INTRODUCTION

Enophthalmos associated with diplopia occurs most commonly following trauma. With fracture and outward displacement of orbital bones, the net volume of the orbit increases, causing enophthalmos from posterior movement of the globe. Diplopia in this setting may occur from limitation of ocular excursions due to muscle entrapment, capsule adhesion to the fracture site, scarring, traumatic myopathy, or neuropathy. Often a single muscle is involved, producing diplopia from restriction or paresis. In such cases, the ocular deviation usually increases when the globe rotates in one direction and decreases in the opposite direction.

In some patients, the orbit becomes enlarged and the globe moves posteriorly without entrapment, restriction, or paresis of a single muscle. The mechanism of diplopia in this situation is not well characterized. We report a patient with delayed onset of diplopia following craniotomy. The operation resulted in enophthalmos from surgical enlargement of the orbit. Clinical observations, computed tomographic imaging, and eye movement recordings suggest that shortening of the rectus muscles caused the patient’s symptoms.

CASE REPORT AND CLINICAL HISTORY

A 55-year-old man reported binocular diplopia for approximately one year. He had single vision in primary gaze and in reading position, but he developed diplopia with lateral gaze or upgaze. He also experienced transient diplopia upon returning to primary gaze.

Medical history was notable for hypertension and chronic obstructive pulmonary disease. He had undergone a left orbitozygomatic pterional craniotomy 2 years earlier for clipping of a basilar tip aneurysm. The diplopia developed months after the aneurysm surgery, but the patient could not recall the exact date.

Visual acuity was 20/20 OU without correction. The pupils were normal. Ductions were full in the right eye. In the left eye, there was subtle limitation...
of elevation, adduction, and abduction (Figure 1). An Hertel’s exophthalmometer revealed 8 mm of enophthalmos OS. The remainder of the ophthalmic examination was normal.

Binocular fixation fields revealed diplopia with upgaze beyond 10 degrees, or 15 degrees to either side (Figure 2). Fusion was relatively preserved in downgaze. These findings were confirmed by eye movement recordings. A translucent screen was placed 57 cm from the patient. A computer-controlled stimulus was rear-projected by a digital light projector. Eye movements were monitored using two infrared video eye-tracking cameras. The patient was instructed to pursue moving spots or to fixate spots in a grid. Experimental details have been published elsewhere (Economides et al., 2007).

Smooth pursuit recordings are shown in Figure 3. The patient preferred to track with his right eye. On horizontal pursuit, there was symmetrical limitation of rotation of the left eye, causing an esotropia on left gaze and an exotropia on right gaze. Ocular separation developed beyond 15 degrees from primary gaze and increased with gaze eccentricity. Vertical smooth pursuit showed marked limitation of upgaze and relative sparing of downgaze. The patient was asked to fixate points in a 5 × 5 grid, separated by 5 degrees (Figure 4).

FIGURE 1 Extraocular eye movements, showing full versions of the right eye and limited elevation, adduction, and abduction of the left eye. Inset: View from below demonstrating 8 mm enophthalmos of the left globe.

FIGURE 2 Binocular fixation fields. Dark shaded area corresponds to single vision and light shaded area represents diplopia. Diplopia field is remarkably symmetric.
The patient always used his right eye to acquire targets. The position of the left eye was collapsed centripetally, especially for upper targets in the grid. Computed tomography scan revealed a large defect in the left zygomatic and sphenoid bones, with a redundant loop of the lateral rectus muscle (Figure 5). The left medial rectus was 36.0 mm long, compared with a right medial rectus of 42.7 mm. The left lateral rectus was 43.8 mm long, compared with a right lateral rectus of 47.7 mm. Sagittal reconstructions of the orbital images could not be obtained, making it impossible to measure accurately the length of the vertical recti.

**DISCUSSION**

Postsurgical enophthalmos may occur following orbitozygomatic pterional craniotomy (McDermott et al., 1990). The lateral wall and roof of the orbit are removed to gain access to the skull base in the parasellar region and middle cranial fossa. If the orbital walls are not adequately reconstructed, there is a net increase in orbital volume. The globe becomes displaced posteriorly and the rectus muscles shorten (Figure 5). In our patient, this produced a distinct pattern of diplopia, which has not been well described in association with enophthalmos.

Shortening of the rectus muscles in enophthalmos is similar to surgical recession, although the insertion of the muscle on the globe is unchanged. The primary effect of recession surgery is to weaken the muscle in its field of action, but the exact mechanism is not understood (Kushner, 2006). Contraction of the antagonist muscle following recession surgery may also change...
muscle dynamics. Most investigators cite a change in the position of the muscle on the length-tension curve as the primary source of muscle weakening (Beisner, 1971; Clement, 1987).

The degree of overlap between actin and myosin filaments determines the maximum tension generated by a muscle. When a muscle is contracted maximally, overlap of filaments impedes further shortening. This flattens the relationship between muscle length and tension. Decreased tension generation has been demonstrated experimentally in mammalian skeletal muscle (Bahler et al., 1968). Shortening of an eye muscle (by posterior displacement of the insertion or enophthalmos) may reduce force generation via a similar mechanism.

Robinson and colleagues (1969) used strain gauges intraoperatively to measure the tension of the lateral rectus muscle fixed isometrically at various lengths. Innervation to the muscle was varied by having the patient fixate on targets with the unoperated eye. With the unoperated eye in primary position, the resting tension of 19 g was reduced to 4 g when the lateral rectus was shortened by 8 mm. Using a similar approach, Collins and coworkers (1975) plotted length-tension curves by recording muscle tension with miniature in-series strain gauges during strabismus surgery at various static positions. Dynamic, continuous length-tension recordings have been reported by Simonsz (1994).

From the length-tension curves of Collins, the tension in the medial rectus of an eye moving from a nasal to temporal direction can be inferred (Figure 6). An approximate 6 mm decrease in muscle length, as in our patient, results in decreased tension in all positions of gaze. The decreased tension generated by the muscle limits the ocular rotation that can be achieved. Diplopia occurs because there is a mismatch of force generated by yoke muscles in each orbit. Thus, as our patient attempts right gaze, the normal right lateral rectus muscle generates more force relative to the shortened left medial rectus muscle. This discrepancy in force generation increases as a function of gaze angle. Near primary gaze, the difference in force generation by yoke muscles is relatively small (Figure 7). Presumably, the patient can adjust innervation levels independently in each orbit to maintain fusion. However, when he looks more than 10-15 degrees from primary gaze (except downgaze), it is no longer possible to compensate for the escalating difference in force generated by yoke muscles.

Interestingly, orthotropia in primary gaze is maintained, presumably because the medial rectus and lateral rectus muscles in the same orbit are affected in a similar fashion. The reduction in force generation is fairly symmetrical and off-setting, preserving eye alignment near primary gaze.

Although immediate changes in the length-tension properties of extraocular muscles have been measured intraoperatively, data after strabismus surgery have not been reported in humans. In rabbits, histological studies have shown atrophy of muscles and their antagonists following surgical recession (Christiansen et al., 1992). Scott (1994) examined monkey extraocular muscles after fixing the globe in abduction for several months. In the lateral rectus, there was no change in sarcomere length despite shortening of the muscle, implying that fibers can reorganize to take up slack by eliminating sarcomeres. It is unknown if such changes occur in rectus muscles from enophthalmos.

In our patient, the pattern of diplopia was quite symmetrical. This suggests a process affecting all the muscles from a change in the volume of the orbit. In the “sunken eyes, sagging brain” syndrome there is marked enophthalmos without orbital trauma, injury, or

![FIGURE 6 Length-tension curves of human extraocular muscle adapted from Collins et al. (1975). The left eye was fixed in seven positions, indicated on the x-axis. The right eye fixated on targets at seven positions indicated on the y-axis (innervation EOMs). Red line indicates the predicted tension in the left medial rectus tracking a target from nasal to temporal gaze.](image1)

![FIGURE 7 Hypothetical plots of muscle tension in the horizontal rectus muscles versus eye position for normal right orbit and enophthalmic left orbit. Red line indicates normal right lateral rectus, blue line indicates slack left medial rectus in right gaze. Tan line indicates normal right medial rectus, purple line indicates slack left lateral rectus in left gaze. Single vision is maintained up to approximately 15° (green shading), beyond which differences in tension between yoke muscles cause diplopia.](image2)
inflammation (Hwang et al., 2011). Surgical overshunting of cerebrospinal fluid results in expansion of the bony orbit with progressive enophthalmos. Shortening of the eye muscles in these patients can give rise to limited ocular movements and diplopia. In the silent sinus syndrome, shrinkage of the maxillary sinus causes orbital expansion. Diplopia associated with the silent sinus syndrome has been described in 28% of patients (Numa et al., 2005). It may be due to shortening of the rectus muscles from enophthalmos.

In our patient, it was not known if a restrictive process contributed to diplopia because forcedduction testing was not conducted. Enophthalmos would shorten all the eye muscles except the superior oblique. Lengthening of this muscle or a change in its angle of insertion might explain the relative preservation of fusion in downgaze.

Abnormal eyelid-globe apposition, pulsatile proptosis, and a cosmetically unsatisfactory appearance are known sequelae of incomplete orbital wall restoration after orbitotomy. Diplopia is an additional complication of this surgery, and underscores the importance of careful orbital reconstruction. We suggest that if orbital volume is enlarged substantially, the resulting enophthalmos can produce a characteristic syndrome of diplopia on eccentric gaze. Its main feature is a symmetrical pattern of limited ocular excursions, relatively sparing downgaze, from shortening of the rectus muscles.

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