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#### **ABOUT COVER**

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#### **AIMS AND SCOPE**

The primary aim of World Journal of Clinical Cases (WJCC, World J Clin Cases) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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CASE REPORT

# Subperiosteal orbital hematoma concomitant with abscess in a patient with sinusitis: A case report

Xi-Hao Hu, Chi Zhang, Yu-Ke Dong, Tie-Chuan Cong

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## Abstract

#### BACKGROUND

Sinusitis-derived subperiosteal orbital collection is uncommon and is usually diagnosed as subperiosteal orbital abscess or, rarely, as hematoma. We report a unique and even rarer case of subperiosteal orbital collection, which is actually a complication of hematoma and abscess.

#### CASE SUMMARY

A 26-year-old female presented with left eyeball pain and ipsilateral chemosis. She had no history of head trauma or upper respiratory infection. Her blood cell count showed an increase in leukocytes. Fiberoptic rhinolaryngoscopy revealed only mucosal edema of the left olfactory crest without apparent discharge. The computed tomography scan results showed an opaque left posterior ethmoid cell with a thickened bony shell and fusiform changes in the periosteal elevation of the medial wall of the left orbit. Emergent surgery revealed an ethmoid mucocele complicated with subperiosteal orbital hematoma and abscess. The pathology of the lamina papyracea between the mucocele and subperiosteal collection was necrotic, and the overlying mucosa was de-epithelialized.

#### **CONCLUSION**

Subperiosteal orbital hematoma with abscess in a patient with sinusitis adds to the current knowledge of orbital complications of sinusitis.

Key Words: Subperiosteal orbital hematoma; Abscess; Sinusitis; Mucocele; Surgery; Case report

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**Core Tip:** As a differential diagnosis of subperiosteal orbital collection, subperiosteal orbital hematoma concomitant with abscess needs to be taken into consideration. Surgery is advocated for the diagnosis and treatment of such pathology. This report adds to current knowledge regarding the orbital complications of sinusitis.

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#### INTRODUCTION

Subperiosteal orbital collection caused by sinus infection is usually diagnosed with subperiosteal orbital abscess (SPOA). In recent years, another diagnostic entity of subperiosteal orbital hematoma that shows clinical manifestations similar to those of SPOA has been sporadically reported in patients with sinusitis or sinus mucocele, and it is believed that the disease could be misdiagnosed as an SPOA if only medically treated [1,2]. Additionally, subperiosteal orbital hematoma mixed with abscess is logically possible when infection spreads to the subperiosteal hematoma. Here, we report a rare condition that was identified in a patient with ethmoid mucocele and subperiosteal orbital collection.

#### **CASE PRESENTATION**

#### Chief complaints

A 26-year-old female presented with left eyeball pain for 2 d (the timeline is shown in Table 1).

#### History of present illness

The patient had a left-sided headache two days before the eyeball pain. She was afebrile and did not display diplopia, visual loss, or any discomfort related to her nose.

#### History of past illness

The patient was otherwise healthy and had no history of head trauma, upper respiratory infection, vascular disease, or coagulation disorder.

#### Personal and family history

The patient had no relevant personal or family history.

#### Physical examination

Ophthalmologic examination showed periorbital edema and conjunctival chemosis in the left eye. Ocular movements of the same eye were normal in all directions. Funduscopic examination yielded results within the normal range. Fiberoptic rhinolaryngoscopy revealed congestion of the left olfactory mucosa without purulent discharge.

#### Laboratory examinations

The patient's white blood cell count was 10400/mm<sup>3</sup>, and her C-reactive protein level was within the normal range.

#### Imaging examinations

Computed tomography with a soft window showed an opaque left posterior ethmoid cell with a thickened bony shell (Figure 1). In contrast to the cell, the orbital periosteum of the lamina papyracea exhibited fusiform changes with isodensity. The lamina papyracea per se showed no sign of erosion. The medial rectus was oppressed and shifted outwards (Figure 1B).

#### **FINAL DIAGNOSIS**

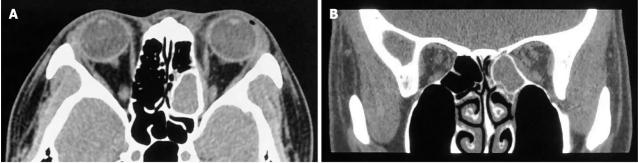
The final diagnosis of the case was infection of the ethmoid mucocele complicated with subperiosteal orbital collection.



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Hu XH et al. Subperiosteal orbital hematoma concomitant with abscess

Table 1 Timeline of events				
Event	Timeline			
Presented with left eyeball pain	-2 d			
Computed tomography showed an opaque left posterior ethmoid cell with thickened bony shell	-1 d			
Admitted to our department and received endoscopic endonasal sinus surgery	0 d			
Free of symptoms	1 d			
Patient was discharged	5 d			
Surgical cavity was completely epithelialized	2 mo			



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Figure 1 Axial and coronal noncontrast computed tomography scans with soft windows showing an opacified left posterior ethmoidal cell with a thickened bony shell. Across the lamina papyracea was a lesion with a fusiform change and isodensity. A: Axial plane; B: Coronal plane.

#### TREATMENT

Ceftriaxone was administered intravenously followed by endoscopic endonasal sinus surgery. Mucocele was ascertained, and purulent discharge with some caseous material was aspirated after marsupialization of the opacified posterior ethmoid sinus (Figure 2). The lamina papyracea exposed in the mucocele cavity, surprisingly, showed an altered blue and white area in which the mucosal lining was devascularized (Figure 2A and B). Medial orbital wall decompression was then performed, and a dark blood clot surrounded and even partially covered by pus appeared inside the exposed subperiosteal space, causing the papyracea to take on the specific blue and white hue. The clot appeared to be partially lysed, and it was easily shaped into threads when aspirated (Figure 2C). During the process of decompression, pus mixed with blood subsequently oozed into the ethmoid cavity from around the blood clot (Figure 2D). Complete drainage of the collection was ensured by applying external pressure to the orbit.

#### OUTCOME AND FOLLOW-UP

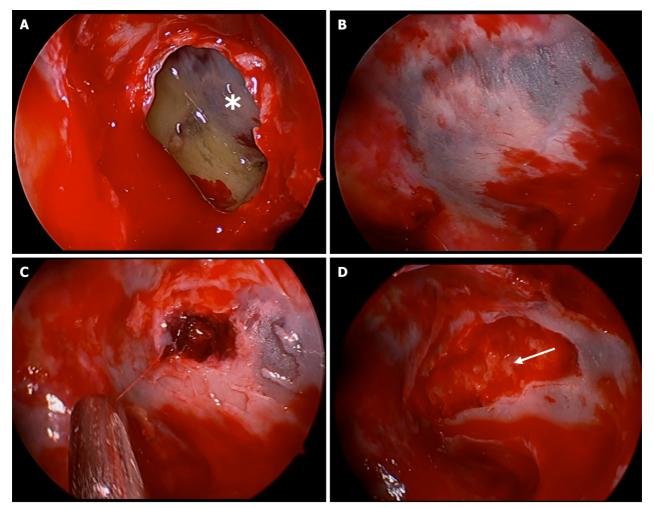
The patient was free of symptoms within 24 h after surgery. Magnetic resonance imaging on postoperative day 5 showed no sign of liquid accumulation in the medial wall of the left orbit (Figure 3A), and the patient was discharged on the same day. Histopathological examination demonstrated de-epithelialized mucosa with bone necrosis of the lamina papyracea. No microorganisms grew from either the subperiosteal orbital or ethmoidal discharges. At two-month follow-up, the surgical cavity was completely epithelialized, and the patient recovered fully (Figure 3B and C).

#### DISCUSSION

Orbital complications of sinusitis, according to Chandler's modified classification, are divided into five groups: preseptal cellulitis, orbital cellulitis, subperiosteal abscess, orbital abscess, and cavernous sinus thrombosis. The most common sinogenic orbital complications appear to originate in the ethmoid sinuses[3-5] and are mainly related to rhinosinusitis or, uncommonly, a mucocele. If not treated in a timely manner, mucocele of the paranasal sinus can induce cranial and/or orbital complications. In



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Figure 2 Endoscopic views of the mucocele and the subperiosteal orbital collection. A: Marsupialization of the mucocele of the left posterior ethmoid sinus. An asterisk indicates the lamina papyracea; B: The lamina papyracea is covered with thin devitalized mucosa and has a blue and white appearance with a geographic pattern. The surrounding mucosa rapidly became hyperemic after the cavity pressure was eliminated; C: A blood clot was uncovered under the blue area of the lamina papyracea, and the periorbita was subsequently exposed; D: Pus (white arrow) oozed from around the blood clot.

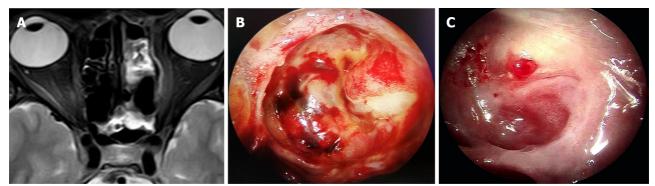
> addition to direct expansion and compression to the orbit, orbital complications of complicated mucocele can also be caused by infection directly through congenital or acquired bony dehiscence or via the neurovascular foramina or through the local valveless veins of the thin lamina papyracea resulting from thrombophlebitic or embolic conditions[6]. Subperiosteal abscess develops between the orbital bone and periorbita, and up to 90% of SPOA cases are caused by complicated sinusitis[7]. The diagnosis of subperiosteal abscess is mainly based on clinical manifestation and radiographic imaging and is generally confirmed by surgical drainage.

> SPOA, however, is only one form of subperiosteal orbital fluid collection that can also present as subperiosteal orbital hematoma. Subperiosteal orbital hematoma is mostly caused by trauma. Nontraumatic subperiosteal orbital hematoma, mostly associated with increased intracranial pressure and bleeding disorders that cause rupture and bleeding of subperiosteal venules[8], is occasionally described in the literature. Sinus infection-related subperiosteal hematoma occurs even more rarely. To date, just over 20 cases of subperiosteal hematoma associated with sinusitis have been surgically identified in the literature, including 4 cases of sinus mucocele[1,2,8,9]. Some authors believed subperiosteal hematoma was an unusual complication of sinusitis. In patients with subperiosteal hematoma associated with sinusitis, no pus was detected, and the results of the hematoma culture were mainly negative.

> To the best of our knowledge, this is the first report of subperiosteal orbital hematoma concomitant with abscess occurring in the same potential anatomical space in an adult patient with complicated sinus infection. We postulate that the pathogenesis of this condition probably involves the rupture of small veins crossing between the orbital bony wall and the periorbita due to adjacent sinus inflammation and the subsequent formation of hematoma, the early spread of pathogenic microorganisms directly through the lamina papyracea to involve the subperiosteal orbital space, and the formation of a localized abscess surrounding the blood clot prior to its complete lysis. Concomitant hematoma and



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Figure 3 Postoperative magnetic resonance imaging and follow-up with endoscopy. A: T2 magnetic resonance imaging scan on the 5th postoperative day shows hyperintensity of the involved medial periorbita of the left orbit on T2-weighted imaging, revealing sufficient drainage of the subperiosteal orbital abscess; B: Endoscopic examination on the 11th postoperative day shows a slightly bulging periorbita that was apparently healthy; C: Endoscopic reevaluation two months after the operation revealed nearly complete epithelization of the lamina papyracea with a small cherry granuloma.

> abscess in the same area are probably transitory conditions and show the process through which the hematoma evolves completely into an abscess. In our case, the de-epithelialized mucosa of the mucocele and the necrotic change in the lamina papyracea involved might indicate a role of direct extension in the development of a subperiosteal abscess.

> Several studies have shown that the age of the patient is an independent risk factor with respect to the choice of treatment for SPOA. Harris found that for patients less than 9 years old without visual impairment, emergent or even urgent surgery was not mandatory as the first-line treatment. Conversely, these patients could be initially administered medical treatment concomitantly with watchful observation. However, in cases of mixed infection with aerobes and anaerobes, which are common in patients more than 9 years old, operative intervention should be initially performed[10,11]. For adult patients with SPOA, endoscopic endonasal sinus surgery is currently advocated as the first choice of treatment due to its advantages of minimal invasion, no facial scarring, and shortened hospital course compared with the external orbital approach. Ethmoidectomy usually suffices when obvious dehiscence of the lamina papyracea is apparent[12]. If dehiscence is not detected during the surgery, the lamina papyracea should be opened for drainage. However, there is controversy regarding the extent of removal of the lamina papyracea. In the early days of endoscopic sinus surgical practice, the lamina papyracea was cracked but not removed[12], or swang back and then medialized in a "swing door" pattern[13], or partially resected[14]. Recently, wide removal of the lamina papyracea with complete ethmoidectomy was performed with satisfactory results[15]. The rationale for wide resection was supported by a report by Ragab et al[6]. In their report, bone sequestrum was identified in all eight cases with the presence of cocci in half; this explained the failure of medical treatment and the necessity for surgery.

> In the case reported here, sequestrum was observed histologically with no pathogens identified, in concordance with the cases reported by Eviatar et al[5], who observed bone destruction and resorption in the lamina papyracea in five of nine children with acute rhinosinusitis and SPOA. These pathological findings strengthen the theory of direct spread for the pathogenesis of SPOA.

> As an approach to diagnosing and treating subperiosteal abscess in patients with sinus mucocele, surgical intervention is advocated, consistent with the treatment modality recommended for cases of subperiosteal orbital hematoma caused by sinus infection. The recovery from subperiosteal hematoma and abscess in the case reported here was uneventful after the surgery.

#### CONCLUSION

Subperiosteal orbital hematoma concomitant with abscess in patients with sinusitis has not been reported previously. It can cause necrotic bony changes, indicating a need for timely surgical intervention.

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#### FOOTNOTES

Author contributions: Cong TC was the patient's surgeon and drafted the manuscript; Hu XH, Zhang C, and Dong YK attributed to the data collection and critically revised the manuscript; all authors read and approved the final manuscript.

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