Case Report

Reduction of Chronic C1-C2 Rotatory Subluxation using Continuous Cervical Halter Traction: A Case Report and Literature Review

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Objective: The purpose of this study was to report on a rare case of progressive chronic rotatory subluxation of C1-C2 (Fielding and Hawkins type III) which was successfully treated non-surgically and to provide a review of the related literature.

Materials and Methods: Retrospective case report

Results: The authors report on a patient who presented with persistent progressive cervical rotation following 5 months of partial treatment. C1-C2 reduction, demonstrated by the symmetry of the lateral atlantodental interval (ADI)in radiographs of the C-spine in open mouth view, was achieved after 3 weeks of continuous halter traction. Following that procedure, 4-poster bracing was used to immobilize the neck for 3 months. The stabilization of the neck permitted normal range of motion.

Conclusion: Based on this case,the authors suggest full time continuous halter traction is an option for reduction and 4-poster brace is an option for immobilization of chronic atlantoaxial rotatory subluxation in cases with no facet joint deformity.

Keywords: Chronic C1-C2 rotatory subluxation, Grisel syndrome, Reduction, Continuous halter traction

J Med Assoc Thai 2018; 101 [Suppl. 3]: S263-S267
Full text. e-Journal: http://www.jmatonline.com

A10-year-old boy presented with progressive head deviation to the left side of 5 months duration associated with mild neck pain and difficulty turning his head to the right. He had no history of upper respiratory tract infection, neck/nasopharyngeal surgery, or trauma. He had previously received medication from a private clinic and had been referred to the Rehabilitation Department, Lerdsin Hospital for cervical traction every weekday for 2 months, but was still unable to turn his head to the right without pain.

Physical examination revealed torticollis without sternocleidomastoid spasm, as well as cock-robin posturing (head tilting, neck rotation). It was not possible to accurately palpate the C2 spinous process. His head rotation to right was restricted due to pain. Open mouth X-ray images showed lateral tilting of the cervical spine. CT scans confirmed the lateral tilting of the spinous processes and the rotation of the atlantoaxial joint with asymmetry on the lateral sides and no detectable abscess. The atlanto-dens interval measured on the lateral cervical spine X-ray was 7 mm.

The patient was admitted and treated with fulltime continuous halter traction at 5 lbs. and non-steroidal anti-inflammatory drugs. After 2 weeks of traction, the clinical appearance had improved, with a lateral ADI of 1.6 mm on the right side and 8.6 mm on left side. After the 3rd week of traction, we discontinued the halter traction and immobilized the patient with 4-poster bracing for 3 months. At the end of that period, he had achieved full recovery of neck motion and normal

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How to cite this article: Kunakornsawat S, Wasinpongwanich K, Pluemvitayaporn T, Pruttikul P, Piyakulkaew C. Reduction of Chronic C1-C2 Rotatory Subluxation using Continuous Cervical Halter Traction: A Case Report and Literature Review. J Med Assoc Thai 2018;101;Suppl.3:

ADI measurement on follow-up C-spine X-rays.

Discussion

The normal human cervical spine can rotate up to 90 degrees; approximately 60% of the rotation is from the atlantoaxial joint combined with the horizontal C1-C2 facet's joint alignment[1,2]. The stability of the atlantoaxial joint is optimized by the C1-C2 capsular ligaments and two main ligamentous structures. The transverse ligament maintains the odontoid opposed to the anterior arch of C1 and prevents the anterior translation of the atlas on the axis. Together with pairs of alar ligaments which are bilaterally attached from the tip of the odontoid process to the occipital condyles, they also prevent excessive anterior subluxation of this joint[1,2]. Atlantoaxial joints in children are easily subluxated because of weakness of the ligament and joint capsule, horizontal joint articulation alignment, and the relatively small size of the supporting muscles[3].

Chronic atlantoaxial rotatory subluxation of C1-C2 is defined as the persistence of symptoms for longer than 2 to 3 months[4,5]. The longest chronicity that had been reported was by Ishii et al[5] at 8.8 months. The incidence of atlantoaxial rotatory subluxation of C1-C2 is rare. In the literature[6], Philips et al reported on a review of 23 cases over a period of 12 years between 1975 and 1986. Similarly, Mifsud, in a review covering 25 years between 1988 and 2014, found only 12 cases[7].

A study by Subach et al[8] reported that most chronic C1-2 rotatory subluxation cases (35%) had a

Figure 1. Photo taken by the patient's mother when the symptoms initially appeared showing the cock-robin posture (head tilting, neck rotation).

Figure 2. Photo taken at the time of admission. Head tilting and neck rotation had progressed and the open mouth X-rays showed rotation of the atlantoaxial joint and asymmetric lateral ADI.

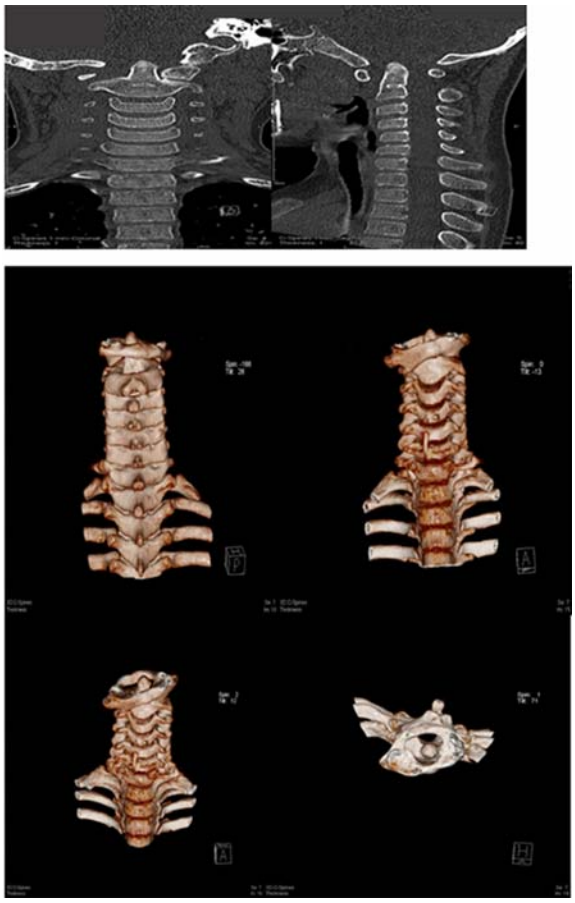

Figure 3. CT scans showed lateral tilting of the spinous process and rotation of the atlantoaxial joint with asymmetry on lateral sides. No C2 facet deformity was detected. In a CT 3D reconstruction, the ADI was measured at 7 mm, a Type III Fielding and Hawkins classification of atlantoaxial rotatory fixation.

history of otitis media or pharyngitis, 20% had undergone head or neck surgery, and 20% appeared after trauma, while 25% had no determined cause[8]. Atlantoaxial rotatory subluxation also occurs in congenital conditions that involve ligamentous laxity such as Down syndrome, Morquio syndrome, and Marfan syndrome[9,10]. Kawasak disease is also related to atlantoaxial rotatory subluxation[11]. Other possible causes of torticollis can be distinguished from atlantoaxial rotatory subluxation, e.g., congenital torticollis due to sternocleidomastoid spasm, congenital anomalies, bone tumor, fracture, infection of the cervical spine or pathological process of the spinal cord and brainstem such as syringomyelia, Arnold-Chiari malformation, and unilateral palsy of trochlear nerve. Torticollis posture can also cause symptoms of diplopia[6].

Atlantoaxial rotatory subluxation typically presents with acquired torticollis[6] as well as cervical pain, restriction of neck motion and cock-robin posturing. There are three clinical signs that can help to diagnose atlantoaxial rotatory subluxation. The first is Sudek's sign, a palpated C2 spinous process deviated to the same side as the head rotation, suggesting an attempt to reduce the deformity. The second sign is a palpable sternocleidomastoid muscle on the same side as the head rotation[6]. The third sign is the inability to rotate the head past the midline in the direction opposite to the lesion[12].

With this patient, the cause of the atlantoaxial rotatory subluxation was considered undetermined because the patient had no history of upper respiratory tract infection, trauma, previous surgery of the head and neck, nor were there any clinical signs indicating any of the syndromes mentioned above. This patient presented with the typical cock-robin and torticollis postures. We found only the second and third signs mentioned above.

The Fielding and Hawkins system[4] classifies atlantoaxial rotatory subluxation into 4 different types based on axial CT imaging of the C1 and C2 vertebrae. According to that classification system, this case was type III: unilateral facet subluxation with anterior displacement of more than 5 mm. More recently, Pang and Li[13] have developed another classification system for atlantoaxial rotatory subluxation based on dynamic CT scans. The latest classification system was established by Ishii K et al[5]. based on the lateral inclination of the atlas on the axis and on facet deformity using 3D CT reconstruction modeling. We did not

Figure 4. After 3 weeks of halter traction followed by 3 months of the 4-poster brace, clinical appearance and X-rays demonstrated the neutral position of the head, indicating reduction of atlantoaxial joint.

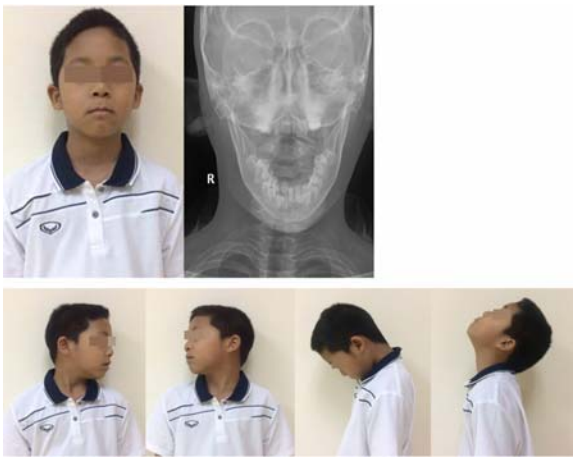

Figure 5. Range of motion of the cervical spine was normal at the 3 month follow-up.

see a C2 facet deformity in this case, and the C1 lateral inclination appeared to be less than 20°, a Grade I classification under the Ishii system.

Because chronic atlantoaxial rotation is quite rare, that treatment is still controversial because it depends on classification type[4], duration of the symptoms before presentation[6-8], and the nature of the C1-C2 facet deformity[5] as well as the compliance of patients and the patient's parents[16].

Unlike acute atlantoaxial rotatory subluxation, which can be spontaneously reduced by simple soft collar immobilization in most cases[3,4,8,15], if there is no improvement within 3 to 4 weeks, skull traction or surgery is considered. For chronic atlantoaxial rotatory subluxation, there is no clear consensus about the treatment, e.g., reduction method, period of immobilization, type of orthosis for immobilization. In the literature, the reported success rate of conservative treatment ranges from 70 to 100%[5-8,16,17]. The factor that predicts failure of reduction is the duration of subluxation before reduction[6-8], e.g., more than 3 weeks in the Subach study[8]. Many studies advocate that if chronic or recurrent atlantoaxial subluxation persists longer than 2 weeks there is a need for further intervention and that in 30% of the cases where atlantoaxial subluxation persists, surgery is required[8]. There is still controversy regarding chronic atlantoaxial subluxation treatment. Park et al reported successful reduction of a 9-year-old girl who had had symptoms of atlantoaxial subluxation for two months. They used 5-lb weight halter traction for 6 weeks followed by bracing for 4 months and intermittent collar use for another 2 months[18]. Hsu et al reported the case of a 7-year-old girl conservatively treated for chronic atlantoaxial rotatory subluxation by intermittent halter traction followed by a rehabilitation program for 4 months[19]. Mifsud described successful reduction in 12 cases with at least 4 weeks of symptoms who were treated with halo traction followed by halo vest or Minerva jacket[7]. Akbay[16] reported using a closed manual reduction maneuver in 12 Hawkins type I and type II patients maintained by a SOMI brace and Philadelphia collar.

In the present case, the patient was treated with intermittent cervical traction, a rehabilitation program 5 days a week, and medication for 2 months, but showed no improvement. Careful clinical and radiological evaluation of the patient revealed no neurological deficits, no facet deformity, and only mild C1 lateral inclination. We then decided to try conservative reduction of the joint. Continuous 5-lb halter traction was started and non-steroidal anti-inflammatory drugs were continued. Clinical evaluations were done daily. There was gradually improvement in reduction of pain and an increase in range of motion. After 2 weeks of traction, lateral ADI was 1.6 mm on the right side and 8.6 mm on the left side. After the 3rd week of traction the neck had achieved a near total recovery to normal with no pain, so we discontinued the halter traction and immobilized the neck with a 4-poster brace for an additional 3 months. The patient achieved full recovery of cervical motion and normal ADI as measured on the follow-up C-spine X-ray.

Reduction by full time continuous cervical halter traction has advantages over other methods because it is a noninvasive method of reducing C1-2 facet loading pressure in an absolute bed rest status and allows spontaneous reduction in cases where the patient has no facet deformity. Furthermore, the patient is not required to undergo general anesthesia or any manual reduction maneuvers that might result in additional injury to the ligamentous structures or otherwise disrupt the healing process. Compared with halo traction, halter traction results in fewer complications. Akbay[16] reported on one case of halo pin skull penetration after a fall. The most significant advantage of this method is greater preservation of the cervical rotation of the spine compared to fusion surgery.

Maintaining the reduction with a 4-poster brace has the advantage of restricting the range of motion of the cervical spine while avoiding potential pin-related complications related to the halo vest[7] and, importantly,achieving greater preservation of cervical rotation motion compared with surgical instrumentation and fusion. The disadvantage is the dependence on the compliance of both the patient and the patient's family and the fact that recurrence of the problem is possible[16].

In a study of 12 cases, Ishii et al evaluated the risk of recurrence after reduction by looking for C2 facet joint deformitieson 3D CT scans[17].The study found no C2 facet deformity and no recurrence at the latest follow-up.

Based on the current case, reduction by full time continuous halter traction should be considered before deciding on any surgical option for chronic atlantoaxial rotatory subluxation, especially inpatients with no facet deformity. The period of solid bracing after reduction should be determined individually for each patient.

Conclusion

Based on this case, we suggest full time continuous halter traction is an option for reduction and 4-poster brace an option for immobilization of chronic atlantoaxial rotatory subluxation with no facet deformity.

What is already known on this topic?

There is still controversy regarding chronic atlantoaxial subluxation treatment. The new classification and treatment has been developing lately.

What this study adds?

For chronic atlantoaxial subluxation with no facet deformity, the full time continuous halter traction and 4-poster brace should be considered as another option for reduction and immobilization before deciding on any surgical option.

Potential conflicts of interest

The authors declare no conflict of interest.

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