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Treatment of Atlanto-Axial Rotatory Subluxation with Manipulation

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Infantile torticollis is a common condition in rehabilitation clinics. Acute torticollis due to atlanto-axial rotatory subluxation, however, is an uncommon condition in children. The diagnosis is usually delayed due to difficulty in the evaluation of roentgenograms, as well as to clinicians’ unawareness of this disorder. Delayed unrecognized atlanto-axial subluxation may make the treatment more complicated. Here, we review three cases who had atlanto-axial rotatory subluxation successfully treated with manipulation. (J Rehab Med Assoc ROC 2001; 29(1): 51 – 58)

Key words: atlanto-axial rotatory subluxation, manipulation, torticollis

INTRODUCTION

Acute torticollis resulting from atlanto-axial rotatory subluxation is an uncommon condition in children. The clinical presentation is an acute unilateral muscle spasm with typical head position rotated to one side and inclined to the contralateral side, or a “cock-robin” position. Treatment, if started early, is usually simple and the prognosis is good. Delayed diagnosis, however, may prevent complete restoration of atlanto-axial function [1].

CASE REPORT

There were four patients with the impression of atlanto-axial rotatory subluxation seen in our outpatient department from June 1996 to June 2000. Three of them are included in this retrospective review. One girl was excluded because her parents refused to accept our management and sought another doctor’s advice.

Case 1

A 9-year-old boy fell down accidentally when he was playing basketball and suddenly developed neck stiffness and torticollis. He was brought to our clinics for help on the next day. Physical examination revealed that his head was fixed in position inclined to the left side about twenty degrees, and rotated to the right side about forty-five degrees. The active range of motion (ROM) was limited. On examination of passive ROM, muscle-guarding sensation was noted. Nevertheless, neurological examination results were normal. Only local heat was felt on the right upper cervical spine. There was neither sore throat nor previous infection of head, neck or upper respiratory tract. Anterior open mouth (Fig. 1) and lateral plain views of cervical spine radiography were...
taken, which established the diagnosis of atlanto-axial rotatory subluxation.

Case 2

A 10-year-old boy developed neck stiffness with the neck fixed in the turned-to-right position when he woke up in the morning one month before he was brought to our clinics. He had visited some clinicians for help, and some medications, such as muscle relaxants and non-steroid anti-inflammation drugs, had been prescribed, but symptoms persisted. The physical examination showed the characteristic torticollis posture with head rotated to the right about forty degrees, and inclined to the left side about thirty degrees when rest. Compared to the right side, left facial muscles were wasting. The patient experienced sore and difficulty in passive range of motion. The neurological examination results were normal. There were neither previous infections such as pharyngitis, otitis media nor any trauma history except partial pancreactomy about 3 years earlier due to a traffic accident. Standard roentgenograms including anterior-posterior open mouth view (Fig. 3), straight lateral flexion and extension view, and computed tomography (CT) of cervical spine were carried out (Fig. 4). The diagnosis of atlanto-axial rotatory subluxation was established.

Case 3

A 7-year-old boy came to our clinics with the chief complaint of left upper cervical area swelling, pain and limited neck ROM for one week. An audible “click” sound was heard when he woke up in the morning before this visit. Some traditional herbal topical drugs had been tried for symptom relief, but in vain. On physical examination, the head was fixed in right rotated position with only some left rotation movement preserved. The movements of extension and left tilting of head were severely restricted. Local heat was found on left upper cervical area. There was no trauma history, or neurological deficit. The roentgenograms including anterior-posterior open mouth view, straight lateral flexion and extension view, and three dimensional computed tomography (Fig. 5) of cervical spine were carried out. Atlanto-axial rotatory subluxation was mostly impressed.

Fig. 1 In case 1, open mouth view shows that the left inferior articular facet of atlas is not articulating with the left superior articular facet of axis.
Fig. 2  In case 1, lateral flexion-extension view shows widening of the distance between atlas and the anterior surface of axis. The distance is measured as 4 mm.

Fig. 3  In case 2, open-mouth view shows that there is a distance discrepancy between the dens and the lateral masses of atlas. Some sclerosing change also can be seen between the articulating surface of left atlas and axis.

Fig. 4  Case 2. CT scan shows that the dens deviates to the right side between the lateral masses.
In case 1 and case 2, we performed the manipulation procedure in the operating room. Under general anesthesia, patients were put in supine position. Manipulation with some axial traction applied to the occiput and chin, combined with gentle rotation toward the left side was attempted [24]. The reduction procedure was smooth, and neck ROM was regained.

In case 3, in the outpatient department, we tried to restore the reduction with manual straight axial stretch. The patient was put in supine position, with our index finger fixing on the C2 articular pillar. A manual contact of thenar eminance on the suboccipital area and a gentle axial stretch were applied before the patient felt pain. Then a sustained force was maintained for five second. Since this procedure was not successful, an anesthetic manipulation procedure was arranged and he was hospitalized on the next day after manipulation. No medication was necessary. On the second day, he felt better. And in the morning of the third day post-visiting, full active and passive ROM were achieved. Without any further intervention, he had complete resolution of the symptoms.

Our three patients were all male. The head position of the three patients were all rotated to the right. The hospital stay ranged from few hours to three days. The clinical data of these three patients are summarized in Table 1.

In case 1, full clinical restoration of cervical motion was found immediately after manipulative closed reduction. Neither collar nor further physical therapy was applied. After two-year follow-up, no complication was found.

In case 2, a palpable "clunk" occurred during reduction. The restoration of passive ROM for rotation and inclination was achieved immediately after manipulation. Active ROM of upper cervical spine was also restored, but there was still a sensation of pain on the right upper cervical spine when the patient actively rotated to the left.
After twelve sessions of physical therapy, including suboccipital relief, sternocleidomastoid muscle stretching, and neck stabilization exercise by ball therapy, he had a complete clinical restoration. No orthosis was prescribed for immobilization. At six months follow-up, there were no further symptoms.

In case 3, although the symptoms gradually resolved on the third day post manipulation, we still needed further clinical experience to determine the effect of this therapeutic technique.

It is a rare condition for a child to acquire acute torticollis. The possible causes, such as congenital abnormalities, upper cervical spine infection, tumors, juvenile rheumatoid arthritis, odontoid hypoplasia, or muscular torticollis should be distinguished from atlanto-axial rotatory subluxation [42]. The etiology of atlanto-axial rotatory subluxation is thought to be ligament laxity secondary to minor trauma, inflammation or infection [43]. In our cases, the etiologies were due to trauma in case 1, and no trauma history except sleep in case 2 and 3. (Table 1)

Plain radiography is usually the first prescribed method for a child who develops acute torticollis. In recent researchs, plain radiography has been shown to be limited advantage in investigating acute torticollis [39]. With the image of open mouth view revealed length discrepancy between odontoid and axis arch. (Table 2) and the clinical presentation, we could make the diagnosis of atlanto-axial rotatory subluxation. Cervical rotation resulting from positioning of the patient can lead to radiographic changes these are indistinguishable from atlanto-axial rotatory subluxation cases. In such cases, the radiographic diagnosis is based on a nonreducible rotation at the upper cervical spine level. For clinicians, this implies the clinical presentation is the most important diagnostic clue, and atlanto-axial rotatory subluxation should be kept in mind. Three dimensional CT scan is now the most specific modality for the diagnosis of atlanto-axial rotatory subluxation. If the diagnosis cannot be established by roentgenograms, three dimensional CT scan should be considered.

Table 1. Clinical Data of the Three Patients

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age</th>
<th>Sex</th>
<th>Cause</th>
<th>Cervical position</th>
<th>Duration</th>
<th>Recovery time after manipulation</th>
<th>Admission Day*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>9</td>
<td>M</td>
<td>Trauma</td>
<td>Rotated to right</td>
<td>1 day</td>
<td>Immediate</td>
<td>0</td>
</tr>
<tr>
<td>2</td>
<td>10</td>
<td>M</td>
<td>After sleep</td>
<td>Rotated to right</td>
<td>1 month</td>
<td>Cervical movement: immediate pain relief</td>
<td>3</td>
</tr>
<tr>
<td>3</td>
<td>7</td>
<td>M</td>
<td>After sleep</td>
<td>Rotated to right</td>
<td>1 week</td>
<td>Cervical movements and pain relief 3 days later</td>
<td>2</td>
</tr>
</tbody>
</table>

Sex: M= male, F= female
Cervical position: the rotational direction of head at visiting
Duration: the time from symptom onset to treatment
*: the duration from admission to discharge

Table 2. The clinical findings of the plain radiography

<table>
<thead>
<tr>
<th>Patient</th>
<th>Open mouth view</th>
<th>Lateral flexion and extension views* *</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Odontoid direction*</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>deviation to the right side</td>
<td>4mm</td>
</tr>
<tr>
<td>2</td>
<td>deviation to the right side</td>
<td>3.5mm</td>
</tr>
<tr>
<td>3</td>
<td>deviation to the left side</td>
<td>4mm</td>
</tr>
</tbody>
</table>

*: odontoid deviation relative to central position
**: the distance between odontoid and the anterior arch of atlas
The general treatment guides for atlanto-axial subluxation, although they are still controversial, have been reported by Philips and Hensinger [3], including head-halter traction, brace or collar immobilization, and more complicated arthrodesis of atlanto-axial joint. Their treatment plans are based on the duration from onset of torticollis. The treatment, if started earlier, is simple and the prognosis is good. If diagnosis making is delayed, the treatment will be more difficult and may prevent complete restoration of C1-2 upper cervical spine function.

For acute subluxation cases less than one week’s duration, Philips and Hensinger suggest immobilization with a soft collar and bed rest. If spontaneous reduction cannot be obtained, hospitalization and cervical traction are indicated, which are also the treatments for those whose duration of subluxation is from one week to one month. For patients whose clinical durations are more than one month, cervical traction is attempted. If this fails, arthrodesis should be considered [4,5].

Manipulation for close reduction of atlanto-axial rotatory subluxation was first reported by Burkus and Deponie [2]. In Philips and Hensinger’s research, manipulative reduction was also mentioned. They suggested that if the onset duration from onset to treatment is short, it is a good indication for manipulative management. However, they think manipulation has a higher risk than bed rest or traction.

In our cases, we performed the manipulative reduction procedure in the operating room under consideration of safety, and neurosurgeons were on standby at bedside. General anesthesia is designed for the relaxation of spasmodic neck muscle, and makes the manipulation procedure smoother. In case 1, the duration from onset to treatment was only one day, and manipulation was a good choice for treatment [3]. In case 2, though the duration from onset to treatment was four weeks, we tried manipulation to correct the rotatory dislocation. As passive range of motion was regained and active range of motion improved, we designed some physical therapies for the neck and suboccipital muscle spasm relief, sternocleidomastoid muscle stretch, and ball exercise for neck stabilization rather than orthotic device immobilization. With manipulation management, the hospital stay was short. (Table 1)

There are no orthoses that can completely immobilize the cervical spine, especially the atlanto-axial joint [9]. Although many of the cited papers [12,5,10] suggest orthotic devices for immobilization after reduction, there is no substantial evidence to show such devices are mandatory for the treatment of atlanto-axial subluxation.

According to the classification system of Fielding and Hawkins [1], our three cases are classified as type I subluxation, the most common type. We did not use any orthosis for post-reduction cervical immobilization, and the results were successful without any complication.

We suggest that if the atlanto-axial subluxation is classified as type I and good reduction has been achieved, orthosis may not be mandatory. However, for patients classified as type III or type IV subluxation, we think orthotic devices, such as halo devices or the Minerva body jacket may be necessary to maintain the alignment of the atlanto-axial joint, and to prevent recurrence. For type II subluxation, it may depend on the clinician’s judgements.

CONCLUSION

In conclusion, when the atlanto-axial rotatory subluxation is in early clinical period, or when the subluxation is type I, manipulation is a adequate procedure to restore atlanto-axial motion and to obtain symptom relief at once. The radiographic image of open mouth views and lateral flexion and extension views of cervical spine can be used for early diagnosis. As well, the clinicians’ awareness of atlanto-axial subluxation is most important way to prevent delayed diagnosis and treatment, which may lead to undesired complications.

REFERENCES

以徒手矯正來治療環椎軸椎之旋轉性脫位

蔡森蔚 周崇頌
台中榮民總醫院復健科

嬰兒斜頸在復健科門診是很常見的病症，然而因環椎軸椎之旋轉性脫位而引起的兒童急性斜頸卻很少見。在診斷上，常因 X 光的判讀困難及臨床醫師對此病症沒有概念而延誤，而此診斷上的延誤常使得治療變的更複雜。在此，我們回顧三個因環椎軸椎之旋轉性脫位而引起兒童急性斜頸的病例，並成功地以徒手矯正來治療。（中華復健醫誌 2001; 29(1): 51 - 58）

關鍵詞：環椎軸椎旋轉性脫位(atlanto-axial rotatory subluxation)，徒手矯正(manipulation)，斜頸(torticollis)