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Infectious surgically induced necrotising scleritis presenting 10 years post-pterygium excision: a case report

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Abstract

Background: Surgically induced necrotizing scleritis (SINS) is a rare sequela that may occur after ocular surgery. We report a case of infectious SINS as a complication of pterygium excision.
Case presentation: A 75-year-old man had undergone pterygium excision in the right eye and developed infectious SINS 10 years post-surgery. The sclera adjacent to the previous site of pterygium excision showed significant thinning with uveal show, complicated with signs of infection including purulent discharge, necrotic sclera, and hypopyon. Conjunctival swab culture yielded *Pseudomonas aeruginosa*. Blood investigations demonstrated positive rheumatoid factor. The patient was successfully treated with a 2-week course of systemic and topical antibiotics with topical steroid.
Conclusion: SINS may develop post-pterygium excision. Early diagnosis and intervention prevent progression and further complications.

Keywords: pterygium, pterygium surgery, scleritis, surgically induced necrotising scleritis

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Jangkitan skleritis menekrosis berikutan pembedahan terjadi selepas sepuluh tahun pembedahan pterygium: satu laporan kes

Abstrak

Latar belakang: Skleritis menekrosis berikutan pembedahan (SINS) adalah komplikasi yang jarang berlaku selepas pembedahan mata. Kami melaporkan satu kes skleritis menekrosis berjangkit yang diperolehi melalui pembedahan sebagai komplikasi pembedahan pterygium.
Kes: Seorang lelaki berumur 75 tahun yang telah menjalani pembedahan pterygium mata kanan mengalami sckleritis menekrosis berjangkit sepuluh tahun selepas pembedahan. Sklera berdekatan lokasi pembedahan pterygium sebelum ini mengalami penipisan yang ketara dengan menampakkan tisu uvea, dan terdapat tanda-tanda jangkitan seperti nanah, sklera yang nekrotik, dan serta pengumpulan nanah (hypopyon). Sampel konjunktiva yang dikultur menunjukkan kehadiran *Pseudomonas aeruginosa*. Ujian darah menunjukkan keputusan positif untuk faktor rheumatoid. Pesakit ini telah berjaya dirawat dengan antibiotik secara sistemik dan ubat titis steroid selama dua minggu.
Kesimpulan: SINS boleh berlaku selepas pembedahan pterygium. Diagnosa dan intervensi yang awal dapat mencegah kesan rebakkan dan komplikasi selanjutnya.

Keywords: pembedahan pterygium, pterygium, skleritis, skleritis menekrosis melalui pembedahan

Introduction

Surgically induced necrotizing scleritis (SINS) is an intense scleral inflammation of a focal area occurring adjacent to the site of previous scleral or limbal incision. It is a rare sequela that may occur after interventions such as trabeculectomy, cataract extraction surgery, retinal detachment surgery, strabismus surgery, and pterygium surgery.[1] We report a case of infected SINS 10 years post-pterygium excision.

Case report

A 75-year-old Chinese man with a history of pterygium excision in his right eye performed 10 years ago presented with redness, foreign body sensation, discharge, and blurred vision in his right eye for 1 week. On examination, the visual acuity was 6/60 in the right eye and 6/9 in the left eye with no relative afferent pupillary defect. The eyelid was erythematous and oedematous with purulent discharge. An area of thinned sclera measuring 6.7 x 9.0 mm with uveal tissue plugging and tissue slough were seen from 2 to 5 o'clock at the previous site of excised pterygium (Fig. 1). Seidel test was negative. A corneal scar was seen at the nasal limbus, corresponding with the history of pterygium excision. There was associated anterior chamber inflammation with presence of hypopyon, fibrin plaque, and peripheral anterior synechiae at 2 o'clock. There was nuclear sclerosis of the crystalline lens in the right eye and fundus examination was normal with clear media on B-scan.

Conjunctival swab at the site of tissue slough yielded *Pseudomonas aeruginosa*, which is sensitive to ciprofloxacin, gentamicin, and ceftazidime. Biochemical parameters showed raised inflammatory markers with C-reactive protein of 14.5 mg/L, erythrocyte sedimentation rate of 40 mm/hr, and raised white cell count with a neutrophil predominance. Rheumatoid factor was positive. Renal profile, liver enzymes, antinuclear antibodies, syphilis screening, full and microscopic examination of the urine, and urine culture were normal. Tumour markers (cancer antigen 19-9, carcinoembryonic antigen, alpha-feto protein, prostate-specific antigen) were normal.

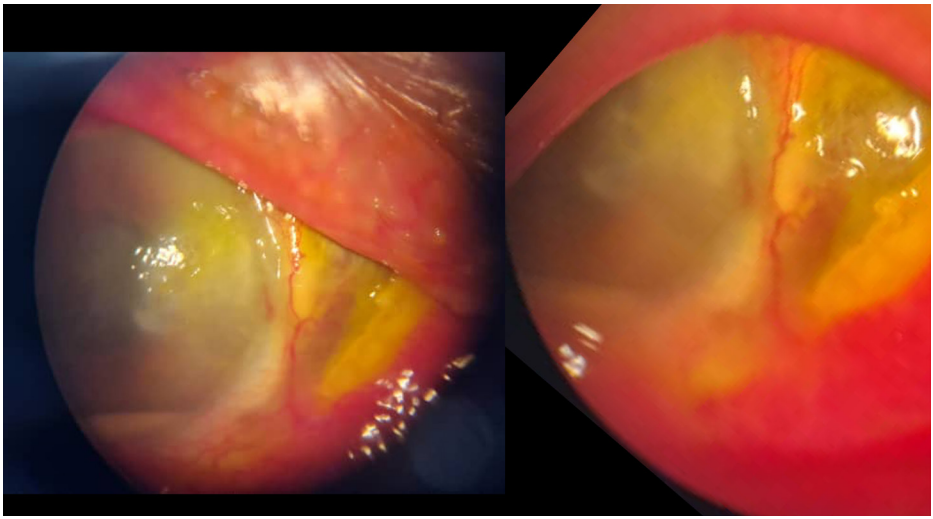

Fig. 1. Area of necrotic and inflamed sclera with uveal show and hypopyon.

He was treated for infectious SINS with severe anterior chamber reaction, and was started on antimicrobial and anti-inflammatory therapies. Empirical moxifloxacin and atropine eyedrops with oral ciprofloxacin and oral ibuprofen were started. Two ocular hypotensive agents were started due to the presence of thinned sclera. He was also referred to a rheumatologist, who concluded that he did not have clinical signs that were suggestive of a connective tissue disease, including rheumatoid arthritis.

There was no improvement seen in the first week of treatment. Ocular necrosis progressed further to developing an area of scleral melting measuring 2.4 mm x 2.5 mm inferior to the previously mentioned thinned sclera with worsening of the anterior chamber inflammation. The patient also developed acute kidney injury attributed to the oral ibuprofen. He was comanaged by a nephrologist, who suggested to stop the oral ibuprofen. Antimicrobial therapy was escalated to gentamicin 0.9% and ceftazidime 5% eyedrops with conversion of the oral ciprofloxacin to intravenous route. Topical non-steroidal anti-inflammatory (NSAID, nepafenac), oral anti-collagenase (oral doxycycline), and oral vitamin C were also started. Systemic steroid was not initiated due to the presence of superinfection.

Clinical improvement was noted 2 days after the initiation of intravenous antibiotic, evidenced by the retardation of tissue necrotization and reduction of anterior chamber inflammation. Repeated conjunctival swab a week after

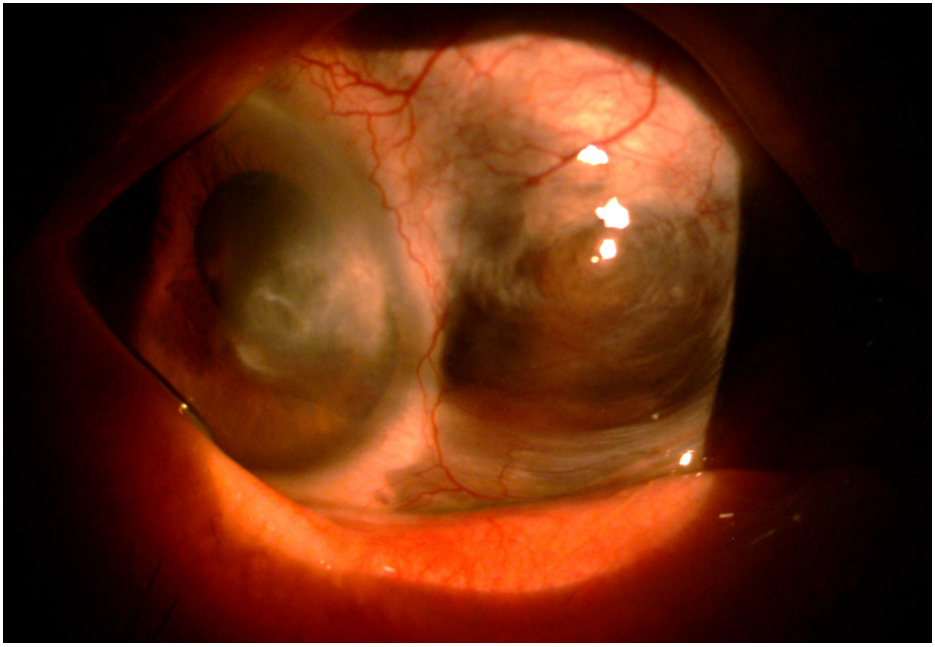

Fig. 2. Stable and epithelialized thinned sclera six weeks after treatment.

treatment had shown clearance from Pseudomonas infection and topical steroid was started. Two weeks following treatment, resolution of scleral thinning was perceptible with the area covered by epithelized tissue. (Fig. 2). The patient was maintained on topical antimicrobial, topical steroids, ocular hypotensive agents, and topical NSAID. Upon review at 6 weeks after hospital discharge, complete resolution of necrotizing scleritis was confirmed.

Discussion

In this case report, SINS occurred 10 years post-pterygium excision with significant infection. We formulated a few hypotheses of the pathophysiology leading to post-operative infectious SINS in this patient.

The first hypothesis was due to the use of adjunctive intraoperative therapy during pterygium excision. It was not known whether the patient's pterygium excision was performed with or without antimetabolite, *e.g.*, mitomycin-C (MMC) or 5-fluorouracil (5-FU), as it was done in a different facility and the patient himself could not recall the details. Antimetabolite use has been found to reduce recurrence rate of pterygium to below 10%.[2] Hence, its use in pterygium excision was widely adopted in Malaysia in the 21st century, which was the time frame of our patient's pterygium excision.

However, antimetabolite use is associated with potentially sight-threatening complications such as scleral necrosis and scleral melting. The suggested pathogenesis included cell apoptosis, followed by vascular disruption and local ischaemia, leading to delayed wound healing.[1] It was associated with increased risk of infection and scleral melting, predisposing the patient to SINS.[1] A review found that 44.8% of 203 cases of scleral necrosis following pterygium surgery had a history of adjunctive beta-radiation or MMC use.[3] Nevertheless, there are also studies that argue that the use of adjunctive therapy is safe with careful patient selection and dosing.[4]

A recent review found that pterygium excision was the most common ocular surgery associated with SINS, and late onset infectious scleral melting was most common post-pterygium excision.[3] Another review found a median latent period of 49 months between pterygium surgery and the onset of scleritis.[5] In cases of infectious postoperative necrotising scleritis after pterygium excision, *Pseudomonas aeruginosa* was the most common bacterium isolated, which corresponded to this case study.[3] Previous studies found that Pseudomonas could produce collagenolytic and proteoglycanolytic extracellular proteases that are capable of damaging ocular tissue, especially in compromised sclera.[6] Pseudomonas infection was also associated with advanced age and immunocompromised patients.[5] A previous study of 55 patients with infectious scleritis found that the median age at diagnosis was 70 years, which is similar to our case.[5]

The incidence of postoperative SINS was found to be higher among patients with underlying systemic autoimmune disease.[1] Rheumatoid arthritis and granulomatosis with polyangiitis are the most commonly associated autoimmune diseases where SINS could be the initial manifestation of the autoimmune disease.[1] It has been proposed that SINS in systemic autoimmune disease is a delayed type III hypersensitivity reaction in which a previous trauma or ischemic event exposes tissue autoantigens, leading to sensitization of immune system and then antigen-antibody immune complex formation around the episcleral vessel walls.[7] A more severe scleral necrosis and ulceration was also found in patients with systemic autoimmune disease as there was overexpression of HLA-DR, localized T-cell activation, and thence deposition of IgM and IgD at the previous surgical site.[8]

However, a positive rheumatoid factor could be attributed to different diseases and conditions, including different connective tissue diseases and infectious diseases. It could be positive in rheumatoid arthritis, primary Sjogren's syndrome, bacterial infections, viral infections, and malignancy.[9] Studies also found that healthy subjects could have positive rheumatoid factors.[9] Hence, rheumatoid factor positivity has to be complemented with suggestive clinical signs before coming to a diagnosis.

As there was mucopurulent discharge and hypopyon in our patient, it was diagnosed as SINS with superinfection. An empirical antibiotic therapy was initiated before switching to targeted therapy. Interestingly, our case had clinical improvement without surgical intervention and systemic corticosteroid treatment. Hodson et. al. found that only 18% of patients with postoperative infectious scleritis responded to medical therapy alone, while most of the cases required early surgical intervention.[5] Poor prognostic indicators include a presenting visual acuity worse than 6/60, concomitant endophthalmitis or keratitis, and fungal etiologies.[10]

Conclusion

We report a case of infectious SINS, which is a rare sequela of ocular surgery. Infectious SINS is a sight-threatening disease in which early diagnosis and prompt intervention are crucial. A complete diagnostic work-up is essential, which includes scleral scraping with culturing, serum inflammatory markers, and autoimmune disease work-up. Aggressive and appropriate antimicrobial therapy are critical in preventing disease progression.

Declarations

Consent for publication
The patient provided informed consent for the use of clinical data and images contained in this case report.

Competing interests
None to declare.

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