CEO Clinical and Experimental Otorhinolaryngology Vol. 6, No. 2: 103-106, June 2013
http://dx.doi.org/10.3342/ceo.2013.6.2.103
pISSN 1976-8710 eISSN 2005-0720

Case Report

Ethmoidal Mucocele Presenting as Oculomotor Nerve Paralysis

Dae Woo Kim[1] · Hee-Young Sohn[2] · Sea-Yuong Jeon[2,3] · Jin-Pyeong Kim[2,3] · Seong-Ki Ahn[2,3] · Jung Je Park[2,3]
Seung Hoon Woo[2,3] · Dong Gu Hur[2,3]

[1]*Department of Otorhinolaryngology, Seoul Metropolitan Government-Seoul National University Boramae Medical Center, Seoul;*
[2]*Department of Otorhinolaryngology, Gyeongsang National University School of Medicine, Jinju;*
[3]*Institute of Health Science, Gyeongsang National University, Jinju, Korea*

A 56-year-old male was admitted with an acute headache and sudden ptosis on the right side. No ophthalmological or neurological etiologies were apparent. A mucocele of the right posterior ethmoid sinus was observed with radiology. After the marsupialization of the mucocele via a transnasal endoscopic approach, the patient's symptoms (oculomotor nerve paralysis and headache) resolved in 4 weeks. Oculomotor paralysis is a rare symptom of an ethmoidal mucocele. In this article, we report this rare case along with a literature review.

Keywords. *Blepharoptosis, Mucocele, Oculomotor nerve*

INTRODUCTION

A mucocele is a slowly progressing benign lesion caused by the retention of mucous secretions and expansion due to the continuous obstruction of the orifice of the sinus or a minor salivary gland [1,2]. Substances such as prostaglandin E2 and collagenase are released at the capsule of the mucocele, which degrades the bone and allows the mucocele to expand to adjacent areas. Mucoceles develop most frequently in the frontal sinus, the anterior ethmoid sinus, and the sphenoid sinus. Common symptoms include exophthalmos, limited eye movement, and headache. However, it is very rare that a mucocele in the ethmoid sinus induces oculomotor nerve paralysis. Such a case is reported here along with a review of the literature.

• Received April 12, 2010
Revised May 31, 2010
Accepted June 24, 2010

• Corresponding author: **Dong Gu Hur**
Department of Otorhinolaryngology, Gyeongsang National University Hospital, 79 Gangnam-ro, Jinju 660-702, Korea
Tel: +82-55-750-8852, Fax: +82-55-759-0613
E-mail: mdhur@hanmail.net

CASE REPORT

A previously healthy 56-year-old man presented with acute headache, ocular pain, and ptosis on the right side beginning one day prior to the visit (Fig. 1). The patient did not complain of deterioration of visual acuity or any nasal symptoms such as rhinorrhea, nasal obstruction, or hyposmia. The patient had no history of surgery or facial trauma, and the previous medical history was unremarkable.

On admission, vital signs were normal. General laboratory tests, including complete blood count, electrolytes, cholesterol, glucose, erythrocyte sedimentation rate, and chest radiography were normal. During the ophthalmological examination, visual acuity was 20/20 oculus uterque (OU). Confrontation visual fields were also normal. Pupils were equal at 4 mm OU with normal reactivity and no relative afferent pupillary defect. Ductions were full oculus sinister (OS). The oculus dexter (OD) would not adduct beyond the midline; supraduction and infraduction were diminished, but abduction was intact. Exophthalmometric, biomicroscopic, and ophthalmoscopic examinations showed normal findings. Weakening of muscle strength and sensory deterioration that may have suggested myasthenia gravis

were not detected in neurological examinations. Serologic tests such as acetylcholine receptor antibody, rheumatoid factor, and antineutrophil cytoplasmic antibody were all negative.

Brain magnetic resonance imaging and computed tomography (CT) documented no lesions on the brain or orbit. However, a mucocele was detected in the right posterior ethmoid sinus. It was located beside the apex of right orbit, and there was suspicious bony erosion but no dehiscence of the lamina papyracea on the CT images (Fig. 2A). The nasal cavity appeared normal upon endoscopic examination.

Endoscopic sinus surgery was performed under general anesthesia. After shrinking the turbinate mucosa with epinephrine-soaked gauze, an endoscopic examination was done with a 4-mm diameter, 0 degree nasal endoscope. The uncinate process and bulla ethmoidalis were serially resected with cold instruments under endoscopic view. Ethmoidectomy was performed through the third lamella to the posterior ethmoid sinus. At this point, the capsule of a mucocele with viscous mucus and yellow fluid with inspissated debris was found. There was no bone dehiscence on the lamina papyracea under the surgical field. Endoscopic marsupialization was performed. Headache and eyeball pain were noticeably reduced immediately after the operation. Improvement of ocular symptoms was observed at an outpatient clinic 2 weeks postoperatively and symptoms were completely resolved 4 weeks after operation. The patient has been followed for more than 30 months and with no recurrence of symptoms.

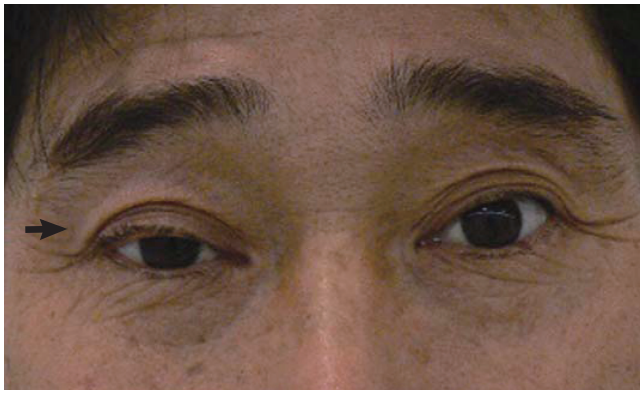

Fig. 1. Preoperative photodocumentation. The arrow indicates ptosis of the right eyelid.

DISCUSSION

Mucoceles in the nasal sinus are induced by mucus retention caused by the obstruction of the natural orifice due to chronic inflammation, allergy, surgery, trauma, and fibrous dysplasia within the nasal sinus [1-4]. Mucoceles are common in the frontal and ethmoid sinuses, but development in the maxilla is rare. Clinical symptoms vary depending on the location, the cyst size, and the involvement of adjacent tissues. Swelling, pain, and other ophthalmic symptoms appear due to the expansion and compression of adjacent tissues by the mucocele or inflammatory infiltration from the mucocele via the ethmoidal vein [1,2,4-6]. Oculomotor nerve palsy is a very rare symptom in mucocele patients. Well-documented cases of oculomotor nerve paralysis complicated by a mucocele have been reported in the English literature (Table 1). The sphenoid sinus (six cases) was most common mucocele site, followed by the frontal sinus (two cases). There was no case of an ethmoidal mucocele reported in the English literature.

Although ptosis was a common symptom among these cases, other oculomotor nerve paralyses showed different characteristics according to the involved sinus. The limitation of upward gaze was usually observed with frontal mucoceles, while sphenoid mucoceles presented complete extraocular oculomotor nerve palsy including pupilloparesis. According to the literature review, two up-gaze, two medial-gaze, one down-gaze and two pupillary limitations were documented in sphenoid mucoceles. This phenomenon can be explained by the anatomy of the oculomotor nerve within the orbit (Fig. 2C). The oculomotor nerve divides in the anterior cavernous sinus into superior and inferior branches, which enter the orbit through the superior orbital fis-

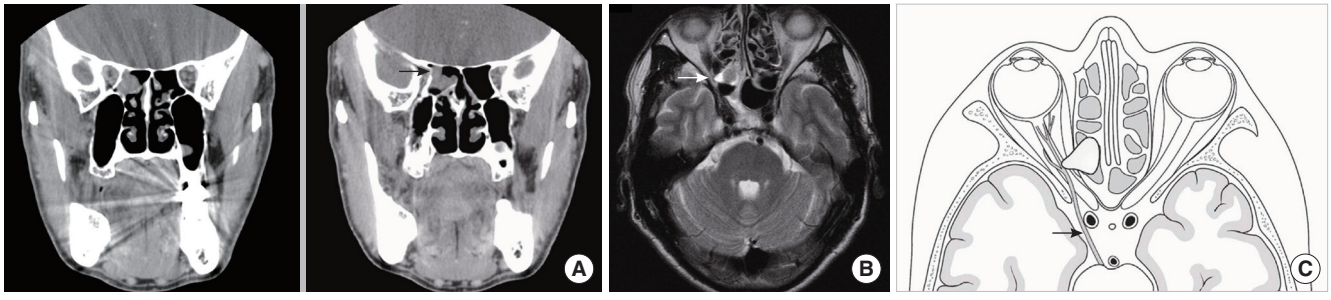

Fig. 2. Preoperative images. (A) Two coronal images of computed tomography without enhancement. A small mucocele was found in the right posterior ethmoid sinus (left). The arrow indicates the erosion of lamina papyracea (right). (B) T2 weighted magnetic resonance image. The arrow indicates the mucocele in the posterior ethmoidal sinus. (C) A simple illustration of the magnetic resonance imaging image. The arrow indicates the suggestive oculomotor nerve pathway from the brainstem. The mucocele resides where the oculomotor nerve spreads its branches superiorly and inferiorly to the external ocular muscles.

Table 1. Cases presenting with isolated oculomotor nerve paralysis due to sinonasal mucocele published in the English literature

Author	Sex/age	Oculomotor symptom	Sinus	Imaging finding	Treatment	Recovery time
Freidmann [7]	M/48	Ptosis complete extraocular 3rd palsy	Sphenoid	Internal carotid: displaced backwards and laterally	Excision via external approach	1 week
Ehrenpreis [8]	M/79	Ptosis limited up-gaze	Frontal	Extension into the orbit	Excision via external approach	4 weeks
Sethi [9]	M/44	Ptosis	Sphenoid	Erosion of posterolateral wall	Endoscopic marsupialization	Several weeks
Sethi [9]	F/61	Ptosis dilated pupil	Sphenoid	Erosion of posterolateral wall	Endoscopic marsupialization	2 days
Lin [10]	M/20	Ptosis limited up-gaze	Frontal	Extension into the orbit	Endoscopic marsupialization	2 days
Akan [11]	F/64	Ptosis limited medial-gaze	Sphenoid	Extension into cavernous sinus and optic canal	Endoscopic marsupialization	4 weeks
Prepageran [12]	M/72	Ptosis dilated pupil	Sphenoid	Erosion of side wall	Endoscopic marsupialization	2 weeks
Vaphiades [13]	F/90	Ptosis limited up-gaze	Sphenoid	Erosion of side wall	Endoscopic marsupialization	4 weeks
This case	M/56	Ptosis limited up-, medial-, down-gaze	Ethmoid	Erosion of side wall	Endoscopic marsupialization	4 weeks

sure. The superior ramus passes superomedially over the optic nerve and supplies the superior rectus and levator palpebrae superioris muscles. The inferior ramus divides into three branches: one passes beneath the optic nerve to the media rectus, another runs inferiorly to the inferior rectus, and the third runs forward to the inferior oblique muscles and is superiorly given off to the lower part of the ciliary ganglion. In summary, the superior division and the nerve to the medial rectus are located on the superiomedial or medial side of optic nerve, and other nerves, including the parasympathetic root, are on the lateral side. Therefore, a frontal mucocele may damage the superior division of the oculomotor nerve, which innervates the superior rectus and levator palpebrae superioris muscles by compressing the orbital structures, while sphenoid or ethmoid mucoceles can compromise the medially located branches of the oculomotor nerve to the medial rectus muscle. In addition, sphenoid mucoceles can present all of the extraocular symptoms of the oculomotor nerve as well as pupilloparesis because the orbital apex around the sphenoid sinus is very narrow and vulnerable to pressure or local inflammation. In our case, ophthalmologic examinations showed upward, medial, and inferior limitations of eye movement with ptosis, indicating the involvement of every oculomotor branch except the parasympathetic fiber. It was a very rare presentation, but one that could be explained by anatomical relationships.

There are some reports about other neuro-ophthalmic manifestations of paranasal mucoceles [14-18]. Sphenoid mucoceles have been associated with optic neuropathy more often than proptosis due to anatomical characteristics [14]. Therefore, sphenoid sinus mucoceles may cause orbital apex syndrome presenting with visual loss and opthalmoplegia without proptosis [15]. Abducens nerve palsy with sphenoid mucocele and trochlear nerve palsy with sphenoethmoidal mucocele were also reported [16-18].

The pathophysiology of nerve palsy in the orbit caused by mucoceles is not well-known. Some authors reported that mucoceles probably causes ischemia by compressing the microvascular supply to the nerve, based on the documentation that papillary sparing, usually observed in sphenoid mucoceles, mimics a vasculopathic process observed in diabetic ophthalmoplegia [19]. Interestingly, frontal mucoceles brake the wall of the frontal sinus and extend into the orbit, but sphenoid mucoceles usually erode the lateral wall without extension into the orbit or the cavernous sinus. These findings might support the mechanism by which oculomotor nerve palsy associated with frontal or sphenoid mucoceles is caused by direct compression or indirect damage, respectively.

The principles of treatment are to resect the cyst, including the sinus mucosa, completely to prevent recurrence and to secure the orifice of the nasal sinus. Either an extranasal or an intranasal approach may be used. An extranasal approach is performed primarily in cases that may be difficult to manage via an endoscopic approach; for example, a cyst in the lateral side of the frontal sinus or one divided by the septum. However, the use of the extranasal approach has been limited because of esthetic problems, difficult postsurgical management and unclear postsurgical radiological interpretations. With the development of nasal endoscopic surgery, the intranasal approach has been performed more widely. This approach reportedly has fewer postsurgical complications and minimal mucosal damage and offers a better postoperative view for close observation [1,6,9]. Most cases were treated with endoscopic marsupialization of the mucocele, usually resulting in rapid regression of the ophthalmic manifestation within 4 weeks.

In conclusion, oculomotor nerve paralysis may be an initial presentation in paranasal mucoceles. We recommend evaluating the paranasal sinuses in cases of oculomotor nerve palsy with-

out definitive neurologic or ophthalmologic etiologies. In these cases, surgical treatment of the mucocele may improve oculomotor nerve palsy.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Kim DY, Kwon BW, Park DE. Clinical evaluation of paranasal sinus mucocele. J Clin Otolaryngol. 2004 Feb;15(1):93-7.
2. Lee KC, Kwon KH, Kang HG, Jin SM, Lee YB. Endoscopic sinus surgery in paranasal sinus mucoceles. Korean J Otolaryngol-Head Neck Surg. 1997 Jun;40(6):855-60.
3. Beasley NJ, Jones NS. Paranasal sinus mucoceles: modern management. Am J Rhinol. 1995 Sep-Oct;9(5):251-6.
4. Kennedy DW, Josephson JS, Zinreich SJ, Mattox DE, Goldsmith MM. Endoscopic sinus surgery for mucoceles: a viable alternative. Laryngoscope. 1989 Sep;99(9):885-95.
5. Canalis RF, Zajtchuk JT, Jenkins HA. Ethmoidal mucoceles. Arch Otolaryngol. 1978 May;104(5):286-91.
6. Kim SS, Kang SS, Kim KS, Yoon JH, Lee JG, Park IY. Clinical characteristics of primary paranasal sinus mucoceles and their surgical treatment outcome. Korean J Otolaryngol-Head Neck Surg. 1998 Nov;41(11):1436-9.
7. Friedmann G, Harrison S. Mucocoele of the sphenoidal sinus as a cause of recurrent oculomotor nerve palsy. J Neurol Neurosurg Psychiatry. 1970 Apr;33(2):172-9.
8. Ehrenpreis SJ, Biedlingmaier JF. Isolated third-nerve palsy associated with frontal sinus mucocele. J Neuroophthalmol. 1995 Jun;15(2):105-8.
9. Sethi DS, Lau DP, Chan C. Sphenoid sinus mucocoele presenting with isolated oculomotor nerve palsy. J Laryngol Otol. 1997 May;111(5):471-3.
10. Lin CJ, Kao CH, Kang BH, Wang HW. Frontal sinus mucocele presenting as oculomotor nerve palsy. Otolaryngol Head Neck Surg. 2002 May;126(5):588-90.
11. Akan H, Cihan B, Celenk C. Sphenoid sinus mucocele causing third nerve paralysis: CT and MR findings. Dentomaxillofac Radiol. 2004 Sep;33(5):342-4.
12. Prepageran N, Subramaniam KN, Krishnan GG, Raman R. Ocular presentation of sphenoid mucocele. Orbit. 2004 Mar;23(1):45-7.
13. Vaphiades MS, Roberson GH. Sphenoid sinus mucocele presenting as a third cranial nerve palsy. J Neuroophthalmol. 2005 Dec;25(4):293-4.
14. Soon SR, Lim CM, Singh H, Sethi DS. Sphenoid sinus mucocele: 10 cases and literature review. J Laryngol Otol. 2010 Jan;124(1):44-7.
15. Kumagai M, Hashimoto S, Suzuki H, Matsuura K, Takahashi E. Orbital apex syndrome caused by sphenoethmoid mucocele. Auris Nasus Larynx. 2003 Aug;30(3):295-7.
16. Ayberk G, Ozveren MF, Yildirim T, Ercan K, Cay EK, Koçak A. Review of a series with abducens nerve palsy. Turk Neurosurg. 2008 Oct;18(4):366-73.
17. Lin JY, Lin SL, Chang YL, Lo SH, Chuang FS, Lin SY. Sphenoid sinus mucocoele presenting with optic neuropathy and abducens palsy: a late complication of radiotherapy to the head and neck. Eye (Lond). 2005 Jun;19(6):697-9.
18. Kimakura M, Oishi A, Miyamoto K, Yoshimura N. Sphenoethmoidal mucocele masquerading as trochlear palsy. J AAPOS. 2009 Dec;13(6):598-9.
19. Johnson LN, Hepler RS, Yee RD, Batzdorf U. Sphenoid sinus mucocele (anterior clinoid variant) mimicking diabetic ophthalmoplegia and retrobulbar neuritis. Am J Ophthalmol. 1986 Jul;102(1):111-5.