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Case Report

역설머리기울임을 보인 선천상사근마비 환자의 수술 결과

Surgical Outcome for Congenital Superior Oblique Palsy with Paradoxical Head Tilt

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Purpose: We report the surgery outcome of three cases with congenital superior oblique palsy (SOP) with paradoxical head tilt after conducting ipsilateral inferior oblique (IO) recession.
Case summary: Case 1 was a 4-year-old boy with right head tilt noted since about 3-4 months of age. He demonstrated right hypertropia of 6 prism diopters (PD). Case 2 was a 4-year-old girl who demonstrated left hypertropia of 6 PD and exotropia of 18 PD. She had shown intermittent left head tilt since about 3 months of age. Case 3 was a 8-year-old boy with right head tilt noted since about 1 year of age. He demonstrated right hypertropia of 10 PD. Case 1 underwent left IO recession and left lateral rectus recession. Case 2 and 3 underwent right IO recession.
Conclusions: Ipsilateral IO weakening was also the effective surgery in congenital SOP with paradoxical head tilt as well as in the one manifesting usual head tilt.
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Key Words: Head tilt; Superior oblique palsy; Surgery

Superior oblique palsy (SOP) is the most frequent isolated paresis of the cyclovertical muscles.[1-4] The usual clinical presentation of SOP is a head tilt away from the affected eye along with a chin depression posture. However, some patients may tilt toward the ipsilateral side with the SOP and the underlying mechanism of paradoxical head tilt remains unclear.[4-6] von Noorden et al[7] reported in 270 patients with SOP, only seven patients (3.4%) showed head tilt toward paralyzed side. Surgeons tend to confuse the situation and even think the other paresis of the cyclovertical muscles because the phenomenon is very rare in SOP and hesitate to perform their own routine surgery even though they are suspicious of SOP associated with paradoxical head tilt. We report the surgical outcome of three cases with congenital SOP with paradoxical head tilt after conducting routine ipsilateral inferior oblique (IO) muscle weakening surgery.

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CASE REPORT

1. Case 1

A 4-year-old boy visited Korea University Hospital because of a right head tilt since about 3-4 months of age (Fig. 1A).

On alternate prism cover test for distance, right hypertropia (HT), measuring 6 prism diopters (PD) was observed in the primary position. And the degree of HT increased to 14 PD on left gaze but was reduced to 2 PD on right gaze (Fig. 1B). Head tilt to the right resulted in 5 PD of right HT, while head tilt to the left result in 6 PD of right HT. Indirect ophthalmoscopic findings showed excyclotorsion in the left eye. IO overaction and superior oblique (SO) underaction in the right eye were noted (Fig. 2A). An impression of congenital SOP with paradoxical head tilt was formed, and we decided to conduct surgery. He underwent 14 mm recession of right IO. Postoperatively, he showed orthotropia, straight head posture and improved extraocular movements (Fig. 2B).

B

	5PD RHT	
2PD RHT	6PD RHT	14PD RHT
	3PD RHT	

Right head tilt
8PD RHT

Left head tilt
5PD RHT

Figure 1. (A) A 4-year-old boy with a right head tilt. (B) Angle of deviation at nine cardinal gazes and head tilt. PD = prism diopters; RHT = right hypertropia exotropia.

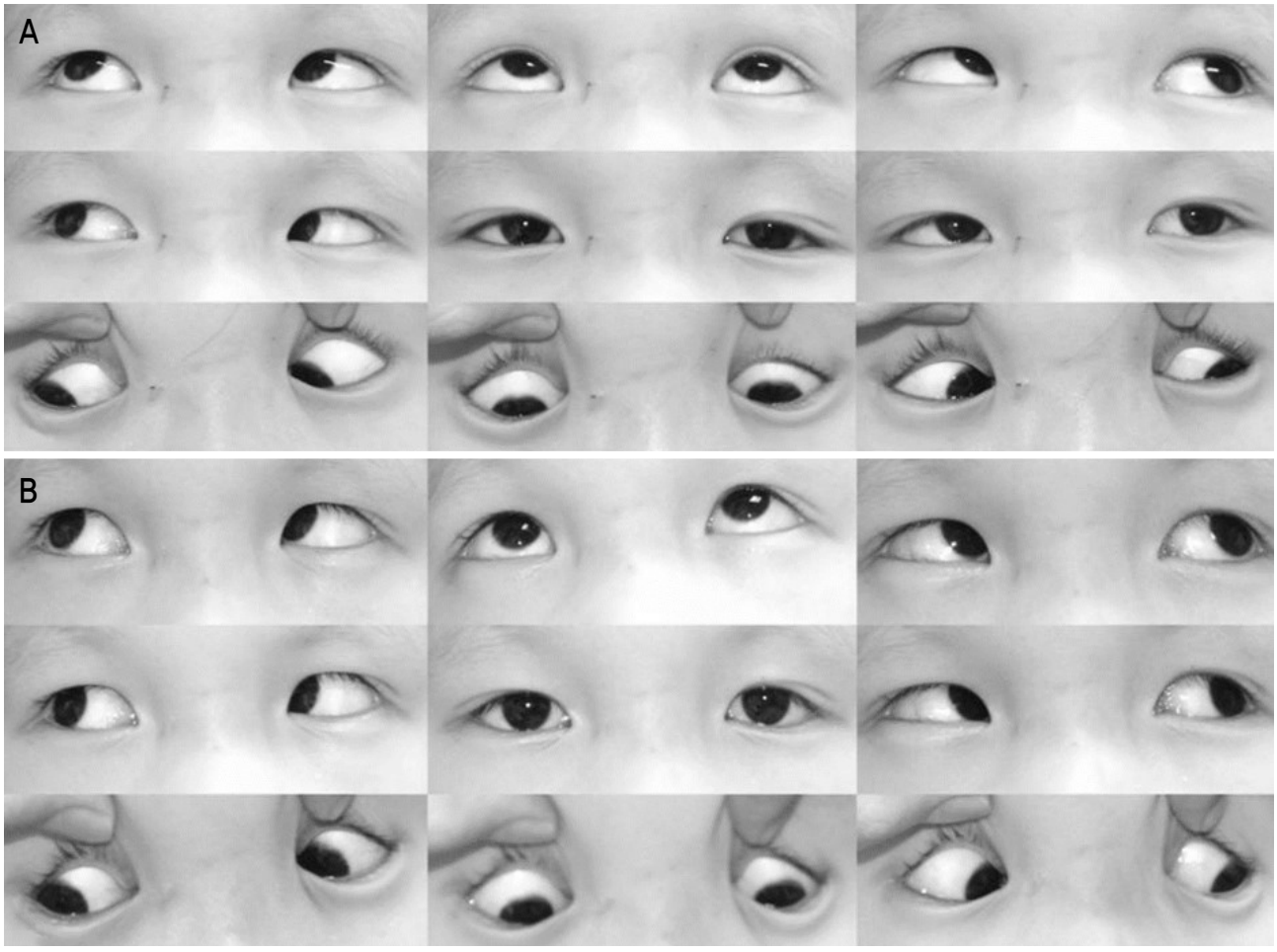

Figure 2. (A) Preoperative eye movement at nine cardinal gazes. (B) Postoperative eye movement at nine cardinal gazes.

2. Case 2

A 4-year-old girl was referred because of an intermittent abnormal head position noted since about 3 months of age and upward deviation since a year ago. On exam, she had an uncorrected visual acuity of 20/20 in both eyes. She had shown left head tilt and right face turn since about 3 months of age intermittently, which was confirmed by her old photograph.

In primary position at distance, she demonstrated a 6 PD of left HT and 18 PD of exotropia. And the degree of HT increased to 8 PD on right gaze but was reduced to 4 PD on left gaze. Head tilt to the left resulted in 8 PD of left HT, while head tilt to the right result in orthotropia. And version at 9 cardinal gaze was consistent with left SOP. Indirect ophthalmoscopic findings showed significant excyclotorsion in the left eye. Surgery was scheduled to reduce the torticollis and HT. A 14 mm of IO recession and left lateral rectus recession were performed. A month postoperatively, no head tilt was noted and there was orthotropia in primary gaze. Postoperatively, she demonstrated equal fixation, maintained orthotropia, rarely exhibited torticollis and improved extraocular movements.

3. Case 3

An 8-year-old boy visited Korea University Hospital because of a right head tilt noted since about 1 year of age. He tilted his head to the right side about 10-15 degrees. On alternate prism cover test for distance, 10 PD of right HT and esotropia of 4 PD were observed in the primary position. And the degree of HT increased to 25 PD on left gaze but was markedly reduced to orthotropia on right gaze. Head tilt to the right resulted in 14 PD of right HT, while head tilt to the left result in 4 PD of right HT. Indirect ophthalmoscopic findings showed excyclotorsion in both eyes. IO overaction and superior oblique underaction in the right eye were +3 and -2 respectively. An impression of congenital SOP with paradoxical head tilt was formed, and we decided to conduct surgery. He underwent right IO recession 14 mm. Postoperatively, he demonstrated orthotropia, exhibited no torticollis and improved extraocular movements.

DISCUSSION

Previously reported unilateral SOP patients with paradoxical head tilt had unstable and intermittent fusion when the head was tilted toward the uninvolved eye, and experienced alternating suppression or diplopia when holding the head toward the paralyzed eye.[1,3] Such patients preferred a head position that disrupted fusion, caused wide image separation to eliminate the discomfort that may have been associated with the constant effort to maintain single binocular vision.[5,7]

Recently, Khan[8] suggested ipsilateral head tilt during fixation with the affected eye in abduction and supraduction allowed a balance between dynamic compensatory counter-rolling and subsequent anticompensatory torsional saccades that was optimal for binocular vision. We think if the difference in degree of HT between right and left head tilt is big, paradoxical head tilt for wide image separation initially can be hard to try because of much discomfort. So SOP patients showing small difference in degree of HT between both sides are tend to present paradoxical head tilt. Our cases support this hypothesis. Case 1, 2, and 3 showed small differences less than 10 PD. And also with the paradoxical head tilt position, ipsilateral superior rectus muscle contracted may allow incyclotorsion compensatory to excyclotorsion.[8] Although these theory explains how the paradoxical head tilt occurs, the underlying mechanism and predisposing factors to paradoxical head tillt are not clear. Further researches of the clinical characteristics with large patients are warranted to elucidate the mechanism of paradoxical head tilt in SOP.

In this study, HT markedly decreased and head tilt disappeared after the routine ipsilateral IO weakening surgery irrespective of direction of head tilt, which means this condition is curable and not contrary to the routine surgical outcome of SOP when the examiner is aware of its existence. In conclusion, we present three cases of SOP with paradoxical head tilit, who underwent surgeries involving IO recession as in the one manifesting usual head tilt, and finally achieved excellent surgical outcomes.

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