CASE REPORT

Sarcoidosis patient: an unexpected reaction to carbonic anhydrase enzyme inhibitor

Yahya A H Khedr,¹ Abdulla H Khedr²

¹Faculty of Medicine, Ophthalmology, Ain Shams University, Cairo, Egypt
²Department of Ophthalmology, Cleopatra Hospital, Cairo, Egypt

Correspondence to
 Yahya Abdulla Hamed Khedr,
 yahya_khedr@hotmail.com

SUMMARY

Ocular diseases are very common in many of the systemic diseases such as sarcoidosis, and may sometimes be the presenting symptom of the disease. In this case report, we present an unusual reaction of the sarcoid granuloma to carbonic anhydrase enzyme inhibitors (CAIs), which was encountered in a patient with ocular sarcoidosis. This observation was taken after a 2-week interval between a CT scan orbits and an MRI orbits which showed a decrease in size from 4×3×4 cm to 2.5×2.5×2 cm, respectively. We suspected the dorzolamide CAI to have had a significant role in the reduction in size. It is suggested that acidotic changes that occur due to the effect of the carbonic anhydrase inhibitor causes electrolyte imbalance, intracellular as well as extracellular, which lead to the reduction in the size of the granuloma.

BACKGROUND

Sarcoidosis is a chronic granulomatous systemic disease with a multisystem affection course. We commonly find lesions in the lungs, skin and eyes.¹ Sometimes ocular affection is the first presenting sign for sarcoidosis.²

This includes orbital diseases (eg, lacrimal gland enlargement), uveitis, optic neuropathy and other

miscellaneous conditions like scleritis, glaucoma and cataracts.³

In this case report we present an unusual reaction of the sarcoid granuloma to carbonic anhydrase enzyme inhibitors (CAI), which was encountered in a patient with ocular sarcoidosis.

CASE PRESENTATION

A female patient in her sixth decade, presented to the ophthalmology outpatient clinic, reporting of heaviness and tearing in the left eye. She had these symptoms for 6 months. She gave a history of having had excision of excess skin of both upper eyelids (dermatochalasis) 2 years ago.

On external examination, the patient revealed left partial blepharoptosis (S-shaped contour). There was also a 5-mm proptosis in comparison to the right eye.

Corrected visual acuity was 1.0 in both eyes. Eye movement showed orthophoric eyes with full range of movement and good convergence.

Intraocular pressure (IOP) was 19 mm Hg in the right eye and 29 mm Hg in the left eye (using applanation tonometer). A left intraorbital space-occupying lesion (enlarged lacrimal gland) was diagnosed causing partial ptosis, proptosis and mechanical pressure raising the IOP.

Figure 1 CT of orbits.

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Figure 2 MRI of orbits 2 weeks later.

The patient was given dorzolamide (CAI) to reduce the IOP and was sent for an orbital CT scan.

She did not present herself again until 2 weeks later. There was some improvement in her left blepharoptosis and proptosis (around 3 mm difference on exophthalmometer reading). IOP also improved, reading 18 mm Hg in the right eye and 23 mm Hg in the left eye.

Her CT performed the day following her first visit, showed heterogenous enlargement of the left lacrimal gland sizing $4 \times 3 \times 4$ cm (figure 1). One of the CT cuts showed minimal right lacrimal gland involvement. Accordingly she was asked to continue the dorzolamide, as it controlled the IOP. She was sent for an orbital MRI to confirm the involvement of the right lacrimal gland. It was decided to assess ACE level as sarcoidosis was suspected.

Two days later, the MRI showed a definite decrease in the size of the left lacrimal gland lesion which was $2.5 \times 2.5 \times 2$ (figure 2) and confirmed the minimal involvement of the right lacrimal gland. Also the ACE was high at 208 U/L (reference range 18–65).

Various radiological and laboratory findings were sufficient to diagnose the patient as a case of sarcoidosis and a course of systemic corticosteroids was started.

OUTCOME AND FOLLOW-UP

The patient is currently on systemic corticosteroids with obvious improvement in her signs and symptoms.

DISCUSSION

Does CAI have a beneficial effect on sarcoidosis?

We performed a literature search and found that this observation was not recorded in any previous paper.

However, there was one article on eHealthMe stating that 2 out of 1136 people reported to have had sarcoidosis when taking acetazolamide (0.18%).⁴ Baring in mind that there was 17 other drugs co-used by these people, which does not necessarily mean that acetazolamide is the definite cause for

sarcoidosis, whereas our patient was only given dorzolamide without any other drugs co-used.

Improvement in the patient's signs and symptoms is definitely not just due to the drop in her IOP, but also because of the definite decrease in the size of the lacrimal gland. This has been proved objectively and subjectively by the decrease in the ptosis, reduction in the exophthalmometer reading and the noticeable reduction in the size of the left lacrimal gland from $4 \times 3 \times 4$ cm as seen in the CT (figure 1), to $2.5 \times 2.5 \times 2$ as seen in the MRI 2 weeks later (figure 2). We suspected the dorzolamide to have had a significant role in the reduction in size.

It is suggested that acidotic changes which occur due to the effect of the carbonic anhydrase inhibitor causes electrolyte imbalance, intracellular as well as extracellular, which lead to the reduction in the size of the granuloma.

Learning points

- ▶ It would be useful to start a case series to investigate the effect of the carbonic anhydrase inhibitors on sarcoidosis.
- ▶ This is a very early observation, and further research into the mechanism of the carbonic anhydrase inhibitor on sarcoidosis will need to be undertaken in the near future.
- ▶ A comparative study between two groups of sarcoid patients—one group on combined therapy of corticosteroids and carbonic anhydrase inhibitor, while the other group on corticosteroids only—will be helpful.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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