

Anterior plagiocephaly with contralateral superior oblique overaction

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Anterior plagiocephaly is a craniofacial anomaly related to premature unilateral synostosis. We present three cases of anterior plagiocephaly with contralateral superior oblique dysfunction. A detailed ophthalmic examination, including orthoptic assessment for the extraocular muscle misalignment, with appropriate radio-imaging was done in all the three cases. All of them showed a right-sided plagiocephaly, with overaction of the left superior oblique muscle, alternating exotropia and a dissociated vertical deviation. Two underwent surgical correction of squint. Both were well aligned after squint surgery. Plagiocephaly has been reported to simulate superior oblique muscle paresis. We report a rare occurrence of contralateral superior oblique muscle overaction in three children with anterior plagiocephaly.

Key words: Craniofacial dystosis, desagittalization, plagiocephaly, superior oblique overaction

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Plagiocephaly involves the unilateral premature fusion of the coronal suture during the intrauterine development. Unilateral coronal suture stenosis provokes a shortening of the orbital roof on one side.¹ Plagiocephaly is known to be associated with ipsilateral overaction of inferior obliques.^{1,2} Retrusion of the

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trochlea leads to desagittalization of the reflected tendinous segment of the superior oblique. This reduces the effective length of the superior oblique but also reduces the infraducting vector of the latter.^{1,3-4} However, contralateral superior oblique dysfunction has not been reported. We report three cases of right-sided plagiocephaly with ipsilateral superior oblique underaction and contralateral (left-sided) superior oblique overaction.

Case Reports

Case 1

A 12-year-old girl presented to us with complaints of squint since childhood. She was a product of full-term caesarean delivery with normal birth weight.

Visual acuity in both eyes was 20/20 unaided. Ocular motility examination showed left eye exotropia with superior oblique overaction in the left eye. She had right-sided inferior oblique overaction [Figure 1a]. In primary position, she measured fixing right eye 18 prism diopters (pd) with left hypotropia 8 pd with superior rectus overaction in primary position [Table 1]. An A pattern of 15 pd was noted along with dissociated vertical deviation (DVD). No hemifacial hypoplasia was noted.

The flattening of frontal bone on the right side was noted [Figure 1b] and a computed tomography (CT) scan was ordered which revealed a right-sided frontal plagiocephaly. The child had undergone CT scan when she was two years old and that too showed right-sided frontal plagiocephaly [Figure 1c]. A diagnosis of alternate exotropia with left superior oblique overaction with DVD was made. A forced duction test was done on table and was negative in the left eye.

Patient underwent left eye lateral rectus recession (9 mm) with posterior tenectomy of the superior oblique. Postoperatively, patient had minimal exotropia with no superior oblique overaction [Figure 1c], with no significant pattern. The DVD persisted postoperatively too.

Case 2

A 15-year-old boy presented to us with complains of squinting since childhood. He was a full-term normal delivery, with no history of birth trauma and a birth weight of 2.8 kg. There was no family history of squint. His best corrected visual acuity was 20/20 in right eye and 20/60 in left eye [Table 1]. There was a

right-sided plagiocephaly with a head tilt to left [Figure 2a].

Ocular motility examination revealed left exotropia of 35 pd for near and 25 pd for distance with left hypotropia of 10 pd



Figure 1a: Left exotropia with left eye superior oblique overaction and an A pattern

[Table 1]. He had a right superior rectus overaction, left superior oblique overaction and an A pattern of 16 pd. Patient also had DVD. A diagnosis of left exotropia with superior oblique



Figure 2a: Right-sided frontal flattening of skull and facial asymmetry

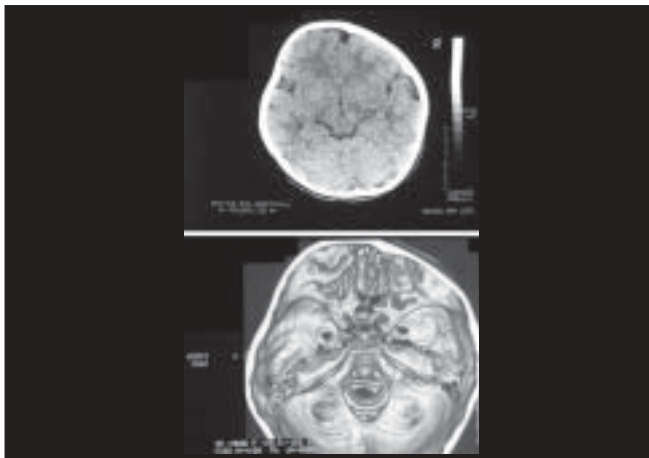


Figure 1b: The upper axial scan shows the right-sided flattening of the skull when the girl was two years old. The lower three-dimensional CT scan clearly shows right side frontal plagiocephaly

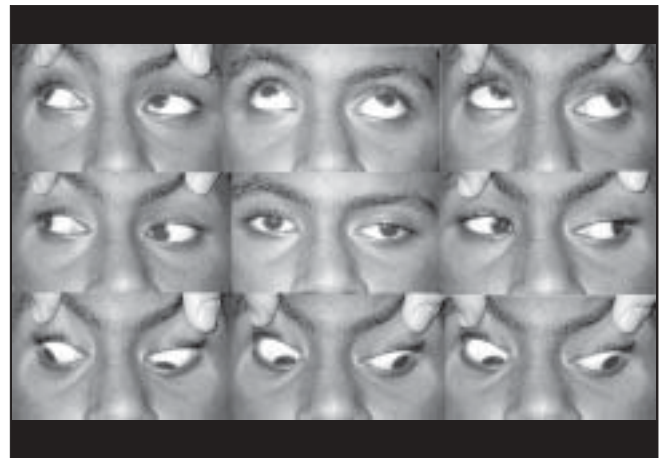


Figure 2b: Exotropia and left-sided superior oblique overaction with an A pattern

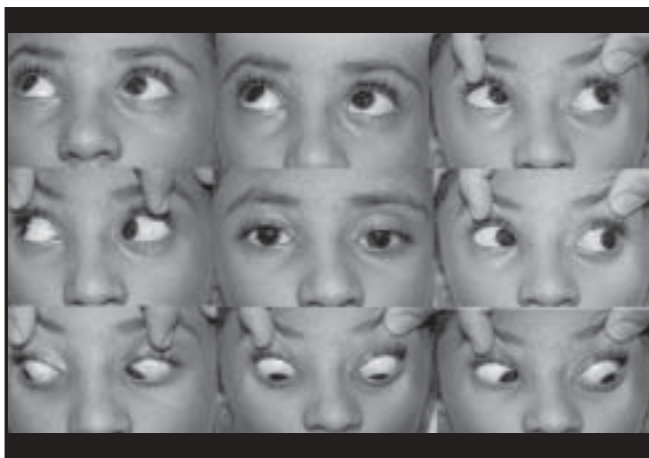


Figure 1c: Postoperative pictures showing excellent correction in primary position and absence of any pattern and superior oblique overaction



Figure 2c: Postoperative correction of exotropia in primary position and a residual superior oblique overaction

overaction, A pattern and DVD was made [Figure 2b]. Forced duction test was negative for left superior oblique.

Patient underwent a left eye lateral rectus recession (6.5 mm) and medial rectus resection (4.0 mm) procedure with posterior tenectomy of superior oblique in the left eye. Patient was well aligned in primary position postoperatively. However, left superior oblique showed mild overaction but no significant pattern and the DVD persisted postoperatively [Figure 2c].

Case 3

A 17-year-old girl presented to us with complaints of watering since one month. She was a full-term normal delivery with no history of birth trauma, with birth weight of 2.6 kg. There was no family history of squint. Her best corrected visual acuity was 20/20 in right eye and 20/80 in left eye [Table 1]. She had a head tilt to the left.

Ocular motility examination revealed left exotropia of 40 pd for near and distance with left hypotropia of 10 pd [Table 1]. She had a left superior oblique overaction and an A pattern of 14 pd [Figure 3 a]. She also had DVD. She had a right-sided frontal plagiocephaly [Figure 3 b]. A diagnosis of left exotropia with right superior rectus overaction, superior oblique overaction, A pattern and DVD was made. She did not want to undergo any surgical intervention.

Discussion

Superior oblique underaction due to plagiocephaly secondary to desagittalization has been well documented in literature.¹⁻⁴ Diamond *et al.*,⁴ found that 11 (32.3%) out of 34 children with plagiocephaly had some form of strabismus. However, only one (2.9%) had inferior oblique overaction and one (2.9%) had superior oblique underaction. The presence of such superior oblique dysfunction with plagiocephaly is rare.^{1,4}

Various theories have been advanced to explain this peculiar motility abnormality. Desagittalization of superior oblique makes it weaker in adduction compared to its antagonist (inferior oblique). The effective length of the superior oblique shortens and hence, makes it weaker in adduction.^{1,4}

Greenberg *et al.*, renamed ocular torticollis with skull and facial symmetry as ocular plagiocephaly⁵ and noted that unilateral superior oblique palsy can give rise to such facial asymmetry. Stevens *et al.*,⁶ in a recent review suggested that

deformational plagiocephaly is indeed distinctively different from the facial hemihypoplasia noted in congenital superior oblique palsy. They suggested that the characteristic facial hemihypoplasia associated with superior oblique palsy develops secondary to gravitational changes and not due to deformational changes. Weiss *et al.*,⁷ reported an imbalance of muscle-pulling forces due to superolateral translation of the

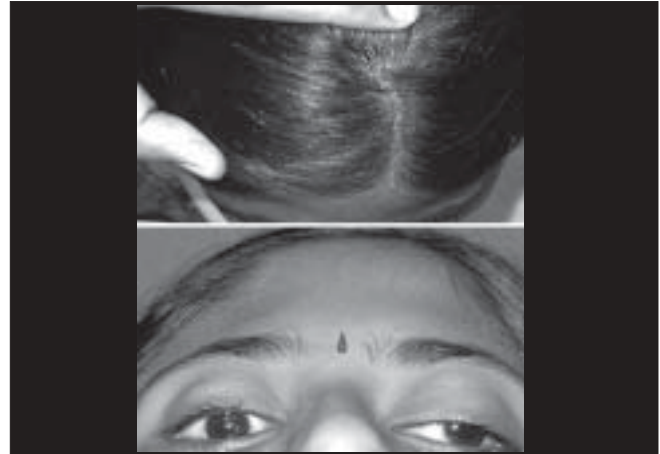


Figure 3a: Frontal flattening of skull on right side

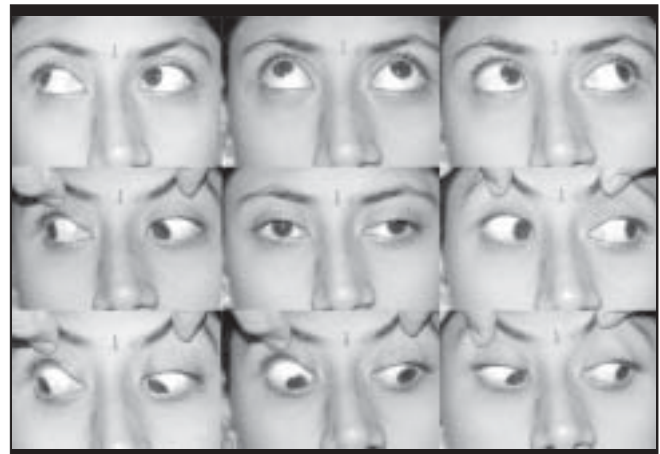


Figure 3b: Large angle exotropia with left eye superior oblique overaction and an A pattern

Table 1: Shows the distribution and measurements of various patients

Case No.	Age/sex	Best correction	Measurements							
			Preoperative				Postoperative			
			Dextro version	Primary position	Levo version	A pattern	Dextro version	Primary position	Levo version	A pattern
1	12/F	RE 20/20 (nil glass)	25 pd BI	18 pd BI	18 pd BI	15 pd	10 pd BI	5 pd BI	4 pd BI	Nil
		LE 20/20 (nil glass)	9 pd BU	8 pd BU	3 pd BU	4 pd BU	2 pd BU			5 pd
2	15/M	RE 20/20 (nil glass)	30 pd BI	35pd BI	30 pd BI	16 pd	5 pd BI	12 pd BI	5 pd BI	Nil
		LE 20/60 (-1.25 D cyl 150°)	16 pd BU	10 pd BU		8 pd BU	5 pd BU			8 pd
3	17/F	RE 20/20 (nil glass)	40 pd BI	40 pd BI	35 pd BI	14 pd	Surgery not done			
		LE 20/80 -1.0D sph/ -1.25 D cyl 1650)	14 pd BU	10 pd BU	4 pd BU					

pd: prism diopters, BI: Base in, BU: Base up, RE: Right eye, LE: Left eye, Sph: Sphere, D: Diopter, cyl: cylinder, M: Male, F: Female, All measurements are fixing right eye and BU implies base up on left eye

superior rectus muscle pulley. They believe that this better accounts for the hypertropia than posterior displacement of the trochlea.

All our cases have right-sided plagiocephaly and left-sided (contralateral) superior oblique overaction. The surgical decision should be considered as the surgeon's personal choice and not as a standard surgery for such cases. Unilateral superior palsy may give rise to contralateral superior oblique overaction secondary to ipsilateral superior rectus contracture.^{3,8} This could well be the explanation for both our cases having right-sided superior oblique weakness secondary to right-sided plagiocephaly. However, while this has been documented for paralytic muscles, our cases were mainly underaction of superior oblique secondary to a mechanical cause. Moreover, none of our cases show any inferior oblique overaction, which should be present. Another possibility could be a rare right-sided plagiocephaly with contralateral inferior oblique paresis giving rise to such incomitance. Two cases underwent a unilateral recession resection surgery with posterior tenectomy of the superior oblique. The cosmetic alignment was good in primary position. Posterior tenectomy of superior oblique is an accepted procedure for moderate A pattern with superior oblique overaction.⁶ Superior oblique tenotomy could have been disastrous, as the patient might have landed in a bilateral superior oblique underaction scenario. Right-sided inferior oblique was not recessed since the patient did not show any significant inferior oblique overaction. However, ipsilateral weakening of superior rectus alone or with contralateral superior oblique weakening could also be tried.

We believe that contralateral superior oblique may show overaction in cases of plagiocephaly, secondary to the underaction

of the ipsilateral superior oblique. Though contralateral superior oblique overaction could be secondary to unilateral superior oblique palsy, we believe that this could also occur in cases of plagiocephaly where the superior oblique dysfunction may be secondary. Importantly, this type of superior oblique overaction may be tackled in appropriate cases which may otherwise be contraindicated in congenital superior oblique palsies.

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