Case Report

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Combined superior oblique palsy and ipsilateral Brown's syndrome after a closed-head trauma: A case report and brief review

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Abstract:

Closed-head trauma is a well-recognized etiology of acquired superior oblique (SO) palsy. However, combined SO palsy and ipsilateral Brown's syndrome after a closed-head trauma is rare. We describe a patient with this ocular motility disorder and conduct a brief review of the literature.

Keywords:

Brown's syndrome, closed-head trauma, diplopia, superior oblique palsy

Introduction

Combined superior oblique (SO) palsy and ipsilateral Brown's syndrome is a rare condition. This ocular motility disorder, referred to as the "canine tooth syndrome," often results from direct damage to the trochlea and SO tendon caused by dog bites. We report the clinical manifestation and treatment of a patient with this condition after a closed-head trauma, but without evidence of direct injury to the trochlear area. We conducted a brief review and compared our case with similar cases reported in the literature.

Case Report

A 49-year-old woman sustained a closed-head trauma in a traffic accident. Brain computed tomography (CT) in the emergency room showed subarachnoid hemorrhage over the left frontal region, quadrigeminal cistern, and along the Sylvian fissure, cerebral falx, and left cerebellum. No significant orbital lesion was noted on CT images. During admission, no consciousness deterioration at the acute stage was observed. However, the patient complained of persistent vertical diplopia. She was then referred to an ophthalmologic clinic after discharge. Her best-corrected visual acuity was 20/20 in both eyes. External eye examinations were normal for both the eyes. Worth's 4-dot test showed vertical diplopia. The Parks three-step test showed a typical pattern of right SO palsy. The double Maddox rod test showed a 9° excyclotorsion, which was further confirmed by fundus photography [Figure 1]. The diagnosis of the patient was traumatic right SO palsy. One month later, the strabismus pattern of right SO palsy was unchanged. However, a newly emerging right eye supraduction limitation, which was more significant in the adduction position, was noted. Both supraduction and infraduction of the right eye were limited in the adduction position [Figure 2]. Restriction due to right side orbital floor fracture with inferior rectus entrapment was highly suspected, but orbital CT showed intact orbital wall and floor in both the eyes.

The patient was observed for 3 months; right hypertropia improved slightly (from 7 prism diopter [PD] to 5 PD in the primary

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position), but the overall strabismus pattern remained unchanged. Strabismus surgery was recommended to eliminate diplopia. The results from the preoperative alternate prism cover test in the nine gaze positions as well as the head tilt position are shown in Figure 3. We performed right inferior oblique (IO) recession. Intraoperative forced duction test of the right eye showed significant restriction of supraduction in the adduction position, confirming the diagnosis of Brown's syndrome. The eye position was orthotropic, and no diplopia was noted postoperatively in the primary position, but right hypertropia in the downgaze position and mild right hypotropia in the upgaze position were unchanged [Figure 3]. Faden operation on the left inferior rectus was suggested to balance the incomitance in the downgaze position, but the patient refused to receive surgery on her nonlesion eye.

Discussion

Combined SO palsy and ipsilateral Brown's syndrome, limiting both supraduction and infraduction, especially in the adduction position of the lesioned eye, is a rare condition typically known as "canine tooth syndrome," because the most common etiology is the direct damage to the trochlea and SO tendon after a dog bite injury.[1-3] Our patient demonstrated similar ocular movement abnormalities after a closed-head trauma, but without the evidence of direct injury to the trochlea or SO tendon. The major differential diagnosis of this condition is orbital floor fracture after trauma. Both supraduction and infraduction limitation can be caused by inferior rectus restriction or injury after orbital floor fracture. However, orbital CT in our patient showed an intact orbital wall.

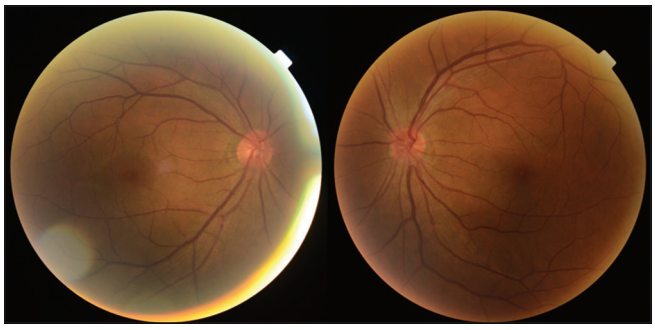

Figure 1: Fundus photography showed significant excyclotorsion, a characteristic of superior oblique palsy

Although SO palsy is a common etiology of vertical diplopia after a closed-head trauma, combined traumatic SO palsy and ipsilateral Brown's syndrome is rare. Few cases have been reported in the literature. A comparison between these reported cases is shown in Table 1.[4-6] Among the three cases that were closely followed immediately after the trauma, two of them manifested Brown's syndrome in a delayed time sequence (our case and the case reported by Schulz[5]); one of them had significant Brown's syndrome immediately after the accident (the case reported by Rowe *et al*.[4]). As for the outcome, two cases had spontaneous resolution of SO palsy; the other three had no complete resolution. None of them had complete resolution of ipsilateral Brown's syndrome. Those three cases who had no complete resolution of SO palsy had symptomatic diplopia at the primary position and finally received strabismus surgery.

Most of the reported cases showed no direct evidence of injury to the trochlea, SO tendon, or orbit on imaging; thus, the pathologic mechanism resulting in this rare condition is still questionable. Schulz postulated that the late-onset Brown's syndrome secondary to traumatic SO palsy was caused by a fibrotic reaction of the SO tendon or adjacent structures, which might be due to the inactivity of the SO tendon or due to concomitant indirect trauma.[5] Rowe *et al*. speculated that an injury in the patient's occipital area had induced a contrecoup injury at the orbit, causing a minor hemorrhage adjacent to the trochlea that resulted in an

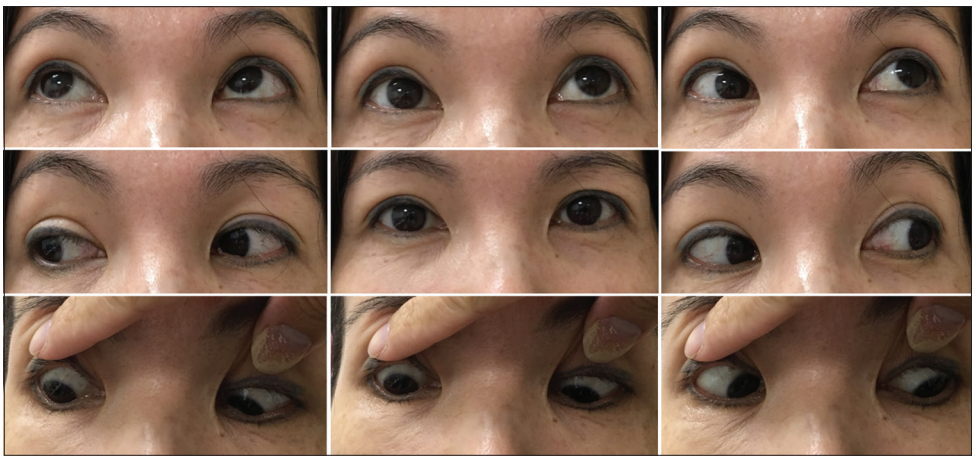

Figure 2: Preoperative photographs of the nine diagnostic gaze positions showed the significant limitation of both supraduction and infraduction of the right eye, especially in the adduction position

Table 1: A comparison between reported cases of combined superior oblique palsy and ipsilateral Brown's syndrome after closed-head trauma

Authors	Our case	Schulz	Schulz	Rowe *et al.*	Wong and Lim
Age	50	28	31	53	59
Gender	Female	Female	Male	Female	Male
Affected eye	Right	Left	Bilateral	Left	Left
Head trauma	Closed-head trauma with SAH	Closed-head trauma	Closed-head trauma with frontal bone fracture, SAH, and ventricular hemorrhage	Closed-head trauma at the occipital area	Closed-head trauma with left superomedial orbital wall fracture
Image-proved trochlear injury	No	No	No	No	Yes
Timing of Brown's syndrome present after trauma	1 month	3 months	Unknown	Immediately after trauma	Unknown
Spontaneous resolution of Brown's syndrome and its timing	No	No	No	No	No
Spontaneous resolution of SO palsy and its timing	No	Yes 2 months	No	Yes 2 months	No
Strabismus surgery	Right IO recession	No	Right SR recession	No	Right IR recession and Faden operation

SO=Superior oblique, SAH=Subarachnoid hemorrhage, IO=Inferior oblique, SR=Superior rectus, IR=Inferior rectus

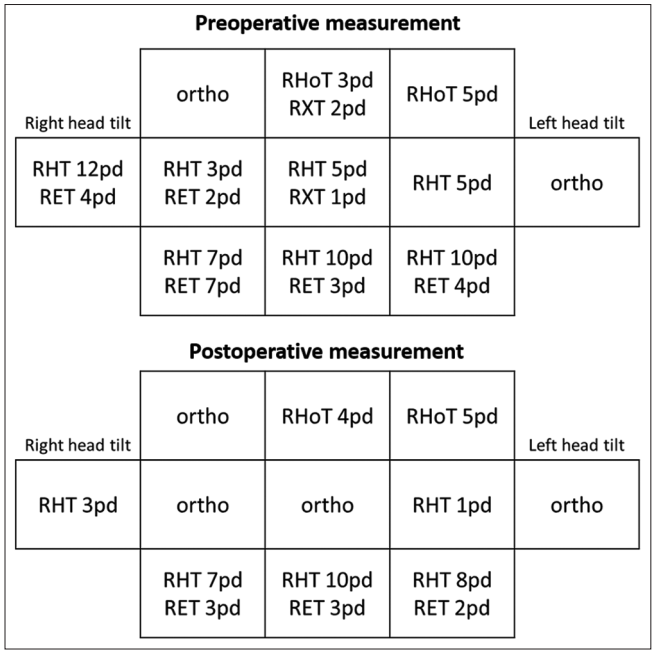

Figure 3: Alternate prism cover test of the nine diagnostic gaze positions and the two head-tilting positions before and 1 month after the strabismus surgery (right inferior oblique recession). pd = Prism diopter, ortho = Orthophoria, RHT = Right hypertropia, RHoT = Right hypotropia, RET = Right esotropia, RXT = Right exotropia

immediate Brown's syndrome.[4] The course of our patient was similar to that reported by Schulz. In our patient, we also speculated that the SO palsy resulted from damage to the fourth cranial nerve due to the closed-head trauma; the late-onset ipsilateral Brown's syndrome resulted from a fibrotic reaction after a concomitant indirect contusion to the trochlear region.

The strategy of strabismus surgery should be carefully considered in such a rare condition. If the manifestation of SO palsy was mainly SO underaction, rather than IO overaction, as in the case of our patient, the strengthening procedure of SO (e.g., SO tucking) is superior to the weakening procedure of IO (e.g., IO recession).[7] However, since ipsilateral Brown's syndrome was intraoperatively confirmed by the forced duction test, SO tucking was avoided because this procedure would inevitably worsen the existing restriction. Therefore, alternative options to correct the diplopia were ipsilateral IO weakening procedure or contralateral inferior rectus recession. Wong and Lim performed a contralateral inferior rectus recession plus the Faden operation in a similar case and achieved a favorable outcome.[6] This procedure corrected both hypertropia at the primary position and the incomitance of downgaze. However, our patient refused surgery for her nonlesion eye. Therefore, we performed an ipsilateral IO recession. This procedure eliminated the patient's hypertropia and vertical diplopia at the primary position without worsening the severity of the ipsilateral Brown's syndrome but had no corrective effect on the hypertropia at the downgaze position.

In summary, combined SO palsy and ipsilateral Brown's syndrome may occur after a closed-head trauma. Detailed examination, careful differential diagnosis, and special considerations in surgical correction are necessary in the management of such a rare condition.

Ethic approval

IRB information: Far Eastern Memorial Hospital (FEMH), Approval No. 108141-C. This case report was conducted in accordance with the Declaration of Helsinki. Informed written consent to publish data and images was obtained from the patient described in this case report.

Declaration of patient consent

The author certifies that he has obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands

that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

The author declares that there are no conflicts of interests of this paper.

References

1. Knapp P. Classification and treatment of superior oblique palsy. Am Orthopt J 1974;24:18-22.
2. Lee WB, O'Halloran HS. A report of canine tooth syndrome. Orbit 2004;23:53-7.
3. Legge RH, Hedges TR 3rd, Anderson M, Reese PD. Hypertropia following trochlear trauma. J Pediatr Ophthalmol Strabismus 1992;29:163-6.
4. Rowe F, Ramasamy B, Noonan C. Canine tooth syndrome following occipital impact closed head injury. Neuroophthalmology 2009;31:23-7.
5. Schulz E. Spontaneous consecutive brown syndrome following superior oblique palsy. Strabismus 1993;1:11-6.
6. Wong CW, Lim Z. A variant of canine tooth syndrome – Presentation and management. Strabismus 2014;22:18-20.
7. Bhola R, Velez FG, Rosenbaum AL. Isolated superior oblique tucking: An effective procedure for superior oblique palsy with profound superior oblique underaction. J AAPOS 2005;9:243-9.