Dystrophic calcification of the sclera following pterygium surgery with adjunctive beta-irradiation: Case report of the surgical management

The purpose of this article was to report a late complication of adjuvant beta-radiation after pterygium surgery. This is a case report of a 56-year-old black female patient who presented with an infectious scleritis associated with scleral necrosis and dystrophic calcification. She had undergone pterygium surgery with adjuvant beta-irradiation six years before. She was managed with topical antibiotics and patch graft, with a good outcome. Beta-radiation is associated with visually significant long-term complications and should be avoided. We recommend an excision with extended tenonectomy and free autograft as the procedure of choice.

Keywords: pterygium; beta-irradiation; strontium-90; dystrophic calcification; scleritis; scleromalacia.

Introduction

A pterygium is a wing-like fibrovascular growth of the conjunctiva that extends onto the cornea.\textsuperscript{1} Prevalence rates range from 7\% to 15\%, with onset mostly in the third to fourth decade of life.\textsuperscript{1,2} It occurs more commonly in hot, dry climates where ultraviolet radiation (UVR) is considered the most important environmental risk factor.\textsuperscript{1,2} Other risk factors include a positive family history, chronic ocular irritation or inflammation, keratoconjunctivitis sicca (KCS) and human papilloma virus.\textsuperscript{1,2}

Clinically, a pterygium is seen as a fleshy interpalpebral wing-like mass, with the apex extending towards the visual axis.\textsuperscript{1,3} There may be evidence of dryness, conjunctival injection and, in severe cases, a dellen may develop.\textsuperscript{1} Vision can be reduced because of dryness, secondary astigmatism or obstruction of the visual axis by the growth.\textsuperscript{1,3}

Pterygia are a degenerative condition with classic elastotic degeneration of the subepithelial tissue.\textsuperscript{1,2} Focal UVR-induced limbal stem cell failure has been thought to be central in the pathogenesis, with conjunctival overgrowth and corneal basement membrane invasion.\textsuperscript{3} Pterygia have demonstrated an over-expression of p53 oncogenes that are thought to be responsible for the epithelial overgrowth.\textsuperscript{1,2} Mutations in this gene are associated with ocular surface neoplasia and, therefore, a 10\% incidence of ocular surface squamous neoplasia (OSSN) as an incidental finding on histology with pterygium surgery has been described.\textsuperscript{4}

Pterygia that are symptomatic or cosmetically unacceptable are managed by surgical excision.\textsuperscript{1,3} The way in which the resulting conjunctival defect is managed is largely responsible for the recurrence rates. This has been shown to be high when leaving the sclera bare (24\% – 89\%) and lowest with a conjunctival autograft (0\% – 5\%).\textsuperscript{1,3} In an attempt to reduce recurrence rates, further adjunctive medical therapies have been employed in the past. These include beta-irradiation, thiotepa, mitomycin-C (MMC), 5-fluorouracil (5FU), anti-vascular endothelial growth factor (VEGF), cyclosporine, collagen implants and doxycycline. Beta-irradiation uses strontium-90 as single or recurrent applications.\textsuperscript{5,6} Significant dose-related complications have arisen from beta-irradiation that includes scleral necrosis, infectious scleritis, corneal perforation, cataract, glaucoma and calcific plaques.\textsuperscript{1,2,7}

This case report describes a complication of pterygium surgery with adjuvant beta-irradiation, presenting 6 years after the primary surgery.
Case report

A 56-year-old black female patient was referred to the cornea unit at St John Eye Hospital with a history of a painful right eye for 8 weeks. Her ocular history was noteworthy for pterygium surgery (with adjuvant beta-irradiation) in the right eye 6 years ago and atypical optic neuritis on the right 37 years ago. She was hypertensive, with no other medical history.

On examination her visual acuity was no light perception in the right and 6/6 in the left, corrected with spectacles. Her right eye was significantly injected with a scleral calcific plaque nasally (Figure 1). She had a cataract, an afferent pupillary defect and a pale optic disc. Her left eye was normal on ocular examination.

Investigations were performed to exclude systemic causes of scleritis. These included a full blood count, erythrocyte sedimentation rate, c-reactive protein, rheumatoid factor, anti-nuclear anti-bodies, angiotensin converting enzyme, treponema pallidum antibodies and chest X-ray. All investigations were found to be within normal limits. The patient was diagnosed as a scleral necrosis with secondary dystrophic calcification following beta-irradiation that developed an infective scleritis. She was admitted on an intensive course of topical ciprofloxacin 0.3% and planned for surgery a week later. At surgery, the calcific plaque was removed, the sclera debrided and a corneal patch graft performed (Figure 1). The patch was covered with vascularised tenons and conjunctiva to ensure optimal healing. She was discharged the following day on topical ciprofloxacin 0.3% and dexamethasone 0.1%. Histology of the specimen showed calcified fibrous tissue with a background of acute inflammation. Two months after surgery, she developed recurrent inflammation that was controlled with a short course of oral prednisone. At last follow-up 6 months after surgery, the eye remained quiet on topical lubricants and prednisolone 1% daily.

Ethical considerations

Ethical clearance to conduct the study was obtained from the University of the Witwatersrand Ethics Committee (Ethical Clearance Number: M1810104).

Source: Anterior segment photos taken by Dr R Höllhumer

FIGURE 1: Anterior segment photos of the right eye. (a) Infectious scleritis resolved after a week of topical ciprofloxacin 0.3%. Residual scleritis with calcific plaque at the time of surgery. (b) At surgery after removal of the calcific plaque and completion of the corneal patch. (c) At surgery after the patch has been closed with a vascular tenons and conjunctiva. (d) At last follow-up, 6 months after surgery.
Discussion

Pterygia are common in the hot, dry South African climate. Surgery has been shown to be an effective solution with the lowest recurrence rates when using conjunctival autografts.\textsuperscript{1} Beta-irradiation was used in the past as an adjuvant therapy to reduce the incidence of recurrence.\textsuperscript{1} There are, however, significant potential complications associated with the use of this modality.\textsuperscript{1,2,3} Our patient presented with scleral necrosis, dystrophic calcification and infectious scleritis as complications.

The method of surgical closure after pterygium excision has been shown to be central in the incidence of recurrence.\textsuperscript{1} Five main approaches have been described for closure, bare sclera, primary closure, rotational autograft, free autograft and amniotic membrane transplant (AMT).\textsuperscript{1} Leaving bare sclera after pterygium removal is universally accepted as a poor management option, with recurrence rates as high as 80%.\textsuperscript{5} The gold standard for conjunctival closure is an autograft from the superior bulbar conjunctiva. Recurrence rates with this technique have been 2% – 12%, and with the P.E.R.F.E.C.T. (pterygium extended removal followed by extended conjunctival transplantation) technique, there were no recurrences.\textsuperscript{5} Amniotic membrane transplant has been shown to be inferior to conjunctival autograft, with variable recurrence rates of 4% – 60%.\textsuperscript{5} Our patient underwent pterygium excision with a free autograft.

Medical adjuvants that have been employed to minimise recurrence rates include beta-irradiation, thiopeta, MMC, 5FU, anti-VEGF, cyclosporine, collagen implants and doxycycline.\textsuperscript{3} Beta-irradiation uses strontium-90 to cause focal radiation with limited ocular penetration.\textsuperscript{3} Studies have shown reduced recurrence rates of 0% – 12%; however, the complications of this therapy may result in irreversible visual loss in up to 13% of patients.\textsuperscript{5,5} These complications often have a long latency period and include scleral necrosis, infectious scleritis, endophthalmitis, corneal perforation, cataract, glaucoma and calcific plaques.\textsuperscript{1,2,3} Infectious scleritis is most commonly caused by \textit{Pseudomonas aeruginosa}; however, other organisms that have been isolated from calcific plaques include fungi, \textit{Streptococcus} and \textit{Stenotrophononas maltophilia}.\textsuperscript{7,8} The calcific plaque acts as a nidus for organisms and, therefore, infectious scleritis can be precipitated by the removal of the plaque as this disrupts the ocular surface integrity.\textsuperscript{7,9} Thiotepa is a radiomimetic alkylating agent that has been used in the post-operative period and reduced recurrence rates to 3% – 28%.\textsuperscript{2} Its use lost favour with the emergence of other adjuvant therapies. Mitomycin-C is an alkylating agent used in chemotherapy. A 5-min application of 0.02% – 0.04% has shown a reduction of the recurrence rate from 32% to 7%.\textsuperscript{2,3} This therapy may, however, also cause significant complications that include delayed healing, scleral thinning and calcific plaques.\textsuperscript{2,3} 5-Fluorouracil is also a chemotherapy agent that has been used topically for OSSN and pterygium surgery. It inhibits tenon capsule fibroblasts, and in a case control study, recurrence rates were reduced from 26% to 6% with the adjuvant use of 5FU.\textsuperscript{2,3} This has also been found to be a safe therapy with minimal risk.\textsuperscript{3} Topical cyclosporine has been used successfully for the management of keratoconjunctivitis sicca, allergic conjunctivitis and other inflammatory disorders. For pterygium surgery, it has been shown to reduce recurrence at a dose of 0.05%.\textsuperscript{3} Our patient had adjuvant beta-irradiation that resulted in scleral necrosis, dystrophic calcification and ultimately an infectious scleritis.

Scleromalacia as a complication of adjuvant therapy can be repaired with corneal or scleral patch tissue, with an overlay of vascularised conjunctiva and tenons.\textsuperscript{10} Our patient was commenced on topical antibiotics to control the infectious scleritis before scheduling surgery. To ensure adequate healing of this area of devitalised tissue, adjacent tenons and conjunctiva were mobilised to cover the patch graft. This resulted in a good cosmetic outcome (Figure 1). Our patient had no visual prognosis because of the previous episode of atypical optic neuritis and, therefore, this was primarily a globe saving procedure.

Conclusion

Pterygium surgery is a commonly performed procedure, with the gold standard for conjunctival closure a free autograft. Many adjuvant therapies have been used in the past with potential negative long-term sequelae. Our patient demonstrates the outcome after pterygium surgery with adjuvant beta-irradiation. We therefore recommend an excision with extended tenonectomy and free autograft as the procedure of choice.

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Author’s contributions

I declare that I am the sole author of this research article.

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