Treatment of Atlantoaxial Rotatory Fixation With Botulinum Toxin Muscle Block and Manipulation

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Slippage after reduction of atlantoaxial rotatory fixation (AARF) is usually treated with repeated cervical traction and brace immobilization. To date, no data have been published on the management of muscle spasm during treatment. Here, we describe the case of a 7-year-old girl with AARF for 1 month who visited our hospital for treatment. During physical examination, spasm of the sternocleidomastoid muscle was noted. The patient was treated with manipulative reduction, and slippage after reduction was managed with botulinum spasticity block of the sternocleidomastoid and splenius capitis muscles, and repeated manipulation. Cervical orthosis immobilization with a rehabilitation program of isometric contract–relax exercise for the neck was conducted for 3 months. The subject had full recovery from AARF at 1-year follow-up. This report demonstrates that, in selected cases of slippage after reduction from AARF, conservative management with manipulation under anesthesia is a good method, and the muscle components may play a crucial role in AARF. [*J Chin Med Assoc* 2010;73(4):222–224]

Key Words: atlantoaxial rotatory fixation, botulinum toxin, manipulation

Introduction

Sudden onset of acute torticollis is a rare condition in children and is usually diagnosed as atlantoaxial rotatory fixation (AARF), which includes conditions such as subluxation or dislocation.[1] It is usually symptomatic, but without neurological disturbance. Treatment procedures include medication such as anti-inflammatory drugs, hard-collar immobilization, and cervical halter traction with immobilization or C1–C2 fusion.[2–4] The major factor predicting the failure of conservative management is the duration of AARF before initial reduction. Patients with long-term AARF are more likely to experience recurrence and require surgery.[4] However, some complications of surgery have been reported, such as pseudoarthrosis, cranial nerve injury, postoperative redislocation, and difficulties in postoperative management.[5–7]

The mechanism that causes AARF is not clearly understood. Trauma, a history of previous upper respiratory infection, and simple muscle spasm have been described in the etiology of this condition.[4] Long-standing cases can have chronic ligamentous changes that cause recurrence of the subluxation, and require surgical fusion to restore alignment.[8]

We present here a case treated successfully using a conservative procedure with manipulation under general anesthesia, combined with botulinum toxin muscle spasticity block and cervical orthosis immobilization.

Case Report

A 7-year-old girl had acute torticollis and neck stiffness for 1 month. Her symptoms occurred suddenly after she went body surfing in a swimming pool. Although some nonsteroidal anti-inflammatory drugs were prescribed, the symptoms persisted. Clinical examination revealed that her neck tilted to the left side, and tightness of the left sternocleidomastoid muscle was found. There were no neurological signs or significant pain if she did not move her neck and head.

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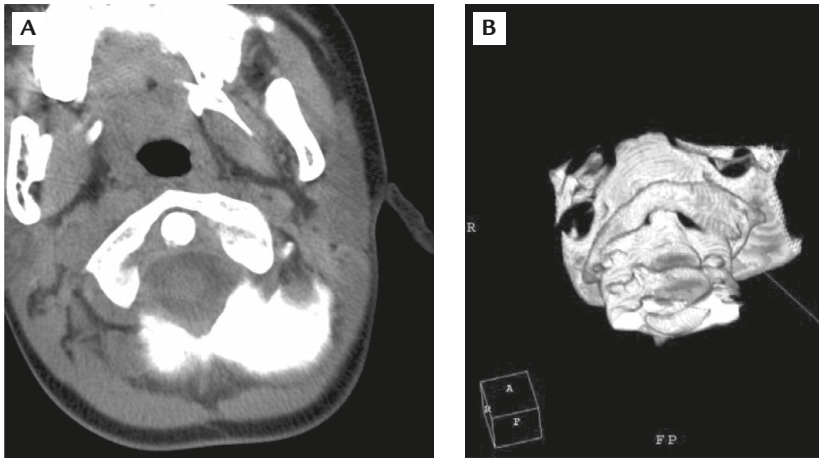

Figure 1. (A) Axial view of left C1–C2 subluxation with anterior displacement of 1.26 mm. (B) Inferior view of 3-dimensional computed tomography reconstruction.

Three-dimensional computed tomography (CT) was arranged, and the bony structure of the atlantoaxial area was reconstructed. The result showed the left atlantoaxial joint subluxation (Figure 1). According to the classification system of Fielding and Hawkins,[1] this case presented as an anterior subluxation of the left lateral articular process, which resulted in rotatory fixation with anterior displacement of < 3 mm. Type I AARF was thus diagnosed.

Manipulation under anesthesia was arranged. In the operating room, the patient was placed in the supine position, and general anesthesia with a muscle relaxant was administered. Manipulation with axial traction was applied to the occiput, as described previously.[9] Complete resolution was gained, but post-reduction slippage occurred on day 2. Tightness and spasm of the left sternocleidomastoid muscle was assumed to be the cause of this slippage.

Under anesthesia, manipulation was performed again smoothly. Botulinum toxin type A (Botox; Allergan Inc., Irvine, CA, USA) was reconstituted with 0.9% normal saline to a concentration of 5 U/0.1 mL. A total of 18 U (1.0 U/kg) was equally divided among 2 sites in the left sternocleidomastoid muscle and 1 site in the left splenius capitis muscle immediately after reduction, as described previously.[10] On the second day, normal neck and head motion was noted. The injected muscles did not show much difference during palpation. The Guilford type of cervical orthosis (2-poster design) and a rehabilitation program of isometric contract–relax and hold–relax exercises for bilateral neck muscles were conducted for 3 months. Muscle relaxant was not given. The patient had full recovery from AARF at 3 months of follow-up and there was no recurrence at 6 months and 1 year.

Discussion

Manipulation under anesthesia is a quick way to treat upper cervical subluxation in pediatric patients.[9,11] In the present case, the time from onset of subluxation to the patient's acceptance of manipulation was 1 month, with noted spasm of the left sternocleidomastoid muscle. According to the classification of Pang and Li,[4] AARF can be stratified by chronicity of pretreatment delay into acute (≤ 1 month), subacute (1–3 months) and chronic (≥ 3 months). Sternomastoid spasm was noted on the chin side in more than half of their cases, which could shift to the side of the lateral head tilt in the chronic phase. Some special aspects of children's spines can predispose them to atlantoaxial rotatory subluxation. Fesmire and Luten have described the developmental anatomy of children, and have pointed out that their ligaments and joint capsules possess sufficient elasticity to allow hypermobility without disruption.[12] Kawabe et al have concluded that the dens-facet angle of the axis is steeper in children than in adults, and meniscus-like synovial folds are found in the C0/1 and C1/2 facet joints of the spines of children but not in those of adults.[13]

Subach et al have suggested that a second mechanism must exist to inhibit spontaneous reduction, such as spasm of the deep cervical musculature, inflammation of the synovium, or disruption of the joint capsule with fragments resisting motion and restoration of normal alignment.[3] Pang and Li have described a new classification for types of AARF with degree of severity measured by motion analysis and they have shown the worst result occurs in type I. However, they did not mention the role of muscle components.[4] In the present case, muscle spasm could have played a crucial role in the mechanism of AARF, and might have been

the cause of slippage of AARF on the second day. Under this assumption, botulinum toxin was used to reduce the spasm of the cervical musculature in the splenius capitis and sternocleidomastoid muscles. Hold–relax and contract–relax rehabilitation exercises were conducted to relieve the tightness of the neck muscles.

The radiographic diagnosis of atlantoaxial rotatory subluxation is usually difficult to establish using only conventional static imaging methods. Rinaldi et al have introduced dynamic CT to offer a more clinically relevant assessment of C1–C2 mobility.[14] Based on our previous experience and other studies, 3-dimensional CT could offer appropriate diagnostic imaging of the upper cervical spine.[9,11,15]

For acute and subacute cases, halter traction in conjunction with muscle relaxant and sedative agents, followed by cervicothoracic orthotic immobilization for 3 months might be the best treatment.[4] Based on our previous experience, patients gain immediate relief after manipulation under anesthesia without immobilization.[9,11] Post-reduction immobilization might not be important in acute AARF when treated with manipulation. In the present case, muscle spasm of the sternocleidomastoid muscle was noted, and after a second reduction with botulinum toxin injection, we did immobilize the patient. However, we could not clearly determine whether or not the post-reduction immobilization resulted in the resolution of her problem, or if the botulinum toxin injection accounted for the good result. This could be a limitation of our report.

Physicians agree that once AARF has been diagnosed, treatment should be undertaken as soon as possible. Subach et al have concluded that there would be no recurrence if reduction were achieved within 21 days of onset of symptoms.[3] Pang and Li also have shown a much higher number of slippages in the subacute group than in the acute group.[4]

In conclusion, manual reduction under anesthesia is a good, quick method of management for acute atlantoaxial subluxation and slippage. When there is muscle spasm involvement on the side of lateral head tilt, botulinum toxin for muscle spasm block could be the best treatment. Although we have shown that manipulation under anesthesia and post-reduction rehabilitation exercise are both effective, further investigations are needed to more fully determine their efficacy in treating AARF.

References

1. Fielding JW, Hawkins RJ. Atlanto-axial rotatory fixation. (Fixed rotatory subluxation of the atlanto-axial joint). *J Bone Joint Surg Am* 1977;59:37–44.
2. Smith MD, Phillips WA, Hensinger RN. Fusion of the upper cervical spine in children and adolescents: an analysis of 17 patients. *Spine* 1991;16:695–701.
3. Subach BR, McLaughlin MR, Albright AL, Pollack IF. Current management of pediatric atlantoaxial rotatory subluxation. *Spine* 1998;23:2174–9.
4. Pang D, Li V. Atlantoaxial rotatory fixation: part 3-a prospective study of the clinical manifestation, diagnosis, management, and outcome of children with atlantoaxial rotatory fixation. *Neurosurgery* 2005;57:954–72.
5. Smith MD, Phillips WA, Hensinger RN. Complications of fusion to the upper cervical spine. *Spine* 1991;16:702–5.
6. Dickman CA, Sonntag VK. Surgical management of atlantoaxial nonunions. *J Neurosurg* 1995;83:248–53.
7. Pinches E, Thompson D, Noordeen H, Liasis A, Nischal KK. Fourth and sixth cranial nerve injury after halo traction in children: a report of two cases. *J AAPOS* 2004;8:580–5.
8. Phillips WA, Hensinger RN. The management of rotatory atlanto-axial subluxation in children. *J Bone Joint Surg Am* 1989;71:664–8.
9. Tsai SW, Chou CS. A case report of manipulation under anesthesia of post-traumatic type II occipital-atlantoaxial rotatory subluxation in a 4-year-old girl. *J Manipulative Physiol Ther* 2005;28:352–5.
10. Liu YC, Fuh JL, Chen RC, Lin KP, Wang SJ. Botulinum toxin type A in the prophylactic treatment of transformed migraine in Taiwanese patients: A review of 30 consecutive cases. *J Chin Med Assoc* 2007;70:535–40.
11. Tsai SW, Zhong JD, Chen YW, Wu SK, Lin YW. Treatment of upper cervical subluxation in pediatric patients. *Man Ther* 2009; 14:448–51.
12. Fesmire FM, Luten RC. The pediatric cervical spine: developmental anatomy and clinical aspects. *J Emerg Med* 1989;7: 133–42.
13. Kawabe N, Hirotani H, Tanaka O. Pathomechanism of atlantoaxial rotatory fixation in children. *J Pediatr Orthop* 1989; 9:569–74.
14. Rinaldi I, Mullins WJ Jr, Delaney WF, Fitzer PM, Tornberg DN. Computerized tomographic demonstration of rotational atlanto-axial fixation. Case report. *J Neurosurg* 1979;50:115–9.
15. Nicholson P, Higgins T, Forgarty E, Moore D, Dowling F. Three-dimensional spiral CT scanning in children with acute torticollis. *Int Orthop* 1999;23:47–50.