CASE REPORT

Orbital mycobacterial infection: a case report

Christina KY Ip, MBChB; Cheong-Yi Fong, MBChB, MRCSEd (Ophth), FCOphth (HK), FHKAM (Ophth); Pui-Fan Lam, MBChB, MPH, FCOphth (HK), FHKAM (Ophth); Shu-Kwan Ip, MBBS, FRCS (Glasg) (Ophth), FCOphth (HK), FHKAM (Ophth)
Department of Ophthalmology, Tuen Mun Eye Centre, Hong Kong

Correspondence and reprint requests:
Dr Christina KY Ip, Department of Ophthalmology, Tuen Mun Eye Centre, Hong Kong. Email: christinaip55@gmail.com

Abstract

A 69-year-old Chinese man presented with proptosis was found to suffer from orbital mycobacterial infection. As acid-fast bacilli were identified from orbital excisional biopsy, the patient was treated with an anti-tuberculous regimen. He responded well and was asymptomatic at 13-month follow-up. This case is documented for its rarity.

Key words: Orbit; Tuberculosis, ocular

Case presentation

A 69-year-old Chinese man presented to Tuen Mun Eye Centre with a 1-month history of right eye proptosis and pain in June 2020. He was a non-smoker with a past medical history of Type 2 diabetes mellitus, hyperlipidemia, and complete heart block with permanent pacemaker implanted. There was no history of ocular trauma or surgery.

The best-corrected visual acuity was Snellen 0.13 OD. Pupils were equal and reactive to light, there were no relative afferent pupillary defect. Exophthalmometer showed right non-axial proptosis by 2 mm. Intraocular pressure (IOP) of both eyes were normal. Anterior segment examination showed right eye mild chemosis. Extraocular movement was full with no diplopia. Fundal examination revealed mild non-proliferative diabetic retinopathy of both eyes and a macula hole over his right eye. There were no signs of optic neuropathy. Blood tests were unremarkable except glycated hemoglobin level of 9.9%. Orbital computed tomography (CT) was arranged.

A follow-up IOP measurement at 4 weeks after initial presentation revealed an elevated IOP of 30 mmHg. There were also lagophthalmos, hypoglobus, and restricted motility over his right eye with markedly reduced elevation and abduction. Anterior segment examination showed exposure keratopathy. Fundus examination findings were similar to those on initial presentation. Imaging with orbital CT revealed a 2.4 cm right orbital post septal rim enhancing mass (**Figure 1**).

The working diagnosis was orbital abscess, right eye lateral canthotomy and drainage yielded yellowish necrotic materials with minimal fluid pus. Histological examination revealed lymphoid rich inflammatory lesions with necrotic features, no obvious malignant cells were identified (**Figure 2**). Gram stain and culture were negative. Postoperatively, intravenous broad-spectrum antibiotics were given but right eye proptosis and exposure keratopathy persisted.

A follow-up CT scan at postoperative 1-week revealed no interval reduction in the size of the lesion. The lesion was excised subsequently with lateral orbitotomy. Proptosis resolved and exposure keratopathy improved. IOP and extraocular movement returned to normal. The excised lesion was 1.8 cm in diameter and well circumscribed (**Figure 3**). The lacrimal gland was left intact. The features of necrotizing inflammation in histopathological analysis and the scanty acid-fast bacilli (AFB) on Ziehl-Neelsen stain (**Figure 4**) were suggestive of mycobacterial infection. Polymerase chain reaction for *Mycobacterium tuberculosis* complex was negative, as were periodic acid-Schiff, Grocott methenamine silver, and mucicarmine stains for other organisms.

On retrospective review, the patient had no history or contact of pulmonary tuberculosis (TB). He was born in Mainland

© 2022 Hong Kong Journal of Ophthalmology. CC BY-NC-ND 4.0

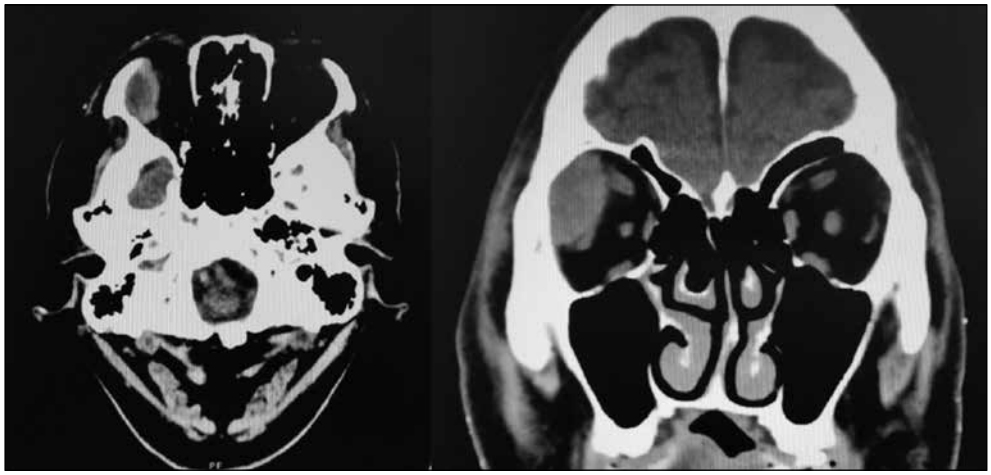


Figure 1. Computed tomography of the orbit showing a 2.4 cm rim-enhancing mass located at the extraconal region of the right orbit in the post-septa region displacing the lateral rectus muscle inferiorly. The mass is abutting on the superior and lateral orbital wall, lacrimal gland, and right globe, causing proptosis. The right lacrimal gland cannot be delineated from this lesion. There is no bony erosion, thickening, or lysis.

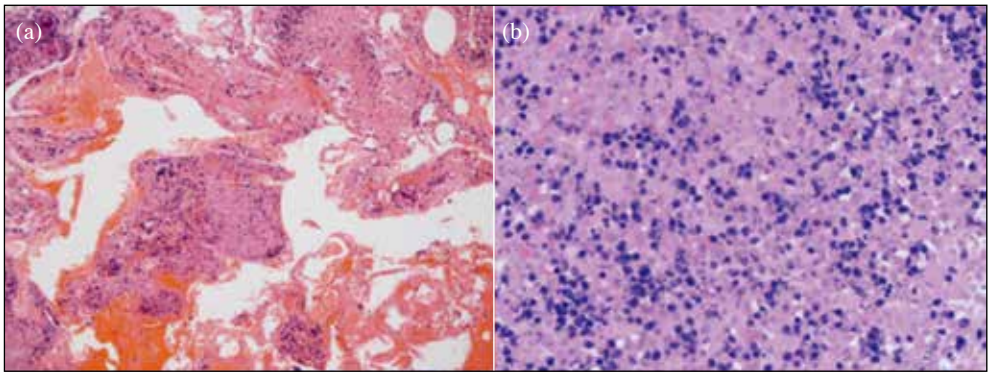


Figure 2. Histopathology showing (a) fibrinous tissue with atypical necrosis (100×). (b) small lymphocytic inflammatory cell infiltrate (400×).



Figure 3. Clinical photograph showing the well circumscribed excised lesion.

China and did not receive Bacillus Calmette-Guérin vaccination. He did not have any signs and symptoms of systemic TB infection; chest X-ray was clear with no features of pulmonary TB. Sputum and blood AFB culture were negative, as was HIV antigen.

The patient was referred to a local chest clinic as microbiologist suggested. He was then treated as extrapulmonary TB with an anti-TB regimen consisting of isoniazid 300 mg daily, rifampicin 600 mg daily, pyrazinamide 1500 mg daily, and levofloxacin 500 mg daily.

Further CT scan at postoperative 1-month revealed no recurrence of proptosis. IOP and extraocular movement remained normal. Follow-up at 13-month showed no clinical evidence of recurrence (**Figure 5**). The patient completed 16-month anti-TB regimen. The lesion resolved completely with no further complications in May 2022. Regular follow-up has been scheduled.

Discussion

Orbital mycobacterial infection is rare. Most of the existing literatures regarding orbital tuberculosis (OTB) and non-tuberculous mycobacterial (NTM) orbital infection are limited to case reports or case series. There were 79 identified cases of OTB in an extensive literature search.[1] In a systematic review, only 11 (2.6%) of 420 cases of NTM ocular infections were orbital infections.[2]

Orbital involvement of TB may be a result of direct spread from paranasal sinuses, or hematogenous spread from a primary source, with or without pulmonary TB.[1,3] Therefore,

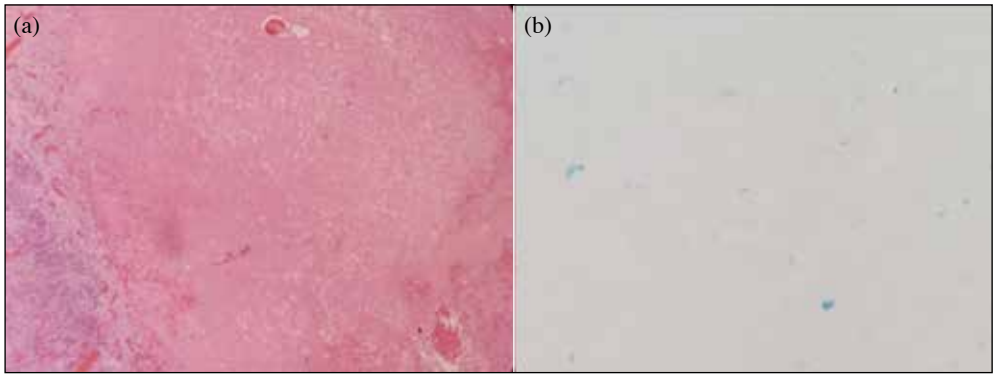


Figure 4. Histopathology showing (a) fibrous stromal tissue with necrotic center rimmed by histiocytes, fibroblastic proliferation and crushed reactive lymphoid infiltrate (100×). (b) Scanty acid-fast bacilli on Ziehl-Neelsen stain (400×).



Figure 5. Clinical photographs showing (a) right eye chemosis and proptosis before lateral orbitotomy. (b) no recurrence of right eye chemosis and proptosis after lateral orbitotomy at 13-month follow-up.

it cannot be excluded in the absence of pulmonary infection. In fact, a case of OTB without primary source identified was reported.[4] Extrapulmonary TB has become more common in immunocompromised patients.[1] As OTB is a rare form of extrapulmonary TB, the association between immunocompromised condition and OTB is uncertain. Increased risk of OTB in immunocompromised patients secondary to poor control of diabetes mellitus is

inconclusive. Our patient did not receive Bacillus Calmette-Guérin vaccination and may be at an increased risk of TB infection, but he had no known TB history or contact.

It was found that orbital fat allowed NTM to escape from the host immune system by protecting them from enzymatic action or phagocytosis,[5] thus predisposing NTM orbital infection. Also, NTM orbital infection is associated with risk factors such as prior corticosteroid therapy, history of interventions or recent accidental ocular trauma.[6] Our patient did not have any recent interventions or ocular trauma or any signs of injury and inflammation, the origin of infection could not be ascertained.

Manifestations of orbital mycobacterial infection is non-specific. Patient may present with an intraconal mass that leads to proptosis and restricted motility, or orbital inflammation such as draining sinus tracts, periorbital cellulitis, and orbital abscess. Extradural abscess formation, soft tissue tuberculoma, caseating granuloma, and orbital apex syndrome can also occur.[2,7] Destructive complications to the bone and soft tissue may be resulted.[8] The wide variety of possible presentations of orbital mycobacterial infection pose a challenge to a straightforward diagnosis.

It is difficult to establish and confirm the diagnosis of orbital mycobacterial infection. Diagnosis is challenging due to its rarity, non-specific manifestations, difficulty in obtaining orbital specimen for histological and microbiological analysis, and low yield from orbital specimen.[8] Extrapulmonary TB infection is often paucibacillary, and false negative may occur. As reported, only 19 of 79 identified cases of OTB yielded positive cultures.[1] In our patient, the delay in diagnosis was accounted for by the absence of systemic symptoms and predisposing risk factors. In addition, the histology of the necrotic materials obtained from lateral canthotomy and drainage was unrevealing. It was the identification of scanty AFB from the orbital excisional biopsy shed light on the diagnosis and the choice of treatment.

There are no specific guidelines for the treatment of orbital mycobacterial infection. Possible treatment options include surgical means such as drainage or excision of lesion, and medical treatment with antibiotics. Surgical excision of lesions can be diagnostic and therapeutic. Medications for OTB are individualized,[8] but mainly comprising multiple anti-TB drugs with duration ranged from 6 to 18 months.[1] For NTM infections, the choice of antibiotics depends on drug sensitivity of the causative agent. In general, slow-growers are sensitive to those for anti-TB, whereas rapid-growers are sensitive to macrolides, fluoroquinolones, and aminoglycosides.[9] Complete surgical excision with anti-TB therapy can be the treatment of choice to achieve resolution of orbital mycobacterial infection as illustrated in this case.

Conclusion

We report a rare case of orbital mycobacterial infection with no primary source identified. Our case demonstrates the possibility of orbital mycobacterial infection in the absence of apparent risk factors and the importance of obtaining appropriate specimen for diagnostic investigations. Ophthalmologists should be aware of the different manifestations of orbital mycobacterial infection and be alert to the possibility of mycobacterial infection.

Contributors

All authors designed the study, acquired the data, analyzed the data, drafted the manuscript, and critically revised the manuscript for important intellectual content. All authors had full access to the data, contributed to the study, approved the final version for publication, and take responsibility for its accuracy and integrity.

Conflicts of interest

All authors have disclosed no conflicts of interest.

Funding/support

This study received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Data availability

All data generated or analyzed during the present study are available from the corresponding author on reasonable request.

Ethics approval

The patient was treated in accordance with the tenets of the Declaration of Helsinki. The patient provided written informed consent for all treatments and procedures and for publication.

References

1. *Madge SN, Prabhakaran VC, Shome D, Kim U, Honavar S, Selva D. Orbital tuberculosis: a review of the literature. Orbit 2008;27:267-77.* Crossref
2. *Kheir WJ, Sheheitli H, Abdul Fattah M, Hamam RN. Nontuberculous mycobacterial ocular infections: a systematic review of the literature. Biomed Res Int 2015;2015:164989.* Crossref
3. *Pillai S, Malone TJ, Abad JC. Orbital tuberculosis.*

Ophthalmic Plast Reconstr Surg 1995;11:27-31. Crossref
4. *Aggarwal D, Suri A, Mahapatra AK. Orbital tuberculosis with abscess. J Neuroophthalmol 2002;22:208-10*. Crossref
5. *Gonzalez-Fernandez F, Kaltreider SA. Orbital lipogranulomatous inflammation harboring* Mycobacterium abscessus. *Ophthalmic Plast Reconstr Surg 2001;17:374-80*. Crossref
6. *Klapper SR, Patrinely JR, Kaplan SL, Font RL. Atypical mycobacterial infection of the orbit. Ophthalmology 1995;102:1536-41*. Crossref
7. *Dalvin LA, Smith WM. Orbital and external ocular manifestations of* Mycobacterium tuberculosis*: a review of the literature. J Clin Tuberc Other Mycobact Dis 2015;4:50-7*. Crossref
8. *Yao A, Aboltins C, McNab AA, Rosa CS, Denholm J, Khong JJ. Orbital tuberculosis: perspectives from Victoria, Australia. Graefes Arch Clin Exp Ophthalmol 2018;256:2443-8*. Crossref
9. *Moorthy RS, Valluri S, Rao NA. Nontuberculous mycobacterial ocular and adnexal infections. Surv Ophthalmol 2012;57:202-35*. Crossref