

## Case Report



# Sphenoethmoid mucocoele: a rare cause of optic neuropathy

Witoon Mitarnun<sup>1\*</sup>, Korrapat Aegakkatajit<sup>2</sup>, Nutdanai Roila<sup>3</sup>, Nithinan Uepechpong<sup>4</sup>

<sup>1</sup>Department of Internal Medicine, Buriram Hospital, Buriram, Thailand

<sup>2</sup>Department of Otolaryngology, Buriram Hospital, Buriram, Thailand

<sup>3</sup> Department of Internal Medicine, Faculty of Medicine, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand

<sup>4</sup>Department of Pathology, Buriram Hospital, Buriram, Thailand

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### Abstract

**Introduction:** The causes of optic neuropathy include ischemic optic neuropathy, infectious agents, inflammatory processes, and compressive lesions. Although a paranasal sinus mucocoele is a rare cause of compressive optic neuropathy, it can be treatable with early diagnosis and appropriate management. This study aims to highlight the importance of recognizing and managing paranasal sinus mucocoeles as a treatable cause of optic neuropathy.

**Case Report:** A 61-year-old woman with diabetes mellitus and dyslipidemia who presented with a one-month history of vision loss in her left eye. Magnetic resonance imaging revealed a cystic lesion compressing the left optic nerve, suggestive of a sphenoethmoid mucocoele. Mucocoele marsupialization and antibiotics improved her vision, highlighting the paranasal sinus mucocoele as a potential cause of optic neuropathy.

**Conclusion:** Recognizing paranasal sinus mucocoeles as a potential cause of optic neuropathy is important, despite their rarity, as they may be treatable. A multidisciplinary approach, including surgical intervention and infection control, is key to their management.

### Introduction

Optic neuropathy is characterized by damage or dysfunction of the optic nerve, which can result in vision loss or impairment. Various etiological factors contribute to optic neuropathy, including ischemic optic neuropathy, infectious agents, inflammatory processes, and compressive lesions.<sup>1</sup> Liu et al. discovered that pituitary adenoma (35%), meningioma (17%), and aneurysm (13%) were frequent causes of compressive optic neuropathy.<sup>2</sup> Additionally, they observed a notable correlation between the duration until treatment and the degree of visual field improvement.<sup>2</sup>

Paranasal sinus mucocoeles are one of the less common causes of compressive optic neuropathy.<sup>3-5</sup> They are cystic lesions lined with epithelium and filled with mucus, which develop due to the blockage of sinus ostia. This blockage can occur due to various factors such as chronic inflammation, infection, polyps, or prior sinus surgery. As mucus accumulates within the sinus, it could lead to cyst enlargement and erosion of the sinus bony wall.<sup>5</sup> They are typically located in the frontal and ethmoid sinuses. The common symptom of paranasal sinus mucocoeles is nasal congestion. Additional manifestations might include facial pain and headaches. If they expand and press on adjacent tissues, patients may encounter vision issues or neurological symptoms.

In the context of this report, we present a rare case of

compressive optic neuropathy due to a sphenoethmoid mucocoele, which exhibited clinical improvement after treatment.

### Case Report

A 61-year-old woman with diabetes mellitus and dyslipidemia, who was taking medication including metformin (2000 mg/d), glipizide (20 mg/d), and simvastatin (10 mg/d), presented at Buriram hospital, a government regional hospital located in Buriram province, Thailand, with a one-month history of painless, progressive vision loss in her left eye. Before this visit, she had been to the private clinic several times regarding her condition, but no definitive diagnosis had been made. The patient's vision problem persisted and progressed further. She had previously been diagnosed and treated for COVID-19 at another hospital, presenting with mild upper respiratory infection symptoms three weeks before the onset of visual symptoms in the left eye. Other aspects of her medical history were not significant. She denied any prior visual problems.

At the outpatient department, an ophthalmologist conducted an examination, demonstrating a relative afferent pupillary defect (RAPD) in the left eye. Her best-corrected visual acuity was 20/20 in the right eye and hand motion in the left eye. Fundoscopic examination revealed no disc swelling. Other neurological examinations,

\*Corresponding Author: Witoon Mitarnun, Email: [miwitoon@gmail.com](mailto:miwitoon@gmail.com)

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including other cranial nerve assessments, motor function tests, sensory examination tests, and cerebellar function tests, as well as the general physical examinations, including cardiovascular, respiratory, and abdominal assessments, were unremarkable. An ophthalmologist initially suspected non-arteritic ischemic optic neuropathy (NAION), considering diabetes mellitus as a predisposing factor. However, other causes of optic neuropathy could not be ruled out. Subsequently, the ophthalmologist consulted the neurologist to admit the patient to the department of internal medicine for further investigations.

The blood analysis, including the complete blood count, along with assessments of kidney and liver function, revealed no concerning abnormalities. Her fasting blood sugar was 171.8 mg /dL, with a hemoglobin A1c level of 10.8%. Her cholesterol level was 160.3 mg /dL, with a low-density lipoprotein level of 86 mg /dL. Serology tests for human immunodeficiency virus and hepatitis B and C virus, as well as the Venereal Disease Research Laboratory test, all yielded negative results. Magnetic resonance imaging (MRI) of the orbit with gadolinium revealed a well-defined cystic lesion (measuring 2.8 cm × 2.2 cm × 1.9 cm) in the left sphenothmoid sinus, accompanied by adjacent bone destruction and compression of the left optic nerve (see Figure 1).

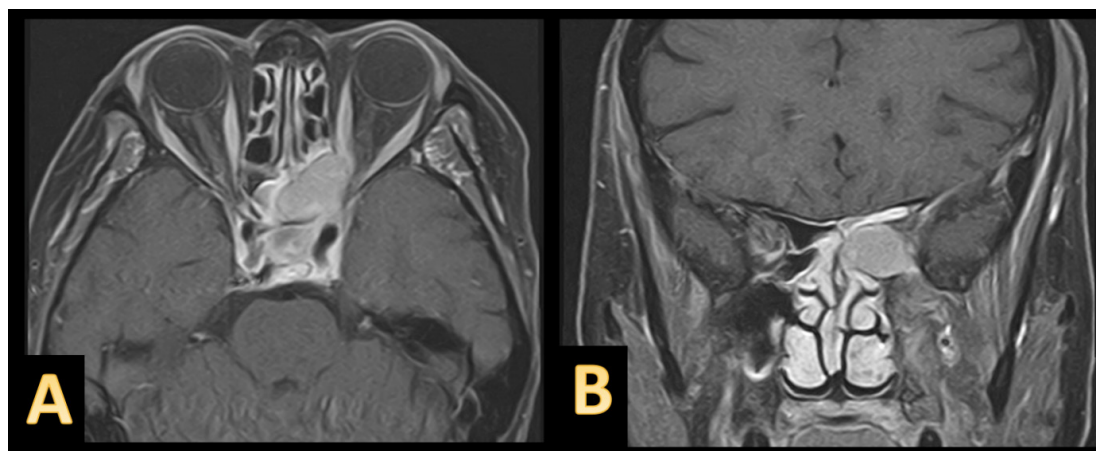
Therefore, an otolaryngology consultation was promptly requested. The endonasal endoscopic surgery, with ethmoidectomy and a large sphenoidotomy to marsupialize the lesion, was performed the following day without any intraoperative or postoperative complications. Intraoperative findings included clear, yellow fluid in the cystic lesion. By histopathological evaluation of the cyst wall tissue, a pathologist confirmed the diagnosis of mucocoele. Post-operation, she received oral amoxicillin/clavulanate (amoxicillin 1,750 mg/d and clavulanate 250 mg/d) for one week to control the infection. One week post-operation, she experienced improved vision in her left eye, with the visual acuity in her left eye improving from hand motion to finger

counting.

## Discussion

The patient experienced painless vision loss in the left eye without swelling of the optic disc. Additionally, there was a positive RAPD in the left eye, leading to a diagnosis of optic neuropathy. In this case, the differential diagnosis of optic neuropathy includes post-COVID-19 optic neuritis, NAION, compressive optic neuropathy, and other potential causes.<sup>1,6</sup> However, an orbit MRI with gadolinium revealed a distinct cystic lesion located in the left sphenothmoid sinus, showing associated bone damage and compression of the left optic nerve, consistent with a paranasal sinus mucocoele. Other potential differential diagnoses, apart from a paranasal sinus mucocoele, encompass paranasal sinus tumors such as inverted papilloma or sinonasal carcinoma. However, most tumors typically exhibit enhancement with gadolinium contrast.<sup>7</sup> Another feasible diagnosis is mucus retention cysts, but these usually do not entirely fill the sinus and typically do not cause any bony erosion.<sup>7</sup> Nonetheless, our case was confirmed as a diagnosis of paranasal sinus mucocoele based on pathological findings.

In our case, the cause of the sphenothmoid mucocoele might be attributed to a previous upper respiratory tract infection due to COVID-19, resulting in the blockage of sinus ostia and accumulation of mucus, subsequently leading to the development of a mucocoele. The mucocoele causes osteolysis through the action of prostaglandins and collagenases, resulting in bony destruction and subsequent damage to adjacent structures, including the optic nerve.<sup>5</sup> The study by Kim et al. investigated ninety-six cases of paranasal sinus mucocoeles in patients presenting with ophthalmologic symptoms.<sup>8</sup> Periorbital swelling and pain were the most prevalent, occurring in 36.4% of the patients. Additionally, eighteen patients (18.8%) were diagnosed with optic neuropathy.<sup>8</sup> Among these eighteen patients, ten experienced improvements in their vision following surgical intervention.<sup>8</sup> The study also revealed



**Figure 1.** subtitle: Gadolinium-Enhanced Orbital MRI of a Patient with Sphenothmoid Mucocoele. Axial (A) and coronal (B) gadolinium-enhanced orbital MRI revealed a well-defined cystic lesion without gadolinium enhancement in the left sphenoid sinus extending to the left ethmoid sinus, with adjacent bone destruction and compression of the left optic nerve, consistent with a sphenothmoid mucocoele

that the presence of infection was the determining factor affecting postoperative visual outcomes.<sup>8</sup> Therefore, in the present case, surgical drainage was performed, and infection was controlled using oral antibiotics, resulting in clinical improvement observed one week after surgery. Our findings highlight that both surgical intervention and the implementation of effective infection control are important strategies when managing mucocoeles associated with optic neuropathy.

## Conclusion

Although paranasal sinus mucocoeles are indeed a rare cause of optic neuropathy, it is crucial to recognize them as a potential cause since they may be treatable. The management and diagnosis of paranasal sinus mucocoeles necessitate a multidisciplinary approach, and surgical intervention along with effective infection control are key strategies in their treatment.

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## Authors' Contribution

**Conceptualization:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Data curation:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Formal analysis:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Investigation:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Methodology:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Project administration:** Witoon Mitarnun.

**Resources:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Software:** Witoon Mitarnun.

**Supervision:** Witoon Mitarnun.

**Validation:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Visualization:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

**Writing—original draft:** Witoon Mitarnun, Korrapat Aegakkatajit,

Nutdanai Roila, Nithinan Uepechpong.

**Writing—review & editing:** Witoon Mitarnun, Korrapat Aegakkatajit, Nutdanai Roila, Nithinan Uepechpong.

## Competing Interests

No conflict of interest in this study.

## Ethical Approval

The ethics committee of Buriram Hospital approved this study (IRB: BR 0033.102.1/23), and the patient provided informed consent.

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