension between 2005-2012 were included in this retrospective study. Intraorbital extension was defined as an invasion of the orbit by the mucocele through an orbital wall defect. The study was conducted in accordance with the tenets of the Declaration of Helsinki by obtaining written consent from all patients, with the approval of the local ethics review board. The ophthalmology department was the first place of application for all the patients. All patients' charts and laboratory studies were reviewed retrospectively, and

clinical features, radiologic data, ophthalmologic manifestation, management, surgical methods, recurrence, complications, and outcomes were recorded and analyzed.

Results

The mean age of the 11 patients was 47.6 ± 15.6 years (ranged from 25 to 69 years); the female/male ratio was 1.2:1. The mean period of follow-up was 18.5 ± 4.8 months. The origin of the orbital mucocele was frontal sinus in 6 patients (54.5%), ethmoidal sinus in 3 patients (27.3%), and maxillary sinus in 2 patients (18.2%). One patient (Case no 1) had bilateral mucoceles who applied to ophthalmology department with the symptoms of enlargement of the left eye and diplopia in the last 6 months. In the ophthalmologic examination of the left eye, best-corrected visual acuity (BCVA) was 0.4, and hypotropia, limitation of upgaze, and macular pucker were detected. The computerized tomography (CT) scans revealed pansinusitis and bilateral frontal sinus mucoceles - one originated from the right frontal sinus and extended to the cranium, while the other was located at the left frontal sinus with intraorbital extension. After the neurosurgery and otorhinolaryngology consultation, this case was treated with osteoplastic flap technique by external approach. During the postoperative follow-up, recurrence was detected which was treated with functional endoscopic sinus surgery (ESS) and no additional complications were observed (Figure 1). Locations were bilateral only in this patient, whereas the rest of the patients had only one mucocele (Table 1). The initial symptoms and signs of all patients are given in Table 2. All patients underwent consultation in the Otorhinolaryngology Department, and for 8 (72.7%) patients whose mucoceles were approachable with endoscopy, the first treatment choice was ESS

Table 1. Demographic and clinical characteristics of the patients

Case no	Age (Year)	Sex	Mucocele origin	Follow-up time (month)	Complication
1	29	M	Bilateral frontal sinus	14	Recurrence
2	44	F	Right ethmoidal sinus	18	-
3	58	F	Right frontal sinus	17	-
4	35	F	Left frontal sinus	13	-
5	69	M	Left maxillary sinus	24	-
6	25	M	Left frontal sinus	28	-
7	44	F	Right ethmoidal sinus	15	-
8	58	F	Right frontal sinus	16	-
9	35	F	Left frontal sinus	15	-
10	69	M	Left maxillary sinus	23	-
11	58	M	Right ethmoidal sinus	21	-

*Abbreviations: M: Male F: Female

Table 2. Presenting ocular symptoms and signs of the patients

Signs or symptoms	n	%
Proptosis	7	63.6
Limitation in ocular movement	6	54.5
Diplopia	6	54.5
Hypotropia	4	36.4
Periorbital palpable mass lesion	3	27.3
Visual loss	3	36.4
Ocular pain	2	18.2
Macular pucker	2	18.2
Ptosis	2	18.2
Optic neuropathy	1	9.1
Exotropia	1	9.1

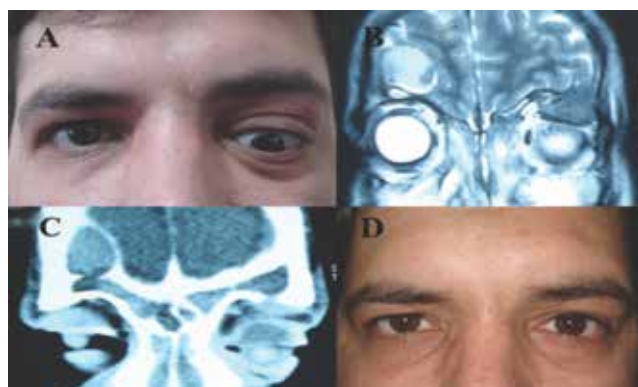


Figure 1. A) Case 1 with hypotropia and proptosis in the left eye. B) Magnetic resonance imaging of bilateral frontal sinus mucocoele with intracranial extension on the right, with intraorbital extension on the left, and mucosal thickening of all paranasal sinuses consistent with pansinusitis of Case 1. C) Computerized tomography of the case showing mass consistent with frontal sinus mucocoele originating from the left frontal sinus with extension into the left orbit which has eroded the orbital roof and with frontal sinus mucocoele originating from the right frontal sinus with intracranial extension which has eroded the roof of the sinus. D) Final postoperative photograph of the case.

and marsupialization of the sinus to the nasal cavity, whereas for 3 (27.3%) patients whose mucocoeles were unapproachable with endoscopy, the treatment was combined with osteoplastic flap technique by external approach. After surgical interventions, all patients' signs and symptoms improved, while only one patient required additional surgery.

Discussion

Paranasal sinus mucocoeles occur with the collection of mucus in a cyst, which is surrounded by the epithelium as a result of the obliteration of the sinus ostium caused by inflammation, trauma, iatrogenic injury, or tumors. They are most likely to be seen in the frontal and ethmoidal sinuses and less frequently - in the maxillary and sphenoidal sinuses.¹⁰⁻¹⁵ The growth of the mucocoele increases the pressure on the surrounding bone wall and may cause thinning. If it is not diagnosed and treated at this stage, it may cause deconstruction of the surrounding bone tissue

which leads to intraorbital, intracranial, and facial soft tissue extensions. Frontal sinus mucocoeles often occur as soft, painless mass in the superonasal and superior orbital region. It can lead to the displacement of the globe downward and forward causing diplopia. Optic neuropathy, diplopia, and visual loss may be seen due to the pressure on the globe and extraocular structure caused by orbital invasion. It may cause meningitis, brain abscess, and cerebrospinal fluid (CSF) fistula by intracranial extension.

Although paranasal sinus mucocoeles may be encountered at any age, it has been previously reported that they frequently occurred in the fourth to fifth decades. In this study, similar to previous reports, the mean age of the eleven patients was estimated as 47.6 ± 15.6 years.¹²

In their case series, Lee et al.¹² encountered complaints or signs on the eyes of 81 (98.8%) of 82 patients who had mucocoeles with intracranial or intraorbital extensions. These complaints were ptosis in 27 (32.9%) patients, periorbital swelling in 24 (29.3%), blurred vision in 20 (24.4%), exophthalmos in 5 (6.1%), and eye pain in 3 (3.6%) patients. In their study that included 15 (62.5%) patients with orbital involvement and 24 mucocoeles, Khong et al.¹³ detected displacement of the globe in 11 patients (73.3%) and non-axial proptosis in 4 (26.6%) patients, diplopia in 6 (40%), eyelid edema in 7 (46.6%), epiphora in 2 (13.3%), vision loss in 1 (6.6%), ptosis in 2 (13.3%), and periorbital palpable mass lesion in 2 (13.3%) patients. Kim et al.¹⁴ detected ophthalmic symptoms in the eyes of 96 out of 97 patients in their 17-year retrospective study, and periorbital swelling and pain were the most encountered symptoms in 35 patients (36.4%). Optic neuropathy was detected in 18 patients (18.8%) who had decreased visual acuity and relative afferent pupillary defect. Ten of these patients showed improvement after the mucocoeles were treated surgically. The authors found that the presence of infection was the only significant factor for visual outcome. In their study of 15 cases with orbital mucocoele, Wang et al.¹⁵ found proptosis in 10 patients (66.7%), diplopia in 5 (33.3%), limitations in ocular movements in 4 (26.7%), periorbital pain in 4 (26.7%), periorbital palpable mass lesion in 4 (26.7%), ptosis in 3 (20%), visual loss in 3 (20%), headache in 2 (13.3%), and relative afferent pupillary defect in 1 patient (6.66%). Orbital mucocoeles arising from the frontal and ethmoidal sinuses frequently present with proptosis or palpable mass in the periorbital area, while the maxillary and sphenoidal sinus mucocoeles are less common and related to optic neuropathy and decreased visual acuity.¹⁵ Similar to the literature, in this study, the most common origin of the mucocoeles were frontal sinus (54.5%) and ethmoidal sinus (27.3%), which due to the mass effect in the orbital cavity, resulted mostly in proptosis (63.6%), diplopia (54.5%), and displacement of the globe (36.4%). We detected decreased visual acuity in three patients, two of which occurred due to macular pucker and one was related to optic neuropathy. In our series, there was only one maxillary sinus mucocoele but no sphenoidal sinus mucocoele, and due to this disruption of the patients, we detected optic neuropathy only in one patient. All of these symptoms improved after treatment in all patients.

A CT scan is an excellent diagnostic tool for identifying the size, location, and extension of the mucocoele. In CT scans, mucocoeles appear as homogeneous isodense lesions that do not enhance with contrast unless they are infected. They are typically space occupying lesions from the paranasal sinus with surrounding bony erosion.¹⁵ Magnetic resonance imaging provides the differentiation of mucocoele from paranasal sinus carcinoma, soft tissue, and dural inflammation visible on magnetic resonance imaging (MRI) in T1 and T2 weighted examination.¹⁶⁻¹⁸ In our department, we preferred CT scanning as the first choice imaging tool, whereas in selected cases such as those who were diagnosed with optic neuropathy, intracranial extension or suspected carcinoma, CT scanning was combined with MRI.

Traditionally, treatment for paranasal sinus mucocoele involved complete removal of the sinus mucosal lining and obliteration of the sinus. Although an external approach by Lynch-Howarth incision or Caldwell-Luc technique was accepted in the past, with the improvements in endoscopic sinus surgery of today, endoscopic surgery of sinus mucocoele is widely preferred with its low rate of morbidity and recurrence.^{5-7,19,20} Different from the endoscopic surgical approach which is common in the literature, Wang et al.¹⁵ practiced transcanalicular surgery in 8 patients, Lynch technique in 3 patients, transforix approach in 2 patients, and functional endoscopic sinus surgery in 1 patient in their study of 15 cases with orbital mucocoele. No recurrence was encountered in any of the patients. Lee et al.¹² treated 77 patients with marsupialization and modified Lothrop procedure combined with intranasal drainage, 4 patients with an external approach, and 1 patient with external combined with endonasal. In our study, we performed functional ESS and marsupialization to the nasal cavity for mucocoeles that were approachable with endoscopy, but for cases located in the lateral side of the frontal sinus, thus being unapproachable with endoscopy, the osteoplastic flap technique with external approach was combined with the endoscopic approach. We tried to use the endoscopic technique as much as possible since it is less harmful to the nasal structures and physiology, leaves no scar aesthetically, and provides an early return to daily life.^{11-15,19,20}

Late diagnosis and treatment of paranasal sinus mucocoele with orbital extension can cause serious complications. In general, ophthalmologic symptoms were the most common clinical presentations of these patients. Therefore, paranasal sinus mucocoeles should be considered for the diagnosis of orbital masses and treated by both ophthalmologists and otorhinolaryngologists. This is particularly important for early diagnosis, and rapid surgical intervention by a collective work of ophthalmologists and otorhinolaryngologists is necessary to prevent patients from having permanent complications. Functional ESS, which has proved to be a successful treatment in the published literature, is recommended as the primary means

of managing paranasal sinus mucocoeles, and the osteoplastic flap technique with external approach gives successful results in patients with endoscopically unapproachable mucocoeles.

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