Photo Essay

Isolated superior oblique myositis causing acquired Brown's syndrome

Muralidhar R, Gautam K[1], Christopher D[1], Vidhya N[1], Ramamurthy D[1]

Case Report

A 9-year-old girl presented to us with complaints of binocular diplopia and pain RE for the past 2 days. On examination, she had a vision of 20/20 OU, left face turn with chin elevation, 30 PD of LE hypertropia (RE fixing). The RE elevation was restricted 4 - in adduction and 3 - in abduction (on a scale of 1–4) [Fig. 1]. The elevation saccades of the right eye were normal with normal levator palpebral superior function. The forced duction test for elevation in adduction (topical anesthesia) was positive. Computed tomography (CT) scan of the orbit showed thickening of the right superior oblique muscle involving the tendon [Fig. 2]. No other orbital/sinus pathology was noted. There was no history of systemic illness, polyuria, polydipsia, weight loss, skin lesion or joint pains. A complete hemogram and erythrocyte sedimentation rate were normal. She was started on oral prednisolone 20 mg OD (body weight 24 kg). After 1-week of therapy, there was an improvement in primary position deviation to 16 PD left hypertropia (RE fixing) with pain relief, though the restriction of elevation in adduction persisted [Fig. 3]. No further improvement was noted at 2 weeks. The patient was counseled for pulse steroids, but was noted to have a high random blood sugar of 208 mg/dl and was referred to a pediatrician for systemic workup. Subsequently she was lost to follow-up and could not be contacted.

Discussion

Isolated superior oblique myositis is a very rare entity. This could be because of relative lack of muscle fibers in strap muscles or because of under-reporting due to technical difficulties in imaging.[1] Diagnosis is by CT/magnetic resonance imaging. Interestingly tendon sparing may be noted.[2,3] Tychsen *et al*. had reported 13 cases of trochleitis with superior oblique myositis. They were all successfully treated by oral steroids or excision biopsy. None had ocular motility restriction at presentation.[4] Salam *et al*. reported superior oblique myositis as a presenting feature of Wegener's granulomatosis with inflammation involving adjacent muscles.[1] Fleischmann *et al*. reported successful treatment of isolated superior oblique myositis with oral prednisolone.[5] Kau *et al*. suggested that while oral steroids produce a prompt improvement, they are often associated with persistent restriction in ocular motility, though pulse steroids produced an improvement even when started late.[3] In our patient, elevation restriction was persistent after 2 weeks of oral steroids, though steroids were commenced

Access this article online

Quick Response Code:

Website: www.ijo.in

DOI: 10.4103/0301-4738.158085

Department of Pediatric Ophthalmology and Strabismus, [1]The Eye Foundation, R. S. Puram, Coimbatore, Tamil Nadu, India

Correspondence to: Dr. Muralidhar R, Department of Pediatric Ophthalmology and Strabismus, The Eye Foundation, 582A, D.B. Road, Coimbatore - 641 002, Tamil Nadu, India. E-mail: rajamanimurali@rediffmail.com

Manuscript received: 13.10.14; Revision accepted: 02.04.15

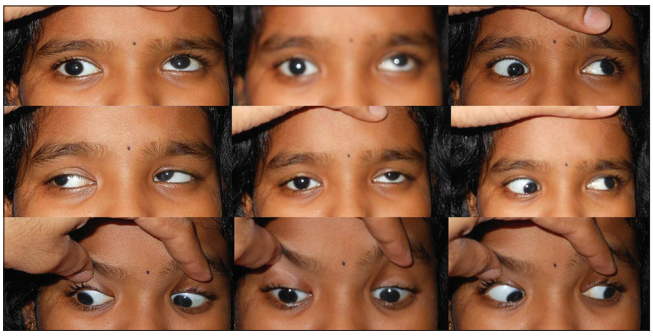

Figure 1: Cardinal gaze photograph at presentation showing left hypertropia with restriction of right eye elevation in adduction. There is a 'Y' pattern divergence on attempted elevation

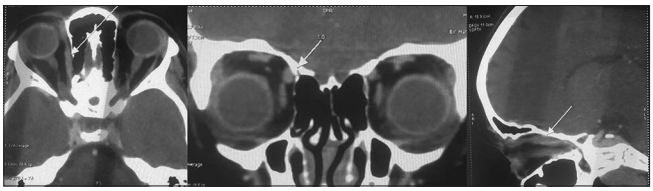

Figure 2: Axial, coronal and sagittal computed tomography scan sections showing enlargement of the right superior oblique muscle

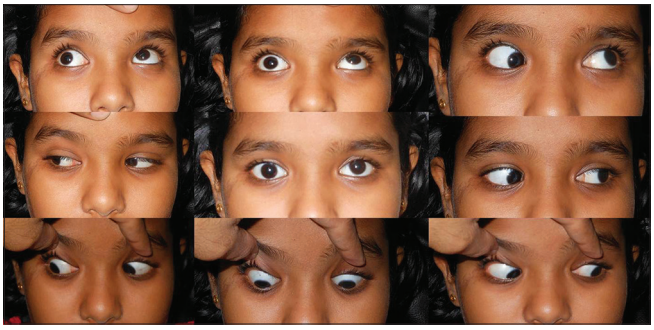

Figure 3: Significant improvement in left hypertropia after 1-week of oral steroids. The restriction of right eye elevation in adduction persists

2 days after presentation. Relief of pain and improvement in primary position deviation argue for a therapeutic role. The high blood sugars were probably a side-effect of oral steroids. Steroids injected in the region of the trochlea were not considered as an option in our case because it would have required general anesthesia. Furthermore, the entire length of the muscle was inflamed. To conclude treatment with oral steroids, even when instituted promptly after diagnosis can leave behind a persistent motility restriction. The role of pulse steroids in this rare condition merits further investigation both as first line and in patients with persistent motility restriction.

References

1. Salam A, Meligonis G, Malhotra R. Superior oblique myositis as an early feature of orbital Wegener's granulomatosis. Orbit 2008;27:203-6.
2. Stidham DB, Sondhi N, Plager D, Helveston E. Presumed isolated inflammation of the superior oblique muscle in idiopathic orbital myositis. Ophthalmology 1998;105:2216-9.
3. Kau HC, Kao SC, Peng CH, Hsu WM, Tsai CC. Methylprednisolone pulse therapy in patient with isolated superior oblique myositis. Eye (Lond) 2006;20:1106-9.
4. Tychsen L, Tse DT, Ossoinig K, Anderson RL. Trochleitis with superior oblique myositis. Ophthalmology 1984;91:1075-9.
5. Fleischmann R, Assmann A, Bohner G, Ruprecht K. Isolated myositis of the superior oblique muscle. J Neurol Neurosurg Psychiatry 2013;84:402-3.

Cite this article as: Muralidhar R, Gautam K, Christopher D, Vidhya N, Ramamurthy D. Isolated superior oblique myositis causing acquired Brown's syndrome. Indian J Ophthalmol 2015;63:340-1.

Source of Support: Nil. **Conflict of Interest:** None declared.