

## BILATERAL BROWN'S SYNDROME ASSOCIATED WITH PREGNANCY: A CASE REPORT

**S E MOORE**, DOBA, DipAppSc(Cumb)

*Department of Ophthalmology, Prince of Wales Hospital & University of New South Wales*

**P J McCARTNEY**, BMedSci, MBBS

*Department of Ophthalmology, Prince of Wales Hospital & University of New South Wales*

---

### Abstract

*Brown's syndrome is recognised to occur in several forms, both congenital and acquired. Similarities have been noted between certain of the acquired forms and stenosing tenosynovitis of the hand. We report a case of bilateral Brown's syndrome with onset related to pregnancy. As stenosing tenosynovitis is known to occur with increased frequency in pregnancy, the observations in this case support the hypothesis that the two conditions share a similar pathogenesis.*

**Key words:** *Brown's syndrome, pregnancy, stenosing tenosynovitis.*

---

Brown's syndrome or superior oblique tendon sheath syndrome was described by Whaley Brown nearly forty years ago.<sup>1</sup> Since then both congenital and acquired forms have been described. In 1969 Sandford-Smith drew the parallel between certain of the acquired types, often the intermittent forms, and stenosing tenosynovitis (DeQuervain's tenosynovitis).<sup>2,3</sup>

We present a case of bilateral Brown's syndrome associated with pregnancy. We suggest that this link with pregnancy supports the concept of a similarity between some forms of acquired Brown's syndrome and stenosing tenosynovitis, as the latter is known to occur more frequently during pregnancy or in the post-partum period.

### CASE REPORT

A 29 year old woman presented with a history of frontal headaches during her first pregnancy, which increased post partum. She complained that on occasion her eyes became fixed in one

position after a rapid change in direction of gaze. These episodes lasted only minutes, during which she noticed diplopia. She had no history of arthritis or local trauma. The pregnancy was complicated by hypertension, fluid retention and proteinuria but proceeded to term with a normal delivery. She was also troubled by painful wrists which persisted for a few months after delivery. She was first seen in our clinic nine months after her first child was born.

She had a face turn to the right, and a gross limitation of either eye on elevation in adduction (Fig 1). There was downshooting of the left eye on dextroversion. A V-pattern was noted in direct elevation with divergence of the right eye. Attempted eye movements in the direction of limitation produced epiphora and pain similar to that complained of as headaches. There was no localised tenderness in the trochlear area; the remainder of the ophthalmological examination was normal. Examination of her wrists revealed no signs of tenosynovitis.

---

*Address for Correspondence:* Ms S E Moore, Department of Ophthalmology, Prince of Wales Hospital, High Street, Randwick NSW 2031.

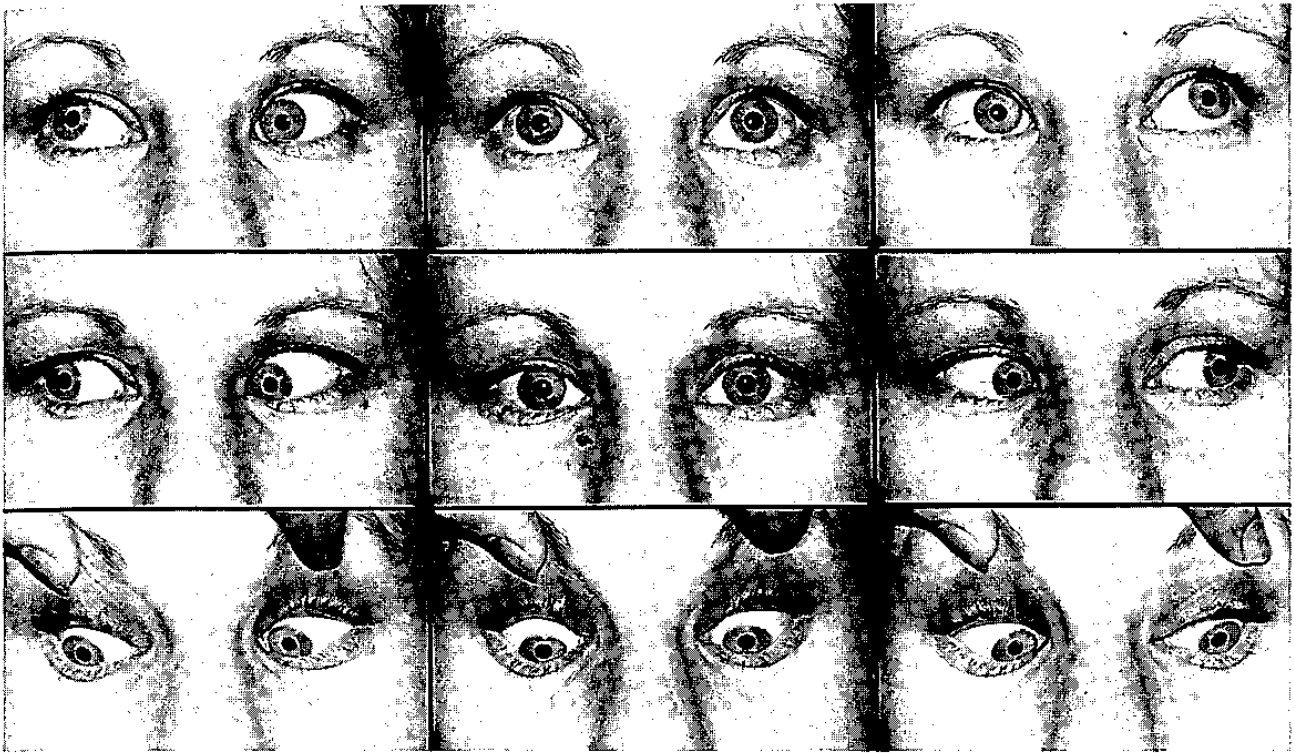


Figure 1: Bilateral Brown's syndrome, with limitation of elevation in adduction more marked in the left eye.

Forced duction testing showed restriction of elevation in adduction. A Hess chart shows a typical bilateral Brown's syndrome, with apparent underaction of the inferior oblique muscles, more marked on the left (Fig 2). There was no overaction of the superior obliques (the ipsilateral antagonists). Antinuclear antibody, rheumatoid factor and erythrocyte sedimentation rate were normal. A CT scan revealed no abnormality of the orbits or adjacent frontal sinuses.

Ocular motility did not change significantly during subsequent visits. Her symptoms increased during a second pregnancy eighteen months later and were only minimally improved by topical dexamethasone eye drops. Post partum, the non-steroidal anti-inflammatory drug ketoprofen 100mg daily produced partial relief of the discomfort. Hess charts showed no significant improvement in motility.

#### DISCUSSION PATHOGENESIS

Mein lists seven possible aetiologies for limitation of elevation in adduction:<sup>4</sup>

1. Short anterior tendon sheath
2. Swelling on the tendon
3. Iatrogenic after tucking of the superior oblique tendon
4. Trauma in the region of the trochlea
5. Anomalous innervation
6. Structural abnormality of inferior oblique
7. Congenital inferior oblique palsy

A short anterior tendon sheath is the original theory suggested by Brown in which initial improvement was obtained after surgical division of the sheath. Such a congenital onset is unlikely in our case given the late presentation without previous suggestive symptoms, although it cannot be conclusively disproved in the absence of a surgical exploration.

Swelling on the tendon was first proposed as an aetiology by Girard, who suggested in 1956 that a circumferential constriction of the trochlea and sheath prevented a locally enlarged tendon from sliding freely.<sup>5</sup> Sandford-Smith states that the cause is a swelling of the tendon of the superior oblique just behind the trochlea at the mouth of the fibrous sheath. Despite muscle

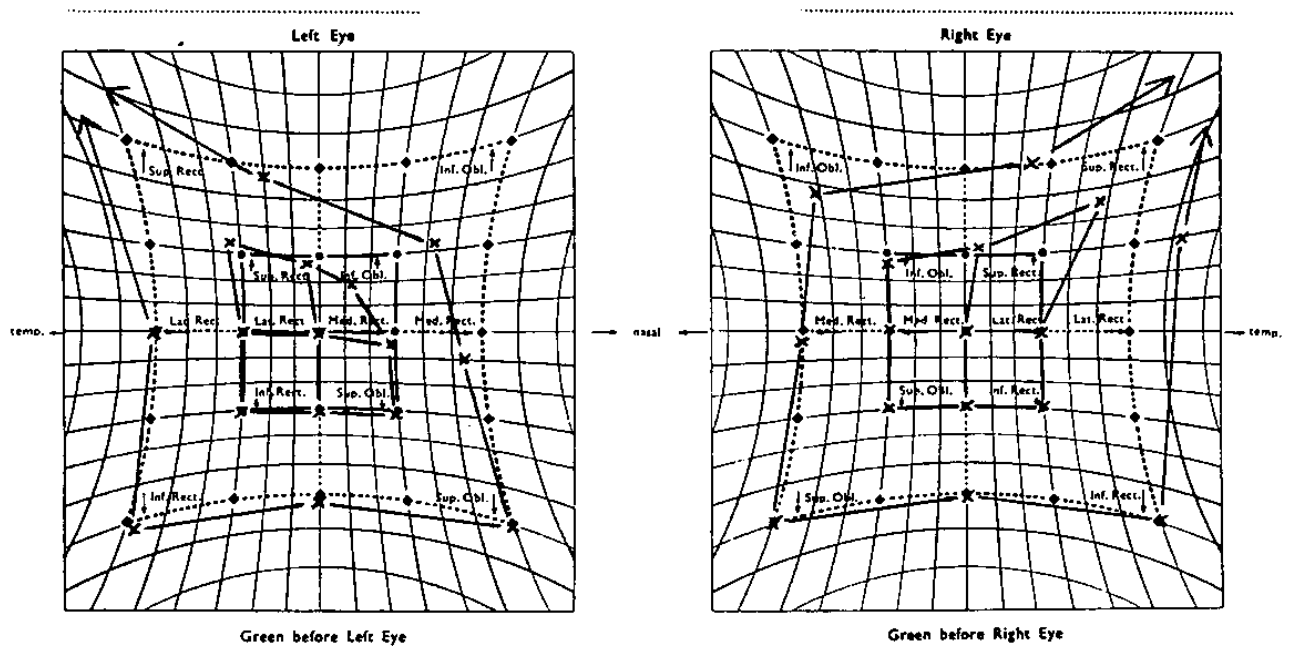


Figure 2: Hess chart showing bilateral Brown's syndrome. Note apparent underaction of the inferior oblique, more marked on the left, but no superior oblique overaction.

relaxation, the swelling is unable to pass into the sheath, causing a restriction of ocular movement. In his original case, the underlying cause was chronic nodular rheumatoid arthritis.<sup>2</sup> However, in 1973 he likened the mechanism of Brown's syndrome to the condition of stenosing tenosynovitis in the flexor tendons of the fingers or abductors and extensors of the thumb.<sup>3</sup> These tendons also pass in fibro-osseous tunnels and develop painful swellings which limit their movement. Sandford-Smith and Waddell<sup>6</sup> hold this to be the cause of the more common type of Brown's syndrome presenting in children, as well as many of the acquired cases, especially those which are intermittent.

Mein describes three other features commonly seen with this aetiology:<sup>4</sup>

- (i) Movement of the eye in the direction of action of the inferior oblique may be improved with practice.
- (ii) Discomfort or pain may be experienced by the patient on attempted elevation in adduction.
- (iii) The patient may describe a clicking sensation resulting in an increase in movement or even an overshoot.

Discomfort and pain when attempting to move

either eye into the area of action of the inferior oblique are marked in our patient. She also complained of locking of the eye in one position on several occasions, but a clicking sensation was not noticed. No increase in movement was obtained during our follow up period. We feel this is the likely mechanism in this case.

An iatrogenic Brown's syndrome after tucking of the superior oblique tendon or trauma in the region of the trochlea were ruled out with a negative history.

Anomalous innervation, structural abnormality of inferior oblique or a congenital inferior oblique palsy are unlikely for several reasons. Innervational defects would not result in symptoms of discomfort. Positive forced duction tests confirmed a mechanical rather than an innervational cause in our patient. The usual sequelae following paralysis of a vertical muscle, that is overaction of the ipsilateral antagonist (the superior oblique of the same eye) was not present. Finally the presence of a V-pattern, according to Mein,<sup>4</sup> is a more reliable method of diagnosis than a forced duction test. With a palsy of the inferior oblique muscle, one would expect an A pattern, since the adducting power

of the affected muscle is lost. However in Brown's syndrome, extra innervation is sent to the inferior oblique in an attempt to elevate the eye and as elevation is mechanically restricted by the superior oblique, adduction occurs with production of a V pattern.

#### ASSOCIATION WITH PREGNANCY

Musculoskeletal problems such as stenosing tenosynovitis, have recently been recognised to occur with greater frequency in pregnant and post partum women.<sup>7</sup> The similarity between Brown's syndrome and stenosing tenosynovitis has already been discussed. Schned found that 25% of female patients with stenosing tenosynovitis were pregnant or postpartum.<sup>8</sup> The reason for this particular susceptibility is not clear. Hormonal effects such as changes in oestrogen and progesterone secretion and the production of relaxin have been implicated in the increased frequency of carpal tunnel syndrome in pregnancy, and Schned suggests these hormonal changes may also play a role in stenosing tenosynovitis.<sup>8</sup> Five of six patients in one series<sup>7</sup> had hand swelling that could have contributed to tendon compression. Mechanical factors involved in caring for newborn babies have also been suggested to play a role.

Our patient had fluid retention and painful wrists during her first pregnancy. Mechanical factors would not seem to be significant in periorbital disease, but hormonal factors and fluid retention may have been involved. Ptosis has been reported in pregnancy, thought to be of the aponeurosis defect type caused by a similar mechanism of fluid retention,<sup>9</sup> but we know of no previous reports of the association of Brown's syndrome and pregnancy.

#### MANAGEMENT

The treatment of Brown's syndrome is controversial and many surgical approaches have been described. While the findings at operation in some cases have helped elucidate some of the causes of the condition, therapeutic results have been disappointing in the long term,<sup>10</sup> probably due to subsequent fibrosis and further restriction of the tendon in its delicate sheath.

We considered medical therapy in our case because the main symptoms were those of discomfort rather than disturbance of functional binocularity. Systemic medications were not used during the pregnancy, and topical steroids produced little effect. Ketoprofen, a non-steroidal anti-inflammatory agent used post partum, has provided moderate relief from symptoms, but no change in ocular motility. Other modalities used for stenosing tenosynovitis in the hand include splinting, surgery and the use of local injection of depot steroid.<sup>8</sup> Splinting is not possible and surgery has been discussed. Depot steroid injections are possibly worth consideration although we have elected not to use them in this case as yet.

#### CONCLUSION

Stenosing tenosynovitis is recognised to occur with a greater frequency in pregnancy. The observation in our case of the onset of bilateral Brown's syndrome related to pregnancy supports the argument that certain of the acquired forms of Brown's syndrome and stenosing tenosynovitis of the hand share a similar pathogenesis.

#### ACKNOWLEDGEMENTS

We thank Dr R McGuinness for the opportunity to examine one of his patients and the Department of Medical Illustration, Prince of Wales Hospital for the photography.

#### References

1. Brown HW. In Allen JH Ed: *Strabismus Ophthalmic Symposium I*. St Louis: Mosby, 1950; 205.
2. Sandford-Smith JH. Intermittent superior oblique tendon sheath syndrome. *Br J Ophthalmol* 1969; 53: 412-417.
3. Sandford-Smith JH. Superior oblique tendon sheath syndrome and its relationship to stenosing tenosynovitis. *Br J Ophthalmol* 1973; 57: 859-865.
4. Mein J. Superior oblique tendon sheath syndrome. *Br Orthopt J* 1971; 28: 70-76.
5. Girard LJ. Pseudoparalysis of the inferior oblique muscle. *Sth Med J* 1956; 49: 342-346.
6. Waddell E. Brown's syndrome revisited. *Br Orthopt J* 1982; 39: 17-21.
7. Schumacher HR, Dorwart BB and Korzeniowski OM. Occurrence of De Quervain's tendinitis during pregnancy. *Arch Intern Med* 1985; 145: 2083-2084.
8. Schned ES. DeQuervain tenosynovitis in pregnant and post partum women. *Obstet Gynecol* 1986; 68: 411-414.
9. Sunness JS. The pregnant woman's eye. *Surv Ophthalmol* 1988; 32: 219-238.
10. Fells P. The superior oblique: its actions and anomalies. *Br Orthop J* 1975; 32: 43-53.