American Journal of Ophthalmology Case Reports 5 (2017) 63–66

Contents lists available at ScienceDirect

American Journal of Ophthalmology Case Reports

journal homepage: http://www.ajocasereports.com/

Case report

Surgical interventions for late ocular complications of relapsing polychondritis

Yuan He*, Huifeng Liu, Zhi Ji, Binliang Zhu

Department of Ophthalmology, The Second Affiliated Hospital of Xi'an Medical University, Ocular Immunology and Inflammation Institute, Shaanxi Provincial Clinical Research Center for Ophthalmology, Xi'an, China

ARTICLE INFO

Article history:
Received 24 February 2016
Received in revised form
17 November 2016
Accepted 7 December 2016
Available online 10 December 2016

Keywords:
Relapsing polychondritis
Late ocular complications
Surgical intervention

ABSTRACT

Purpose: To report a case of surgical interventions for a patient with relapsing polychondritis who presented with late ocular complications.
Observations: A 44-year-old male was diagnosed to have relapsing polychondritis on the basis of recurrent acute auricular chondritis, deformity of the ear, saddle nose deformity and painful nasal chondritis, acute ocular inflammation with conjunctivitis, episcleritis and keratouveitis, laryngotracheal chondritis, erythema nodosum in the skin, a history of polyarthritis, and abnormal blood examination findings. The acute ocular and auricular inflammation was resolved with oral corticosteroid treatment. Intraocular pressure (IOP) of the left eye was 60 mmHg as measured by Goldmann applanation tonometer. Gonioscopic observation revealed the presence of peripheral anterior synechiae and plateau iris configuration. Express drainage screw implantation was applied to the left eye, because topical and systemic medicines failed to decrease the IOP. After 12 months, complicated cataract aggravated in the right eye, and phacoemulsification operation was performed with corticosteroids administered during the perioperative period. His corrected visual acuity was 20/20 for the right eye, and the IOP remained below 21 mmHg for the left eye. The patient has been healthy, without any recurrence, for 36 months.
Conclusion and importance: The present case of relapsing polychondritis is the first to be reported wherein late ocular complications were alleviated by surgical interventions. Routine use of corticosteroids is necessary for successful anti-glaucoma and phacoemulsification operations.

1. Introduction

Relapsing polychondritis is an unusual multiorgan autoimmune disease characterized by recurrent inflammation and progressive destruction of cartilaginous structures and collagen-rich tissues, including the eyes, ears, nose, skin, renal, musculoskeletal, heart, and respiratory systems.[1–7] Ocular manifestations, including proptosis, eyelid edema, conjunctivitis, episcleritis, keratitis, uveitis, secondary open glaucoma, and retinopathy often occur in more than half of patients with relapsing polychondritis.[1–8] We experienced the case of a patient with relapsing polychondritis who presented with ocular complications, including conjunctivitis, episcleritis, keratouveitis, secondary angle closure glaucoma, and complicated cataract. Among the published reports of relapsing polychondritis with ocular manifestations, very few have described surgical management and outcome for late ocular complications. Here, we report the case of successful express drainage screw implantation and phacoemulsification operations in a patient with relapsing polychondritis with late ocular complications.

2. Case report

A 44-year-old man visited a local ophthalmic clinic with the complaint of recurrent bilateral and painful red eyes, swollen eyelids, and blurred vision that had started 5 months earlier. He had been treated with antibiotics and anti-allergy medicine topically and systemically for bacterial and allergic conjunctivitis; however, the treatment could not alleviate the inflammation. On the contrary, the symptoms of redness and pain in the eyes were aggravated. He was also being treated with topical corticosteroids for interstitial keratitis for a long period of time, and the inflammation was controlled only for a very short duration. An ophthalmic examination showed acute ocular inflammation with redness and

* Corresponding author. The Second Affiliated Hospital of Xi'an College of Medicine, 167 Fang Dong Street, Xi'an 710038, China.
E-mail address: openji7127@hotmail.com (Y. He).

http://dx.doi.org/10.1016/j.ajoc.2016.12.007

mixed conjunctival congestion in both eyes with dilated vessels, conjunctival swelling (Figs. 1 and 2), and auricular chondritis that led to a swollen, inflamed, painful, and soft left ear (Fig. 3). There was also a saddle nose deformity and erythema nodosum in the skin (Fig. 4). His corrected visual acuity was hand motions (HM)/30 cm for the right eye and 20/20 for the left eye. The intraocular pressure (IOP) for the two eyes was 16 mmHg (right) and 60 mmHg (left), as measured by a Goldmann applanation tonometer. An ocular examination revealed diffuse conjunctivitis, episcleritis, and keratouveitis with dense whole intrastromal corneal infiltrate in the right eye (Fig. 1). The left eye presented with diffuse conjunctivitis and episcleritis (Fig. 2). The posterior section of the right eye could not be observed clearly because of the cloudy cornea. The anterior chamber angles were shallow for both the eyes. Ultrabiomicroscopy (UBM) (Fig. 5) revealed narrow anterior chamber angles for both the eyes and accumulation of inflammatory cells at the corner of the anterior chamber angles. The flow of inflammatory cells in the aqueous humor also indicated anterior uveitis. Both narrow anterior chamber angles and anterior uveitis were caused by this secondary IOP elevation. Gonioscopic observation revealed the presence of bridge-like peripheral anterior synechiae that discontinuously occupied more than 60% of the angle and plateau iris configuration in the left eye.

A corneal scrape was taken at the presentation time but failed to show any growth on standard culture media. A blood examination revealed that serum total protein levels, total hemolytic complement activity (CH50), complement C3, C-reactive protein levels (39.5 mg/L, the normal value is less than 5 mg/L), and Westergren sedimentation rate (46 mm/h, the normal value is less than 20 mm/h) were elevated with mild leukocytosis. The test results for rheumatoid factor, antinuclear antibodies, antimitochondrial antibodies, HLA-DR4-DNA, HLA-DW53-DNA, and HLA-B27-DNA were normal. A computed tomography (CT) scan showed lower airway stenosis (Fig. 6) and calcified laryngeal cartilages (Fig. 7).

A diagnosis of relapsing polychondritis was made on the basis of recurrent acute auricular chondritis with marked redness; swelling, warmth, and tenderness of the pinna; deformity of the ear that followed recurrent acute attacks; saddle nose deformity and painful nasal chondritis; acute ocular inflammation; laryngotracheal chondritis; erythema nodosum in the skin; a history of polyarthritis; and abnormal blood examination findings.[9,10] The acute ocular and auricular inflammation was resolved by treatment with 1 mg/kg/day oral prednisone (Fig. 8).[8] Methotrexate was started at a dose of 7.5 mg/week, and the corticosteroids were tapered.[8] Because 0.5% timolol ophthalmic solution, travoprost, brinzolamide, and 2% brimonidinetartrate failed to decrease the

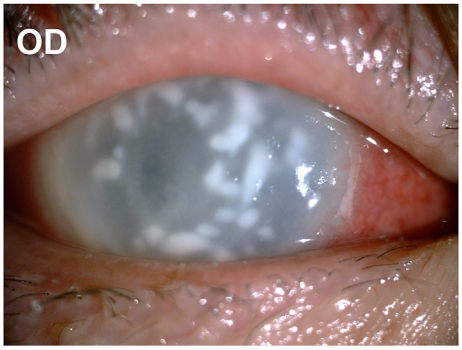

Fig. 1. Red right eye with conjunctival and episcleral congestion. Ocular examination revealed diffuse conjunctivitis, episcleritis, and dense whole intrastromal corneal infiltrate in the right eye. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

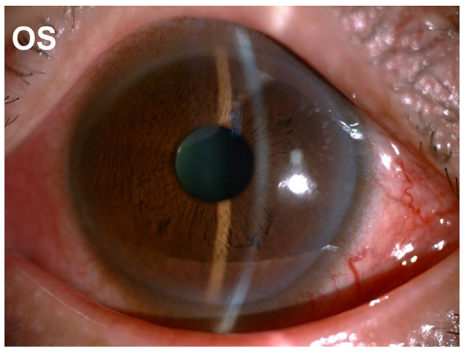

Fig. 2. Diffused conjunctivitis and episcleritis in the left eye.

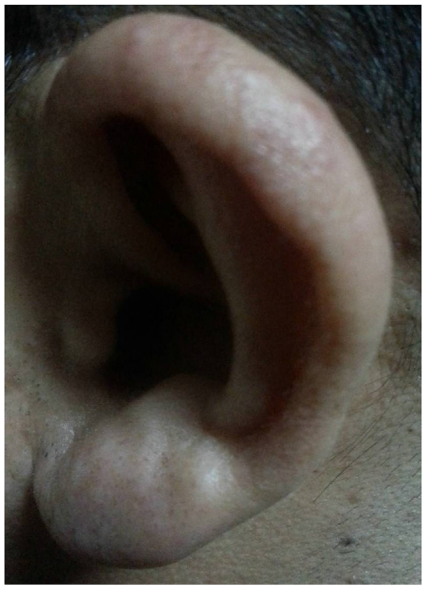

Fig. 3. An inflamed, swollen, and tender left ear.

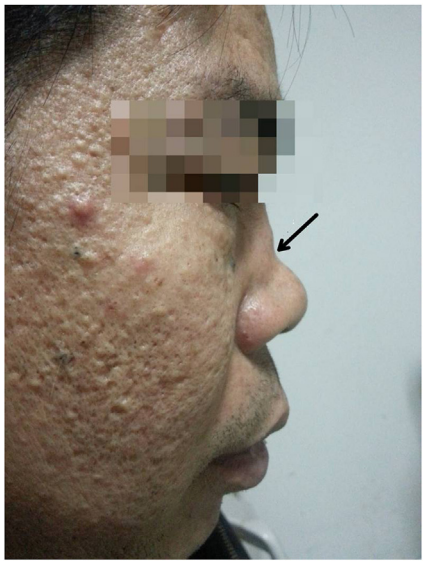

Fig. 4. Saddle nose deformity and inflammation. A type of inflammation with nodular, erythema, and pustule present in the skin.