CASE REPORT

Primary orbital chronic granulomatous reaction to deep staphylococcal infection due to trauma in immunocompetent

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SUMMARY

A 53-year-old man presented with a slow-growing mass in the right orbit and temporal area of 1 year duration following a road traffic accident. Examination revealed a nodular fungating mass involving the orbit, right temporal area with multiple discharging sinuses causing proptosis and restricted ocular motility. The patient denied light perception in the right eye. There was exposure keratopathy and optic atrophy. Imaging showed mass lesion of the orbit extending intracranially. Tissue biopsy and culture revealed a chronic granulomatous reaction due to deep infection with coagulase-positive staphylococci. The patient showed remarkable improvement to systemic antibiotics and anticonvulsants. Mortality is high in such cases due to intracranial extension if there is a delay in treatment. This case report summarises a rare aetiology and successful treatment of a case of chronic granulomatous reaction due to deep bacterial infection following trauma and is deemed rare as cases of primary orbital infection in immunocompetent have not been reported so far.

BACKGROUND

'Botryomycosis' arises from chronic infections produced by low-virulence organisms in an altered host environment. The term was coined by Rivolta in 1870 and refers to the grape-like granules (Gr. botryo = grapes) and the erroneously presumed fungal aetiology (Gr. mykes = fungus).¹ Staphylococci have also been implicated causing tumefaction, deformity, sinus formation, fistulous tracts, deep abscesses and ulcerations. It can be confused with other mycetomas or fungal infections. Hence, this term botryomycosis is no longer used for this condition as it is a pseudomycetoma and not a true fungal infection as the term suggests. The diagnosis can only be established by tissue culture and biopsy.² There is no conclusive evidence regarding the type or duration of treatment which makes the management of this condition even more difficult. This case report shows how tissue culture and biopsy aided in the diagnosis and successful treatment of this rare case.

CASE PRESENTATION

A 53-year-old man presented with ab axial proptosis of 1 year duration which gradually progressed to involve the lids and the right temporal area. Nine months later he developed redness, chemosis and gradual painful loss of vision in the right eye. The patient gave history of an extensive laceration

over the right temporal area 3 years ago due to a road traffic accident for which he underwent debridement and repair. He was asymptomatic for a year. Patient was moderately built and nourished with no premorbid illness. On examination, patient denied perception of light in the right eye, left eye vision was 6/6, N6. There was ab axial proptosis, mechanical ptosis, multiple nodules and discharging sinuses over the lids, restriction of ocular motility with 45° of exotropia in the right eye. Extraocular movements in the left eye were normal. Slit lamp examination of the right eye revealed congestion and chemosis of conjunctiva and exposure keratopathy. Anterior chamber was quiet. There was afferent pupillary defect in the

(a)

(b)

Figure 1 (A) Before the start of the treatment: lesion of the right orbit and frontotemporal region showing tumefaction with multiple nodules and discharging sinuses, deep abscess and ulceration. (B) Lateral view showing ab axial proptosis, bullous chemosis with multiple discharging sinuses and nodules.

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Figure 2 (A) Axial section of the inferior orbit showing a soft tissue lesion of the orbit extending into the right maxillary sinus and extracalvareal soft tissues. (B) MRI: coronal section of the face showing soft tissue lesion involving the right orbit and temperoparietal soft tissues extending into the right maxilla and subdural area causing localised meningeal enhancement. (C) MRI: showing soft tissue mass over the frontotemporal bone with extensive intracranial involvement and midline shift.

right eye. Fundus examination showed pale disc suggestive of optic atrophy. There was a nodular fungating mass over the right temporal area also with multiple draining sinuses (figure 1A,B). Left eye examination was unremarkable. Intraocular pressure in both eyes was normal. Based on these findings a diagnosis of inflammatory proptosis was made. Systemic examination did not reveal anything significant.

INVESTIGATIONS

Hemogram showed a markedly elevated ESR of 108 mm/h and a white cell count of 12 000 cells/mm³ with 40% of lymphocytes. Other parameters such as liver function tests, renal function tests were normal. HIV monospot was negative. Blood cultures were also negative. Discharge from the sinuses revealed coagulase-positive staphylococci sensitive to vancomycin, piperacillin and tazobactam, linezolid, teicopenem, rifampicin, clindamycin and gentamycin. The organism was resistant to erythromycin, cotrimaxazole, amoxicillin, ampicillin and ciprofloxacin. The microbiologist recommended the patient to be started on vancomycin, piperacillin, tazobactam and linezolid based on the organism sensitivity pattern. MRI showed a soft tissue lesion in the right orbit extending into the extracalvarial soft tissues overlying the frontotemporal bone with extension into the maxilla and subdural area causing localised cerebral oedema and meningeal enhancement (figure 2A–C). Tissue biopsy from the upper lid showed staphylococcal granular colonies surrounded by necroinflammatory infiltrate and giant cells (figure 3A,B). On the basis of these findings orbital chronic granulomatous reaction to deep staphylococcal infection was diagnosed.

DIFFERENTIAL DIAGNOSIS

Important differential diagnosis of this condition include mycetoma, tuberculosis, infections with atypical mycobacteria, nocardiosis, cat-scratch disease, syphilis and deep fungal infection which were ruled out based on culture of the pus from the draining sinuses which showed colonies of staphylococci. Tissue biopsy showed typical staphylococcal granular colonies.

TREATMENT

To optimise the antibiotic coverage a medicine consultation was sought and the patient was started on injection piperacillin–tazobactam in a dose of 4.5 g 8 hourly to which the patient developed nausea, vomiting, diarrhoea and gastrointestinal

intolerance. Hence the drug had to be stopped. The patient was then started on injection vancomycin 1 g intravenously twice daily for 2 weeks, followed by tab linezolid 600 mg twice daily

Figure 3 (A) Staphylococcal granular colonies surrounded by necroinflammatory infiltrate, scattered round cells and giant cells. Within these fragments are multiple basophilic granules of variable sizes with peripheral radiation (H&E, 10 $\times$). (B) Staphylococcal granular colonies surrounded by granulomatous inflammation (May-Grunwald-Giemsa stain, 20 $\times$).

for 6 weeks with monitoring of serum electrolytes, blood urea and serum creatine, along with anticonvulsants to which the patient responded well. Topically he was put on clindamycin skin ointment over the sinuses three times daily for 6 weeks along tear supplements and tobramycin eye ointment to the right eye.

OUTCOME AND FOLLOW-UP

The patient showed remarkable improvement with reduction in the size of the swelling and proptosis (figure 4A,B).

DISCUSSION

Winslow subcategorised botryomycosis into integumental and visceral forms. The integumental form is more common and is characterised by localised granulomatous skin infections often associated with trauma, foreign body and wound contamination. Visceral form affecting the liver, lung, kidney, prostate, caecum, brain, lymph tissue and orbit is rare.³

(a)

(b)

Figure 4 (A) Showing resolution of the skin lesions with significant reduction in proptosis and chemosis with exposure keratopathy. (B) Lateral view showing significant reduction in proptosis, healed skin nodules and sinuses with exposure keratopathy.

Our 53-year-old patient following trauma presented with proptosis (a primary orbital lesion) with intracranial involvement, involvement of the fronto temporal area and paranasal sinuses. Considering the diagnosis of granulomas such as actinomycosis and mycetoma biopsy from the discharging sinuses overlying the skin and periorbital tissue was carried out. Histopathological examination and biopsy of the specimen revealed a chronic granulomatous reaction to deep bacterial infection caused by *Staphylococcus aureus*.

Although extensive surgical debridement with systemic antibiotics is the treatment of choice⁴ in view of the significant intracranial extension, decision was taken to try medical line of management. Patient responded to intensive antibiotic therapy with vancomycin for 2 weeks and linezolid for 6 weeks along with 6 weeks of topical clindamycin therapy.

It is quoted that the mortality is high in case of intracranial extension.⁵ In this case on presentation there was extensive intracranial involvement causing midline shift. Despite this the patient responded very well to medical management.

Chronic granulomatous reaction to deep staphylococcal infection is common in immune compromised status like diabetes mellitus, AIDS, cystic fibrosis,^{6,7} but in our patient trauma was the only precipitating factor.

The case presented illustrates how chronic granulomatous reaction to deep bacterial infection was diagnosed by biopsy and successfully treated with systemic antibiotics without resorting to surgical debridement.

Learning points

- ▶ Chronic suppurative infection due to a granulomatous response to bacterial pathogens occurs mostly in immunocompromised.
- ▶ The term Botryomycosis is no longer used as it is a pseudofungal infection, not a true mycotic infection. It may be difficult to distinguish from mycetoma and other chronic granulomatous infections.
- ▶ Diagnosis is by finding bacteria in the pus draining from the sinuses.
- ▶ Treatment may require surgical debridement and long-term antibiotic therapy.

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