Ethmoid pneumocele presenting with exophthalmos 15 years after endoscopic sinus surgery

Michelle Song, D.O.,1 Sun M. Ahn, M.D.,1 Douglas R. Reh, M.D.,1 and Josef Shargorodsky, M.D., M.P.H.1,2

ABSTRACT

Introduction: A pneumocele is an abnormal dilation of an air-containing sinus beyond the normal margins of bone, with associated bony thinning. A delayed ethmoid pneumocele after sinus surgery has not previously been reported.

Methods: A case report of a patient with a delayed ethmoid pneumocele after sinus surgery. The diagnostic workup, operative approach, and postoperative results were evaluated.

Results: A 57-year-old female with a history of endoscopic sinus surgery 15 years prior presented with right eye proptosis and severe orbital and facial pressure. A maxillofacial computed tomography showed a markedly expanded air-filled right anterior ethmoid space with a dehiscent lamina papyracea, consistent with a pneumocele. Marsupialization of the pneumocele as well as a revision ethmoidectomy were performed, with a visible return of the orbital contents to a more natural position. The patient experienced worsened diplopia immediately postoperatively that resolved within two weeks.

Conclusions: This case demonstrates that a pneumocele can present even years after endoscopic sinus surgery, and acute but temporary development or worsening of diplopia can result from surgical decompression of the pneumocele as the eye returns to its natural position.

(After Rhinol 6:e129–e132, 2015; doi: 10.2500/ar.2015.6.0123)

A pneumocele is an abnormal dilatation of an air-containing sinus beyond the normal margins of the bone, with thinning of the associated sinus bony wall.1 This is a rare disorder that has most commonly been described in the frontal sinus.2,3 The likely mechanism involves a one-way valve allowing air into a sinus space, creating positive pressure and associated expansion within this space. It is important to differentiate a pneumocele from pneumosinus dilatans, a condition that describes chronic air-filled expansion of a paranasal sinus but without the associated bone thinning.4,5 Pneumoceles of the anterior ethmoid sinus are extremely rare, with only a single case reported in the literature.6 That case reported a pneumocele that occurred acutely after endoscopic drainage of an ethmoid sinus mucocele. We report a case of an expansile anterior ethmoid sinus pneumocele causing lateral displacement of the medial orbit wall, which occurred 15 years after endoscopic sinus surgery.

CASE REPORT

A 57-year-old African American female, with a history of endoscopic sinus surgery 15 years prior, presented to a tertiary medical center Emergency Department with right eye proptosis and severe pressure and pain increasing in severity for 2 weeks. Her previous sinus operation, performed at a different institution, was for recurrent sinusitis, and consisted of bilateral endoscopic maxillary antrostomy, total ethmoidectomy, and sphenoidotomy. Her sinusitis episodes decreased in frequency and severity after her operation. Before this event, she denied any ophthalmologic abnormalities. On examination, she had intact visual acuity but with right side proptosis and exophthalmos, with significant gaze restriction, minimal extraocular motion, and diplopia. An ophthalmologic evaluation noted normal visual acuity but with an elevated intraocular pressure, which normalized with topical Latanoprost. The remainder of her head and neck examination was normal, with no significant sinonasal inflammation on anterior rhinoscopy or nasal endoscopy.

From the 1Department of Otolaryngology-Head and Neck Surgery, Johns Hopkins University School of Medicine, Baltimore, Maryland, and 2Coastal Ear Nose and Throat, Neptune, New Jersey
The authors have no conflicts of interest to declare pertaining to this article
Address correspondence to Josef Shargorodsky, M.D., M.P.H., Coastal Ear Nose and Throat, 3700 Route 33 Suite 101, Neptune, NJ 07753
E-mail address: jshargor@gmail.com
Copyright © 2015, OceanSide Publications, Inc., U.S.A.
was entered, with immediate associated air-bubbling (Fig. 2). The lamina papyracea was noted to be absent. The patient was discharged home postoperatively, but later that evening started noticing worsening diplopia. The diplopia worsened for approximately the next three days. At her one-week visit, she was noted to have significantly improved extraocular motion but was severely bothered by the diplopia. Nasal endoscopy demonstrated a more medial orbital position as compared with its intraoperative position. An ophthalmologic evaluation demonstrated normal visual acuity, intraocular pressure, and extraocular motion. At her two-week visit, the diplopia had resolved, and at the one month visit, her endoscopic examination showed her right globe to be in a normal anatomic position (Fig. 3).

DISCUSSION

We report the first case of a delayed ethmoid pneumocele after endoscopic sinus surgery. A pneumocele is a lesion characterized by abnormal expansion of an air-containing sinus. Urken et al. developed a classification system for the expansion of the paranasal sinuses based on a spectrum of radiologic and clinical findings. The feature that differentiates pneumocele from pneumosinus dilatans is the loss of integrity of part, or all, of the bony sinus wall due to bony erosion. Pneumosinus dilatans refers to an air-containing sinus cavity that is abnormally expanded but with intact sinus walls of normal thickness. In contrast, a pneumocele refers to an aerated sinus with either focal or generalized thinning of the bony sinus wall. In previous reports, however, the two terms have been used interchangeably. Furthermore, it may sometimes be difficult to differentiate pneumosinus dilatans from pneumocele radiographically. Reports vary as to the frequency of affected sinuses, but the frontal sinus has been the most commonly reported affected sinus.

The pathogenesis of pneumoceles is not clearly understood, although several etiologies have been suggested. Postulated causes of pneumocele include iatrogenic, developmental (arachnoid cyst), inflammatory, neoplastic (optic nerve sheath meningioma, fibro-osseous disease), and traumatic, leading to obliteration of the sinus and subsequent hyperinflation. Wolfensberger and Herrmann were the first to document the trap valve hypothesis in a patient with a maxillary sinus pneumocele. The theory is that redundant mucosa or a stricture may create a one-way valve between the nasal cavity and the affected sinus, and produce chronic air trapping, intrasinus pressure disequilib-

Figure 1. A, Coronal computed tomography scan shows the right anterior ethmoid sinus pushing the right medial rectus and displacing the optic nerve and globe. Right side Lamina papyracea is not visible. Thinning and erosion of the bone of the lateral wall of the anterior ethmoid air cell suggests that this is a pneumocele rather than pneumosinus dilatans. B and C, Axial computed tomography showing protrusion of the right anterior ethmoid pneumocele laterally, with erosion of the lamina papyracea and compression of the orbital contents.
Pneumoceles tend to remain asymptomatic over long periods of time. Repeated air trapping due to positive pressure results in progressive dilation of the sinus cavity. Symptoms occur when the pressure on neighboring structures, such as orbital contents or the trigeminal nerve, reaches a critical level. The expansion may progress over years and may worsen with atmospheric pressure changes such as flights or scuba diving. There are no signs of sinusitis or inflammation in most reported cases. The treatment of a pneumocele is surgical, consisting of decompression of the involved sinus with a large sinusotomy. Postoperative care should be taken to ensure the patency of the sinus opening.

Our patient had a remote history of endoscopic sinus surgery 15 years before presentation, and a recent upper respiratory infection that coincided with the onset of symptoms related to her pneumocele. A possible disease mechanism is that inflammation from the upper respiratory infection created a one-way valve into a partially intact ethmoid air cell. Then Valsalva or sneezing could have created positive pressure and pushed air through the open valve resulting in the pneumocele, with associated thinning of the lamina and lateral displacement of the orbit. It is also possible that this was a more chronic process, with an accelerated acute expansion of the pneumocele with Valsalva from sneezing or nose blowing. In our case, the revision ethmoidectomy released the positive pressure from the anterior ethmoid air cell. The patient’s worsened postoperative diplopia was likely due the resulting medial shift of the orbital contents. The diplopia resolved as the patient adjusted to the normalization of the orbital position.

In conclusion, this case demonstrates that, although rare, a pneumocele can present even years after endoscopic sinus surgery. Acute but temporary develop-
ment of diplopia can result from surgical decompression of the pneumocele, as the position of the orbital contents normalizes.

REFERENCES

1. Urken ML, Som PM, Lawson W, et al. Abnormally large frontal sinus. II. Nomenclature, pathology, and symptoms. Laryngoscope 97:606–611, 1987.
2. van Schayck R, and Niedeggen A. Pneumosinus dilatans after prolonged cerebrospinal fluid shunting in young adults with cerebral hemiatrophy. A report of two cases and review of the literature. Neurosurg Rev 15:217–223, 1992.
3. Appelt EA, Wilhelmi BJ, Warder DE, and Blackwell SJ. A rare case of pneumosinus dilatans of the frontal sinus and review of the literature. Ann Plast Surg 43:653–656, 1999.
4. Malinvaud D, Halimi P, and Bonfils P. Pneumosinus dilatans associated with nasal polyposis. B-ENT 7:283–287, 2011.
5. Acar M, Yucel A, Degirmenci B, et al. Pneumocele vs. pneumo-sinus dilatans: Review of the literature with a case of frontal sinus pneumocele. Tohoku J Exp Med 202:295–297, 2004.
6. Gupta A, Durkin SR, Muecke JS, et al. Ethmoidal pneumocele following drainage of an ethmoidal mucocele. Orbit 27:161–163, 2008.
7. Walker JL, and Jones NS. Pneumosinus dilatans of the frontal sinuses: Two cases and a discussion of its aetiology. J Laryngol Otol 116:382–385, 2002.
8. Boulos PR, Bernardino CR, and Rubin PA. Pneumocele—a rare cause of air in the orbit. Am J Ophthalmol 138:168–169, 2004.
9. Patel AC, Hammoudeh JA, and Urata MM. Pneumosinus dilatans: A rare cause of frontal sinus expansion—case report and review of literature. J Oral Maxillofac Surg 66:2380–2386, 2008.
10. Wolfensberger M, and Herrmann P. The pathogenesis of maxillary sinus pneumoceles. Arch Otolaryngol Head Neck Surg 113:184–186, 1987.
11. Braverman I. Pneumocele of the maxillary sinus with orbital and trigeminal nerve involvement: Case report and review of the literature. J Otolaryngol Head Neck Surg 38:E35–E38, 2009.
12. Vines FS, Bonstelle CT, and Floyd HL. Proptosis secondary to pneumocele of the maxillary sinus. Neuroradiology 11:57–59, 1976.