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DOI: 10.46903/gjms/21.03.1348

CASE REPORT

ACUTE UNILATERAL OPTIC NEUROPATHY SECONDARY TO ONODI CELL MUCOCELE: A CASE REPORT

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ABSTRACT

Introduction: Optic neuropathy refers to damage or impairment of the optic nerve, which can result in vision loss. Onodi cell mucocele is a rare condition that can lead to optic neuropathy and other health-threatening complications. Here we present a case of successfully treated unilateral optic neuropathy secondary to Onodi cell mucocele associated with chronic sinusitis.

Case report: A 45-year-old woman presented with a 4-day history of right eye pain associated with decreased vision and photophobia with chronic nasal obstruction. Physical examination showed pain on downward extraocular muscle movement, vision acuity of 6/60 and 6/9, and right eye Relative Afferent Papillary Defect (RAPD). CT scan findings led to the diagnosis of right optic neuropathy secondary to Onodi air cell mucocele. Emergency endoscopic sinus surgery was successfully performed in the form of anterior and posterior ethmoidectomy and bilateral sphenoidotomy with drainage of Onodi cell mucocele to release pressure on the optic nerve.

Conclusion: Overall, optic neuropathy from Onodi air cell mucocele is a rare but serious complication that requires prompt diagnosis and treatment to prevent permanent vision loss. This case report details the clinical and radiological presentation and management of Onodi cell mucocele and associated optic neuropathy, highlighting the need for healthcare providers to consider it, especially in patients with optic neuropathy and chronic sinusitis.

KEY WORDS: Onodi cell; Mucocele; Acute optic neuropathy; Optic Nerve Decompression.

Cite as: Assiri AM, Kadasah SK, Al-Qannass AM, Aldhabaan SA, Dlboh SSA, Al-Malki AQ. Acute unilateral optic neuropathy secondary to Onodi cell mucocele: a rase report [case report]. Gomal J Med Sci 2023 Jul-Sep;21(3):197-200. https://doi.org/1046903/gjms/21.03.1348

INTRODUCTION

Mucoceles are chronic benign epithelial cystic lesions arising from the paranasal sinus mucosa.[1] Its etiology is still controversial, but it seems to be correlated to a chronic inflammatory process occurring in a closed space, causing asymptomatic ostial obstruction and accumulation of mucus secretions and, later on, present as insidious ocular or intracranial complications.[2,3] Onodi cell mucocele affects frontal sinuses in 65% of cases, ethmoid cells in 25%, the maxillary sinuses in 10%, and the sphenoid sinuses. Onodi cell mucocele is a rare case with 3-42% global prevalence, and more prevalent in Asians (50-60%).[4-6]

The sphenoethmoidal cell, known as the Onodi cell that was initially described by Onodi in 1904, is a posterior ethmoid air cell that has pneumatized superolaterally into the sphenoid sinus in a close area to the internal carotid artery and optic canal, creating an optic nerve bulge in the posterior ethmoid.[7] Onodi cell mucocele can compress optic nerve endings because of their pneumatization pattern and anatomic closeness to the optic nerve, resulting in ophthalmologic problems.[7,8] Due to the anatomical complexity, endoscopic marsupialization of the mucocele is the most widely accepted treatment protocol.[9] Compressive optic neuropathy from Onodi cell-related mucocele is a rare complication, with only a few cases reported in the literature.[8,10] Therefore, we presented a rare case of unilateral visual loss due to an Onodi cell mucocele.

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Date Submitted: 30-05-2023
Date Revised: 16-07-2023
Date Accepted: 19-08-2023

CASE PRESENTATION

We present a woman aged 45 with a 4-day history of right eye pain associated with decreased vision and photophobia. She had a long-time history of nasal obstruction but no foul-smell discharge or any other remarkable medical history.

Physical examination revealed a conscious, alert patient with normal vital signs. No meningeal irritation signs, cranial nerves were intact, and normal muscle power and reflexes as well as normal coordination. Ophthalmic examination showed pain on downward left extraocular eye movement, leading to the suspicion of right optic neuritis with sinusitis as a differential diagnosis.

Brain Computed Tomography (CT) scan showed right sphenoethmoidal cell (Onodi cell) opacification with asymmetrical sphenoid sinuses and bone thinning, bilateral minimal mucosal thickening of sphenoid and maxillary sinuses, and bilateral inferior turbinate hypertrophy (Figure 1a). Further evaluation by Magnetic Resonance Imaging (MRI) was suggested. While waiting for the MRI results, the patient was put on vancomycin 500 mg IV every 6 hours, ceftriaxone 1000 mg IV every 12 hours, intranasal steroid 3 drops nasal three times a day, nasal decongestant 2 puffs before bedtime, and oral paracetamol 1000mg every 6 hours. Emergency endoscopic sinus surgery was performed in the form of anterior and posterior ethmoidectomy and bilateral sphenoidotomy with drainage of Onodi cell mucocele (Figure 1b) to release pressure on the optic nerve together with septoplasty, and the patient's pain and headache started improving afterward; however, the right eye vision kept deteriorating.

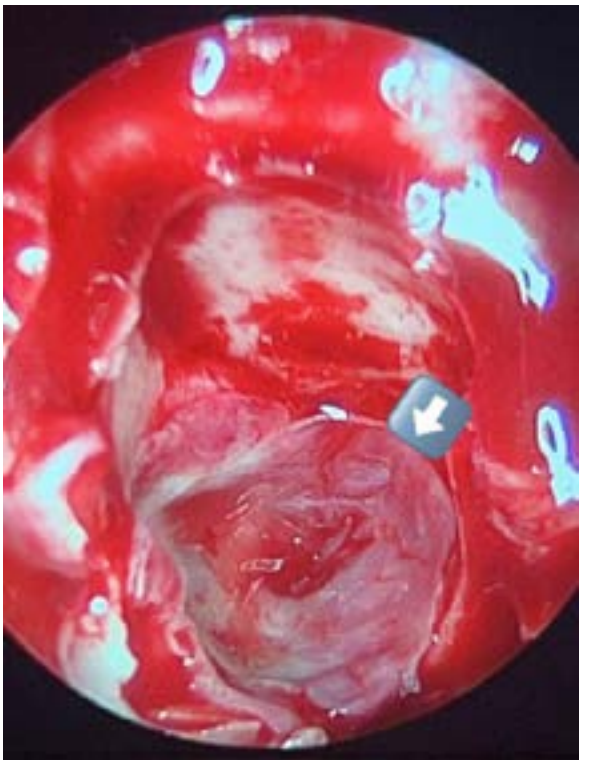

Figure 1b: Drainage of Onodi cell mucocele

Pre-surgical intervention MRI showed mild mucosal thickening in paranasal sinuses, normal optic nerve, and degenerative cervical spinal changes with multilevel discopathy at C4/5-C6/7 spinal levels. The continuously deteriorating vision led to the second ophthalmic examination that revealed the Right Eye

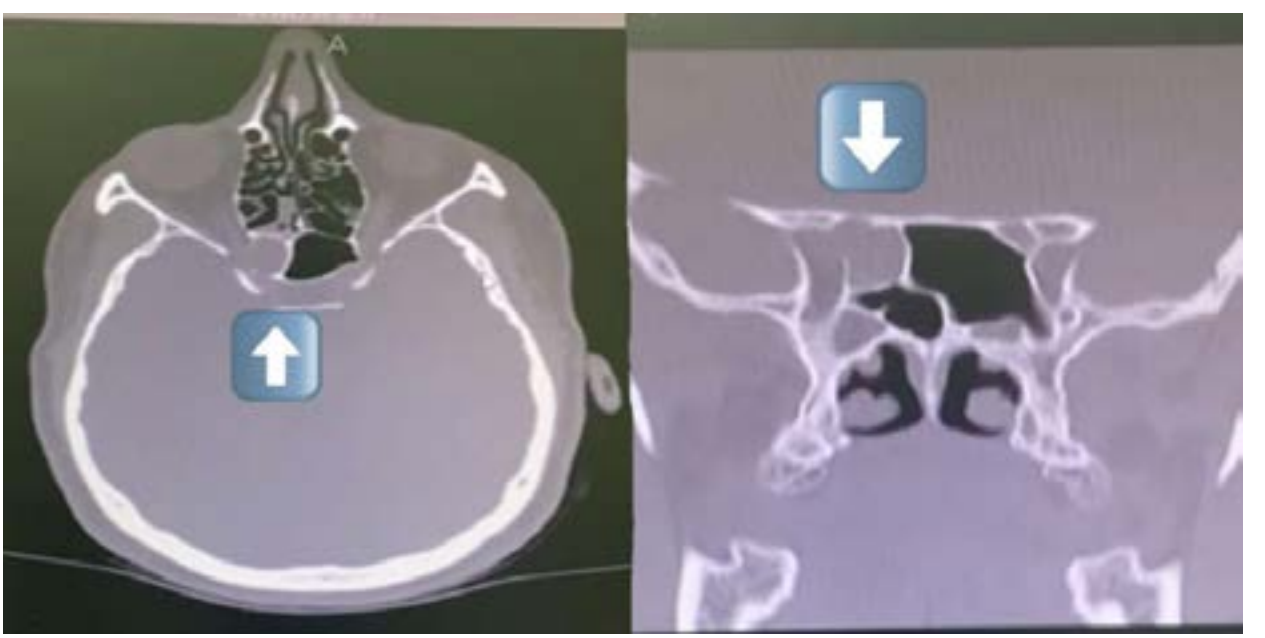

Figure 1a: CT scan showing right sphenoethmoidal cell (Onodi cell) opacification and other features of pansinusitis.

Relative Afferent Papillary Defect (RAPD) and consideration of right retrobulbar optic neuropathy. The second CT scan of the paranasal sinuses showed open sinuses but there was a recollection of fluid despite the previous drainage.

The family requested to be transferred to a higher-level hospital instead. The patient was referred to the King Fahad Medical City in Riyadh, where ophthalmic examination showed visual acuity of 6/60 and 6/9, right eye RAPD with color vision affected and NFL defect on both OCT and perimetry, fundus not swollen, and normal extraocular muscle movement.

The CT scan at the King Fahad Medical City showed post-drainage residual opacification of the right sphenoethmoidal air cells (Onodi air cells) with thinning of its right/lateral wall bone and small focal lateral bulge towards adjacent posterior right orbit, indenting the posterior aspect of the adjacent part of the right optic nerve. On arrival at the King Fahad Medical City, no procedure was done because the patient's symptoms improved, and she was discharged with a follow-up appointment at the Rhinology Skull base clinic.

DISCUSSION

This case report details the presentation of a patient with right-eye vision. The patient had a history of chronic sinusitis, and imaging revealed an Onodi air cell, a mucocele, a type of sphenoethmoidal air cell located near the optic nerve. The mucocele pressed the optic nerve, leading to optic neuropathy and subsequent vision. Our case is similar to other cases previously reported in the literature, presenting with unilateral visual problems following optic neuropathy from Onodi cell mucocele.[11–3]

Onodi air cell mucocele is a rare condition, with an estimated incidence of 0.3-6.3% in patients with chronic sinusitis,[7,8,10] as they are thought to arise from the accumulation of mucus within the air cells due to obstruction of the sinus drainage pathway.[13] A previous review of 16 Onodi air cell mucocele cases found that 12 (75%) were among Asian patients, which aligns with previous reports.[10]

Research has demonstrated that 0.7-8% of all sphenoid sinuses develop optic nerve dehiscence.[10] The majority (86%) of those cases occur in patients with posterior ethmoid or sphenoid sinus mucocele, indicating expansion of the mucocele, which directly compresses and absorbs the optic canal bone wall.[10,14] This aligns with the CT scan findings for our patient that showed right/lateral sphenoethmoidal air cell wall bone thinning. Another previous case report of similar pathology showed a mass invading the orbital cavity, leading to the compression of eyeball [9].

Optic neuropathy from Onodi air cell mucocele can occur due to several mechanisms. First, the mucocele can grow in size and compress the optic nerve, leading to nerve damage, which was the mechanism for our patient's neuropathy. Second, the mucocele can cause inflammation within the sinus, leading to compression and damage of the optic nerve. Third, the mucocele can cause pressure changes within the sinus, leading to changes in the blood supply to the optic nerve, resulting in nerve damage due to ischemia.[15]

The symptoms of optic neuropathy from Onodi air cell mucocele can vary depending on the severity of the nerve damage. Patients may experience blurry vision, loss of peripheral vision, complete vision loss in the affected eye, and facial pain.[3,12,13] Other symptoms may include headache, eye pain, and color vision changes,[13] of which were our patient's main symptoms.

Diagnosis of optic neuropathy from Onodi air cell mucocele typically involves a combination of imaging studies, such as CT or MRI scans, and ophthalmologic exams to assess visual function.[12,16] The imaging modalities help visualize the mucocele and its relationship to the optic nerve and guide the interventions. Treatment options depend on the severity of the compression and may include mucocele surgical removal or optic nerve decompression through a minimally invasive endoscopic procedure.[12] In some cases, vision loss may be irreversible, and supportive measures such as visual aids or rehabilitation may be necessary.[13]

Endoscopic sinus drainage is the most frequent surgical technique for Onodi cell-associated optic neuropathy because it is less intrusive, reduces the risk of complications, and has a shorter recovery period for patients.[11] This serves as decompression, relieving pressure from the optic nerve with subsequent neuropathy resolution. The decompression surgery assisted by endoscopy remains the most used technique, and it was also used to manage similar cases reported in the literature.[11-13] Endoscopy technology helps prevent associated morbidity and longer hospitalization.[9]

When treating Onodi cell mucoceles, especially during a surgical procedure, the optic nerve's expansion into the sphenoid sinus should be considered and monitored. The prevalence of optic nerve protrusion ranges from 4.1 to 35%, with Asian populations having a greater frequency (up to 69%).[10] This protrusion is seen as an extension of the optic canal into the sinus cavity, exposing more than half of the circumference of the nerve.

CONCLUSION

Optic neuropathy secondary to Onodi air cell mucocele is a potentially serious complication. Healthcare providers should be aware of this potential complication in patients with chronic sinusitis, particularly those with visual symptoms. Early diagnosis and treatment are crucial in preventing irreversible damage to the optic nerve. Patients with Onodi air cell mucoceles should be monitored closely for symptoms of optic

neuropathy, and prompt referral to an ophthalmologist or neurologist should be made if these symptoms arise. Further research is needed to better understand Onodi cell mucoceles pathophysiology and develop effective treatment strategies for this condition.

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CONFLICT OF INTEREST
Authors declare no conflict of interest.
GRANT SUPPORT AND FINANCIAL DISCLOSURE
None declared.

AUTHORS' CONTRIBUTION

The following authors have made substantial contributions to the manuscript as under:

Conception or Design:	AMA, SKK
Acquisition, Analysis or Interpretation of Data:	AMA, SKK, AMA, SAA, SSAD, AQA
Manuscript Writing & Approval:	AMA, SKK, AMA, SAA, SSAD, AQA

All the authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.