



A case report of unrecovered monocular vision loss after anterior clinoid mucocele resection

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ABSTRACT

Purpose: To describe a patient of anterior clinoid mucocele presenting with orbital apex syndrome and to propose the reason for her unrecovered monocular vision loss after operation, we hope this case report can provide treatment experience for the similar rare disease.

Conclusions and Importance: Mucocele of the anterior clinoid process remains a rare cause of optic neuropathy. They usually present with acute, rapidly progressive, painless monocular visual loss. Urgent surgical decompression play a vital role in the recovery of neurological function.

Observations: A 64-year-old woman presented with sudden painless loss of vision in her left eye for about 36 days. Ophthalmologic exam at admission revealed normal in the right eye and no perception of light in the left eye. There was an afferent pupillary defect on the left. Imaging data revealed an expansile soft-tissue mass in the left anterior clinoid process with compression of the optic canal. Then optic canal decompression and histopathological examination were conducted. Histopathological examination confirmed the diagnosis of anterior clinoid mucocele. No bacteria or other microorganisms were identified in microbiological examination. Cytologic examination showed no evidence of neoplasia. At 6 months follow-up, the patient had no improvement of vision in the left eye and there was no evidence of residual or recurrence of the anterior clinoid mucocele.

1. Introduction

Pneumatization of the anterior clinoid process (ACP) occurrence with a frequency of approximately 9.5%. Anterior clinoid mucoceles usually develop when the ostium of a pneumatized ACP becomes obstructed as a result of mucosal thickening, fibrosis or bony overgrowth. Mucocele of the ACP can result in compression on the adjacent structures in the optic nerve canal or superior orbital fissure, causing optic neuropathy (compressive or inflammatory) or ophthalmoplegia with dysfunction of third, fourth and/or sixth cranial nerves ACP.

Mucoceles in the anterior clinoid location are exceptionally rare, the cases that cause symptoms such as optic neuropathy or ophthalmoplegia are even more rare.¹⁻⁴ So far, no more than 20 cases have been reported.^{5,6}

1.1. Case report

A 64-year-old woman with a history of diabetes and hypertension presented with sudden painless loss of vision in her left eye, it was about 36 days when she admitted for examination after onset, with no prior history of sinus disease or surgery. Because we have not been exposed to this kind of disease before, the patient was not diagnosed definitely before the operation. 3 days after hospitalization, she underwent optic canal decompression via left lateral supraorbital approach. A cystic lesion containing yellow viscous liquid was excised. The cystic wall was taken for pathological examination, and the cloudy cyst fluid was taken for cytologic examination and microbiological examination. There were no other cranial neuropathies. Clinical history revealed hypertension and diabetes mellitus for 2 years.

Ophthalmological examinations: Ophthalmologic exam at admission revealed normal in the right eye and no perception of light in the left eye. There was an afferent pupillary defect on the left. Extraocular movements were full with no nystagmus or ophthalmoplegia.

Imaging examination: Computerized tomography (CT) scan and

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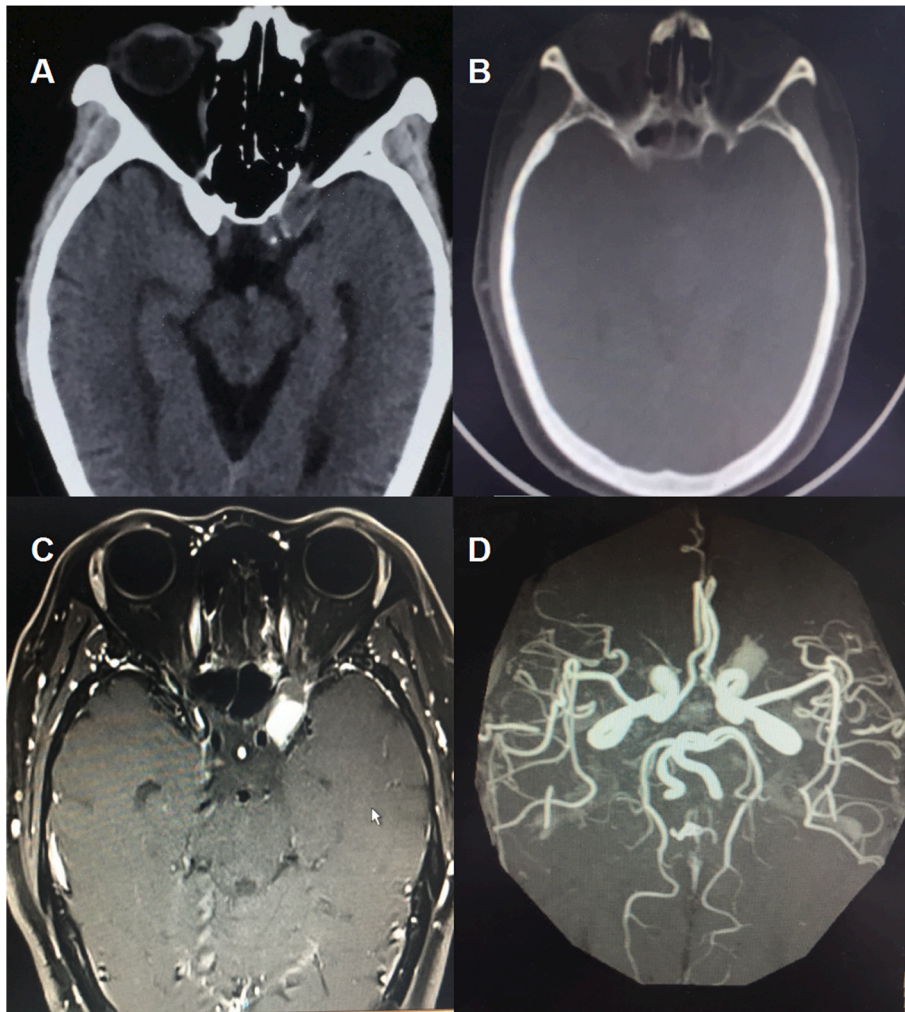


Fig. 1. CT scan revealed an expansile soft-tissuemass in the left anterior clinoid, and the bony margin of the lesion appeared to be eroded and partially absent (A B). MRI shows nodular enhanced tissuemass in the left ACP with compression of the optic canal (C). MRA shows high density mass in the left anterior clinoid, no obvious arteries (D).

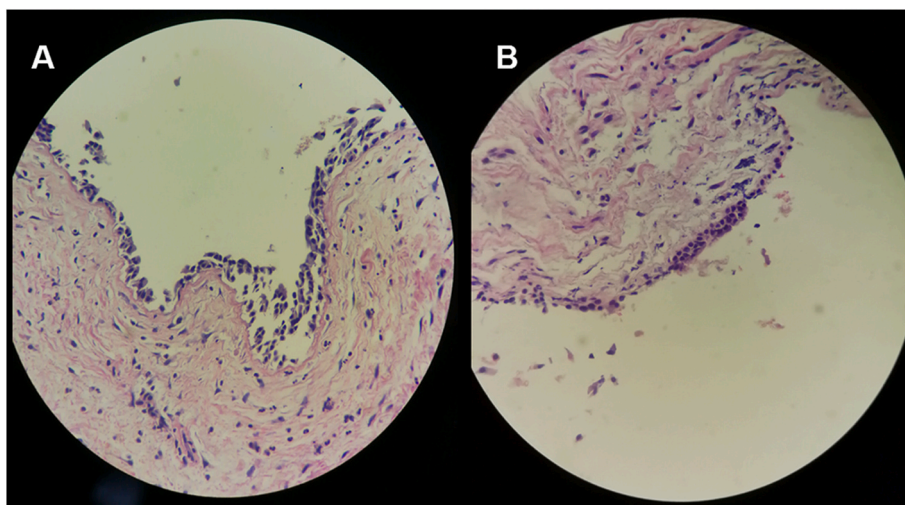


Fig. 2. Histopathological examination showed the lesion had a fibrous wall with ciliated columnar epithelium, it was consistent with a mucocele, a diagnosis of anterior clinoid mucocele was made.

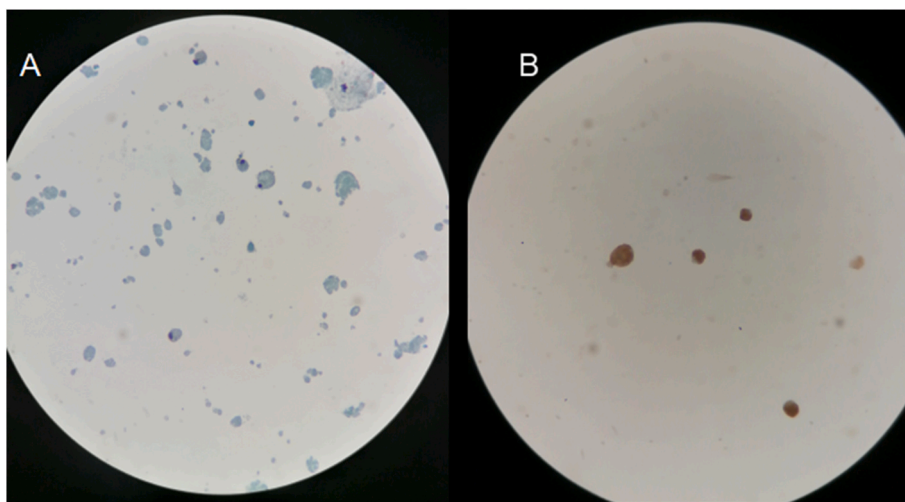


Fig. 3. Cytologic examination showed no evidence of neoplasia.

magnetic resonance imaging (MRI) of the brain revealed an expansile soft-tissue mass in the left ACP with compression of the optic canal. On the bone windows of the CT, the bony margin of the lesion appeared to be eroded and partially absent. In addition, the contralateral ACP was also well pneumatized (Fig. 1).

Histopathological and Laboratory examinations: Histopathological examination showed the lesion had a fibrous wall with ciliated columnar epithelium, it was consistent with a mucocele, a diagnosis of anterior clinoid mucocele was made (Fig. 2). No bacteria or other microorganisms were identified in microbiological examination. Cytologic examination showed no evidence of neoplasia (Fig. 3).

Outcome and follow-up: At 6 months follow-up, the patient had no improvement of vision in the left eye and there was no evidence of residual or recurrence of the anterior clinoid mucocele on CT and MRI of the brain.

2. Discussion

Anterior clinoid mucocèles are very rare, accounting for only 1% of paranasal sinus mucocèles. Pneumatization of the ACP is a normal anatomical variant in sphenoid sinus development. Obstruction of the sinus ostium has been considered as a result of mucosal thickening, fibrosis, bony overgrowth or tumors causing retention of mucous with erosion and remodeling of the adjacent bones.¹ This can make them susceptible to obstruction and secondary inflammation or infection which can lead to the development of mucocèle.^{7,8}

The expanded mucocele can cause symptoms of oppression, such as the symptoms including retro orbital pain, blurring of vision, monocular visual loss, with variable degree of visual field defect, diplopia due to compression 3rd, 4th or 6th cranial nerves, and sensory deficits of trigeminal nerve.^{6,9,10} Symptoms can be due to direct compression on the cranial nerves causing ischemia and dysfunction, or possibly due to extension of the inflammatory process from the mucocele to the nerve fibers causing neuritis.⁶

Johnson et al. found that delay of more than 7 days from the beginning of visual deficiencies to surgical decompression can cause irreversible damage to the optic nerve.¹¹ The latest articles also take a similar view that the good prognosis of anterior clinoid mucocele is related to early diagnosis and early decompression.^{5,6,12,13}

The visual dysfunction caused by mucocele seldom return to normal when there is a significant delay in treatment. In our case, the patient's clinical symptoms did not improve significantly after surgery because of late detection. The progressive deterioration of the patient's vision led to diagnostic difficulties, which also provided an a diagnostic approach to ophthalmologist. In addition, patients with diabetes may affect the

patient's prognosis, which maybe related to diabetic eye complications, but the patient has no history of diabetes, we can not further confirm. After the operation, we treated the patient with Mecobalamin and other neurotrophic drugs. Six months later, the patient showed no obvious improvement, accompanied by optic atrophy. This atrophy of the optic nerve would be expected to accompany her unrecovered vision. Unfortunately, we were not observe the optic nerve before surgery. So far, the mechanism of vision loss caused by this disease has not been clear. We speculate that its mechanism may be consistent with optic nerve damage caused by trauma. Previous studies have confirmed that mucous cysts increase in optic nerve injury caused by trauma, and the vascular and hemodynamic disturbances of the central retinal artery play a vital role in the pathological process. Animal experiments have confirmed that inflammation, lipid peroxidation, bradykinin release, cell apoptosis and axon demyelination aggravate the initial damage, further damage the axons of retinal ganglion cells, leading to vision loss.¹⁴⁻¹⁶

Rapidly deteriorating vision is a bad prognostic sign, and urgent imaging and management should be undertaken immediately, in our report, the unrecovered monocular vision loss maybe the delayed examination and surgery.

3. Conclusion

Mucocele of the ACP remains a rare cause of optic neuropathy. They usually present with acute orbital apex syndrome. Urgent surgical decompression play a vital role in the recovery of neurological function.

3.1. Patient consent

Consent to publish this case report has been obtained from the patient in writing.

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Author declaration form

We have been attach the copy of author declaration form signed as e-Component.

Declaration of competing interest

The following authors have no financial disclosures: Shizhong Zhang, Zhen Li, Hong Ye, Han Zhao, Qiong Wu, Xiubao Zhang, Guojun

Wang.

Authorship: All authors attest that they meet the current ICMJE criteria for Authorship.

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