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CASE REPORT

Sunil Kumar

Bilateral ankle edema with bilateral iritis

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Abstract I report two patient presented to me with bilateral symmetrical ankle edema and bilateral acute iritis. A 42-year-old female of Indian origin and 30-year-old female from Somalia both presented with bilateral acute iritis. In the first patient, bilateral ankle edema preceded the onset of bilateral acute iritis. Bilateral ankle edema developed during the course of disease after onset of ocular symptoms in the second patient. Both patients did not suffer any significant ocular problem in the past, and on systemic examination, all clinical parameters were within normal limit. Lacrimal gland and conjunctival nodule biopsy established the final diagnosis of sarcoidosis in both cases, although the chest x-rays were normal.

Keywords Acute iritis · Ankle edema · Sarcoidosis

Articular symptoms are not uncommon in patients with uveitis. Uveitis is seen in 25% of patients suffering with HLA B-27 arthritis, e.g., ankylosing spondylitis and Reiter's syndrome. Traditionally, when investigating a patient having uveitis with articular symptoms, we do not consider sarcoidosis in the differential diagnosis, especially if chest x-ray is within normal limit. I report two patients of biopsy-proved sarcoidosis in whom bilateral ankle edema and bilateral iritis were the presenting features of the disease. In both patients, there was no clinical evidence of pulmonary sarcoidosis. I would like to emphasize the strong possibility of sarcoidosis in patients having bilateral uveitis and bilateral ankle edema of recent onset and short duration.

I do not have any financial or proprietary interest in any method or material mentioned.

S. Kumar (✉)
Department of Ophthalmology, Mohammad Dossary Hospital,
P.O. Box 335 Al Khobar 31952, Kingdom of Saudi Arabia
e-mail: sunkaru79@hotmail.com
Tel.: +966-3-8945524
Fax: +966-3-8950735

Case number 1

This 42-year-old female of Indian origin attended out patient department (OPD) with complaints of redness and mild pain in right eye of 2-day duration. She reported a past episode of diarrhea and burning pain during micturition followed by bilateral ankle edema and pain in the sole of her feet of 1-month duration. Two weeks after ankle edema, she developed stiffness of fingers and bilateral wrist edema. Prior to development of ocular complaints, she consulted a rheumatologist, and follow-up investigations were done. Her hemogram was normal, erythrocyte sedimentation ratio (ESR) 43 mm at 1 h, C-reactive protein (CRP) 32 mg/L, antinuclear antibody (ANA) and rheumatoid factors were not detected, urine analysis reveal 3–4 pus cells and RBC. Urine culture was sterile. A clinical diagnosis of seronegative rheumatoid arthritis was made by a rheumatologist.

On ocular examination, during first visit, her best-corrected visual acuity was 6/9 OU. Intraocular pressure was 18 mmHg in both eyes. Slit lamp examination showed bilateral findings of moderate (2+) cells and nongranulomatous keratic precipitates.

Detailed history and thorough clinical examination did not suggest any systemic illness presenting as iritis. The patient was not on any medication and did not suffer any significant ocular problem in the past.

As per the guidelines of Royal College of Ophthalmologist of UK, in the absence of strong suspicion of a systemic disease, the patient was not investigated further, instead a provisional diagnosis of Reiter's syndrome was made. Patient was treated with prednisolone acetate 1% eye drop and prednisolone 0.5% ointment.

On her ophthalmic examination 1 week later, anterior chamber reaction was essentially unchanged. Fundus examination revealed bilateral mild blurring and splinter hemorrhage of disc margin. As the iritis was not controlled on topical steroid, tablet prednisolone 60 mg/day was added for 3 days and tapered by 10 mg every third day.

While her iritis was controlled on topical steroid, she comes back with swelling of right upper lid. Examination

revealed dacryoadenitis. Chest x-ray was advised which was within normal limit. Biopsy of lacrimal gland (Fig. 1) established a diagnosis of sarcoidosis.

Case number 2

A 30-year-old female from Somalia was referred to me with the complaints of painful red left eye of 1-week duration. She was being treated at some polyclinic with the diagnosis of acute congestive glaucoma. She did not report any significant past medical or ocular history. Detailed history and thorough clinical examination were not rewarding. On ocular examination, her best-corrected visual acuity was 6/24 in left eye and 6/6 in right eye. Intraocular pressure in left eye was 40 mmHg, and in right eye, it was 20 mmHg. Slit lamp examination of left eye showed that conjunctiva was congested, cornea was edematous, anterior chamber was deep having moderate (2+) cells, and pupil was semidilated and nonreactive to light. All clinical parameters in right eye were within normal limit. Based on the presentation, the patient was thought to be suffering from either Possner Schlossman syndrome or acute nongranulomatous iritis with ocular hypertension. She was treated with I/V mannitol, tablet diamox, and prednisolone 1% eye drops. After regression of corneal edema, gonioscopy revealed 360° angle of grade 3 or 4. On fundus examination, bilateral disc was pink and well defined with small cup. She was discharge on prednisolone eye drop and timolol + dorzolamide combination for 1 week. At the time of discharge, her intraocular pressure in left eye was 10 mmHg.

The patient came back to us 1 month later with complaints of painful red right eye. Her best-corrected visual acuity was 6/9 OU. Her intraocular pressure in both eyes was within normal limit. Slit lamp examination revealed bilateral acute iritis. Despite review of history and repeat systemic examination, there was no evidence of any systemic disease presenting as iritis. Prior to starting systemic steroid to treat bilateral acute iritis, follow-up investigations were done. ESR was 6 mm at 1 h. Hemogram was within normal limit. Venereal Disease Research Laboratory (VDRL) was nonreactive. Chest x-ray was within normal limit. In the absence of suspicion of any systemic disease, the patient was not investigated further. A provisional diagnosis of acute nongranulomatous iritis was made, and the patient was treated with tablet prednisolone 60 mg/day for 3 days, tapered by 10 mg every third day, and prednisolone 1% eye drops.

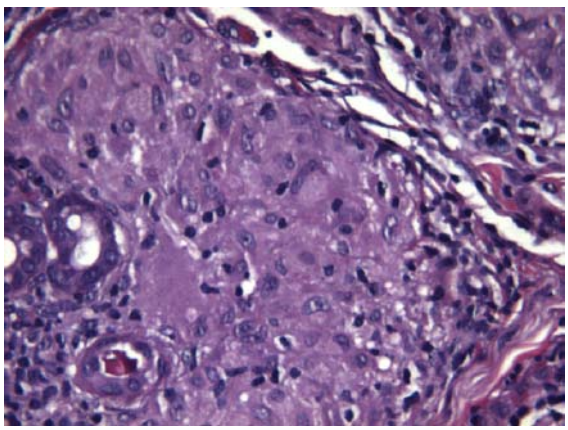

Fig. 1 Histopathological section of lacrimal gland

The patient came back again 5 weeks later with painful redness in both eyes and bilateral ankle edema of 1-week duration. On ocular examination, intraocular pressure was within normal limit. A small pink nodule was documented in left inferior palpabral conjunctiva. Anterior chamber was having 2+ cells.

Considering my past experience, I advised 24-h urinary calcium and serum angiotensin converting enzyme (ACE) level to rule out the possibility of sarcoidosis presenting as bilateral iritis and ankle edema. Twenty-four-hour urinary calcium was 680 mg/24 hours, and serum ACE level was 80 U/L. In view of bilateral iritis and ankle edema with high 24-hour urinary calcium and high ACE level, a provisional diagnosis of sarcoidosis was made. Biopsy of conjunctival nodule of left palpabral conjunctiva established the diagnosis of sarcoidosis.

Discussion

Sarcoidosis is a granulomatous inflammation of unknown origin that varies in severity and distribution from single-organ involvement and self-limiting disease to chronic multisystem inflammation and organ failure; a mortality rate of up to 4.8% has been recorded from referral centers [1]. Limited sarcoidosis is well recognized, and there is a plethora of reports of single-organ involvement in the literature; in these cases, diagnosis is often difficult. Although lacrimal gland and conjunctival involvement are frequent during the course of sarcoidosis, they are generally asymptomatic [2].

The ophthalmologist may be presented with ocular features in keeping with sarcoidosis but without supporting systemic symptoms or signs. Sarcoidosis involves the eye in up to half of patients; manifestations vary enormously and include the following: granulomatous or nongranulomatous anterior uveitis; intermediate uveitis; retinal periphlebitis, usually nonocclusive; multifocal choroiditis; papillitis, optic nerve granuloma, or papilledema; orthoptic manifestations of neurosarcoidosis; lacrimal gland enlargement and dry eye; and, rarely, orbital involvement or scleritis [3].

Uveitis may precede the systemic manifestation of sarcoidosis by many years [4]. Uveitis is the primary manifestation of sarcoidosis in 1.5% of sarcoidosis cases [5]. Uveitis may be granulomatous or nongranulomatous. Fifty percent of patients with sarcoidosis had osteoarticular manifestation, and in 5.7% of patients, such manifestation revealed the disease [6]. Ankle joint involvement is a common presentation of sarcoidosis of acute onset [7]. All sarcoid arthritis patients had bilateral ankle joint involvement. Nine percent of patients with possible reactive

arthritis turned out to have sarcoid arthritis [8]. ACE levels are high in sarcoid arthritis and permit differentiation with seronegative polyarthritis [9]. Among uveitis patients, the sensitivity and specificity of ACE level are 73% and 83%, respectively [10]. According to a large prospective cohort study of early arthritis, presenting in early arthritis clinic, conducted in Netherlands, in a patient below 40 years of age with symmetrical ankle arthritis of less than 2-month duration and erythema nodosum, the probability of sarcoid arthritis is 75% if four of three above-mentioned criteria are present [11].

By correlating the available information in the literature pertaining to sarcoid arthritis and uveitis with my own observations, I feel I am justified in suggesting that bilateral ankle edema of recent onset in females below 40 years of age who are suffering from bilateral acute uveitis is highly suggestive of sarcoidosis.

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