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# Atlantoaxial rotatory subluxation/displacement presented with torticolis

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*Atlantoaxial displacement is one of the most common causes of childhood torticollis. The problem occur spontaneously or may follow upper respiratory tract infection or minor trauma such as sneezing or dental procedures. Although Atlantoaxial displacement is common, rotatory displacement is rarely found. If the physicians misdiagnose, the patients could get wrong treatments especially physical therapy procedures which include cervical traction that may lead to life threathening result. Therefore, the evaluation for the underlying condition and appropriate plan of management will reduce the unnecessary operation and risk of complication.*

**Keywords :** *Torlicolis, Cervical spine displacement.*

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*กระดูกสันหลังส่วนคอเคลื่อนเป็นสาเหตุที่พบบ่อยในเด็กที่มีอาการคอบิดเอียง ซึ่งปัญหานี้อาจเกิดขึ้นโดยไม่มีสาเหตุ พบได้ในผู้ป่วยเด็กเล็กถึงเด็กโต โดยมักพบตามหลังอาการติดเชื้อของระบบทางเดินหายใจ การไอจามอย่างรุนแรง หรือการทำหัตถการในช่องปาก และมีผู้ป่วยเด็กบางคนมีการเคลื่อนบิดของกระดูกสันหลังส่วนคอ ซึ่งพบภาวะนี้ได้ไม่บ่อย ถึงแม้ว่าปัญหากระดูกคอเคลื่อนที่พบได้บ่อย แต่ในกรณีที่แพทย์ที่ไม่เคยมีประสบการณ์ในการเห็น หรือพบผู้ป่วยกรณีเช่นนี้ และให้การวินิจฉัยผิดว่าเป็นเพียงภาวะกล้ามเนื้อเกร็ง และส่งผู้ป่วยไปรับการรักษาทางกายภาพบำบัดที่ผิดวิธี โดยเฉพาะการดึงคอ ซึ่งอาจทำให้เกิดอันตรายถึงชีวิตได้ ดังนั้นการศึกษาถึงสาเหตุและวางแผนการรักษาที่ถูกต้องจะลดการผ่าตัดที่ไม่จำเป็น รวมถึงปัญหาแทรกซ้อนต่าง ๆ ได้*

**คำสำคัญ :** *คอบิดเอียง, กระดูกสันหลังส่วนคอเลื่อน*

## Case report

A 12-year- old Thai girl patient came to the Department of Rehabilitation Medicine of King Chulalongkorn Memorial Hospital. Her main presenting symptom was head and neck tilt for 7 months. She had the neck tilt with chin shift to the left and neck pain after wake up for 7 months. She did not have any weakness and/or numbness. The patient was aware of the problem for the first time when she went to see the dentist and was transferred to have the physical therapy at a government hospital. Ultrasonography, manipulation and cervical traction were done. She felt less pain but still had neck tilt. Then she came to our hospital for a second opinion. On physical examination, she had head rotated to the left and tilt to the right (Figure 1). Her neck range of motion was full on flexion and left lateral rotation but limited on extension and right lateral rotation (movement can not pass neutral position). Muscle spasm and trigger points were found at the right upper trapezius and right sternocleidomastoid muscles. Neurological examination revealed no cranial nerve deficits. Motor strength and sensation were normal in all extremities. Deep tendon reflex and Babinski's test were also normal.

X-ray film of the cervical spine in anter-oposterior, lateral and open mouth views showed rotation of C1 on C2 with right anterolateral subluxation of C1 and C2, and left lateral subluxation of craniocervical junction (Figure 2-A). Three dimension CT scan (3-D CT) showed compression of anterior subarachnoid space and mild impingement on the upper cervical cord by the tip of odontoid process. (Figure 2-B)

**Figure 1.** The patient presenting with head rotated to the left side and tilt to the right side.

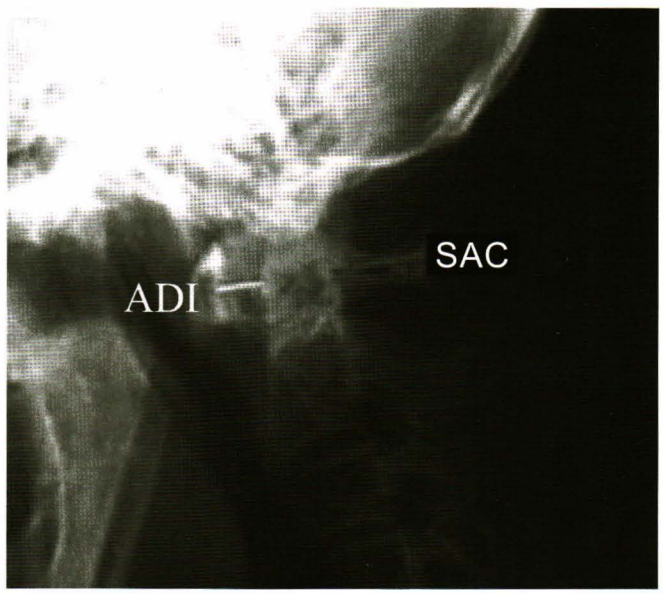


**Figure 2-A.**

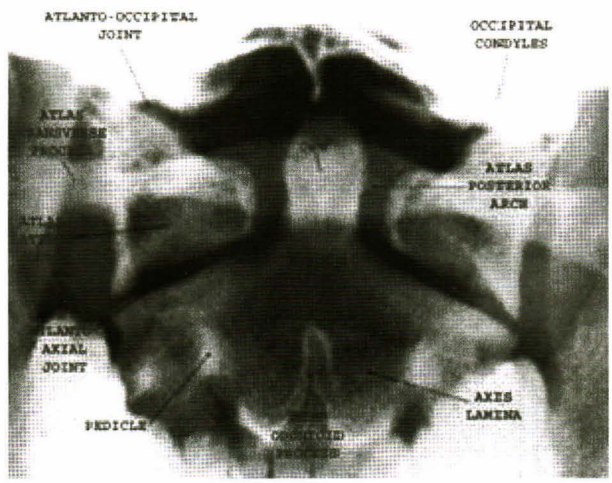


**Figure 2-B.**

**Figure 2.** Radiograph of cervical spine from third patient A: X ray open- mouth view: rotation of C1 on C2 with right anterolateral subluxation of C1 and C2 left lateral subluxation of craniocervical junction. B: Coronal image.

The treatments received in this hospital were as follows. We advised the patient to wear soft collar for relaxing the neck muscles and preventing the neurological damage. The neurosurgeon was consulted for correction because her conditions were in type III that be classified by Fielding and Hawkins[1] classification.

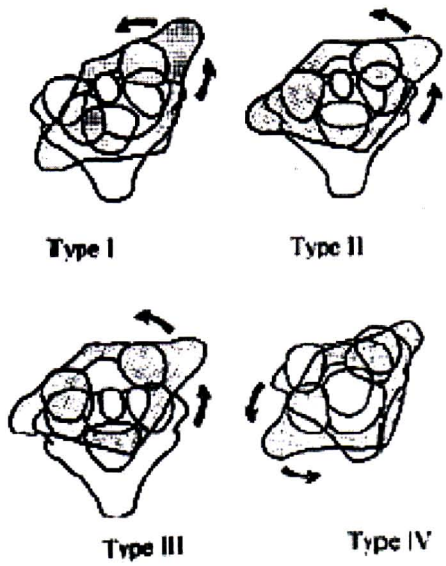


**Figure 3.** Atlantoaxial Rotatory Displacement.
: Classification by Fielding and Hawkins

Atlantoaxial Rotatory Displacement.
: Classification by Fielding and Hawkins. [2-6](Figure 3)

- Type 1 – Rotatory fixation without anterior displacement of the atlas
- Type II – Rotatory fixation with anterior displacement of the atlas of 3-5 mm
  - greater than 3 mm in older children and adults
  - greater than 4 mm in younger children
- Type III – Rotatory fixation with anterior displacement of more than 5 mm
- Type IV – Rotatory fixation with posterior displacement

Transoral lysis adhesion with facet fusion on the right side by iliac crest graft and posterior C1-2 wiring with rib graft were done. The patient conditions after the surgery and rehabilitation programs were satisfactory. After the operation, the patient was prescribed the philladephia collar to immobize the neck and to support and prolong stretching neck muscles.

Finally, after immobilization by the philladephia collar, the patient was suggested to do the isometric neck muscle and stretching exercise.

## Discussion

Atlantoaxial displacement is one of the most common causes of childhood torticollis whereas rotatory displacement is not common.[2-6] The problem can occur spontaneously or follow an upper respiratory tract infection or minor trauma such as sneezing, dental procedures.

Clinical manifestations of atlantoaxial rotatory displacement are torticolis, typical head position of *"Cock - Robin" (male bird eyeing worm)* [3-5], lateral flexion to one side, rotation toward the opposite side with slight flexion.

Limited or diminished ranges of motion by pain and neck muscle spasm are also found. Fixed deformities or persisted torticollis along with decreased neck motion can be found if the patients do not receive appropriate treatments.

The pathophysiology of alantoaxial rotatory displacement is not well understood.[2-5] In some cases, direct connection between the periodontoid venous plexus and the pharyngovertebral vein and suboccipital epidural sinuses are found. Children appear to be more susceptible secondary to the steeper dens-facet angle and rich vascular folds in the atlantoaxial and lateral atlantoaxial joints.

Radiography shows failure of superimposition of two elements of posterior arch of C1 in true lateral view.

Open-mouth view shows widening of the odontoid–lateral mass interval and absence of a fracture of the atlas. Lateral displacement of the dens by more than 4 mm is suggestive of atlanto-axial rotatory fixation. (Figure 4)[4,7-10]

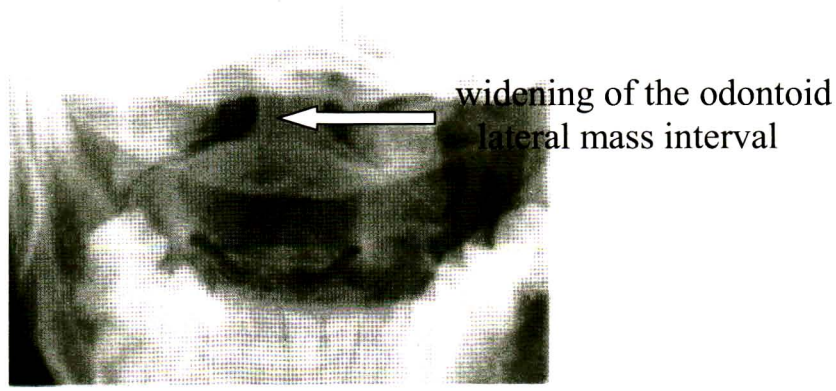


**Figure 4.** Open - mouth view shows widening of the odontoid - lateral mass interval.

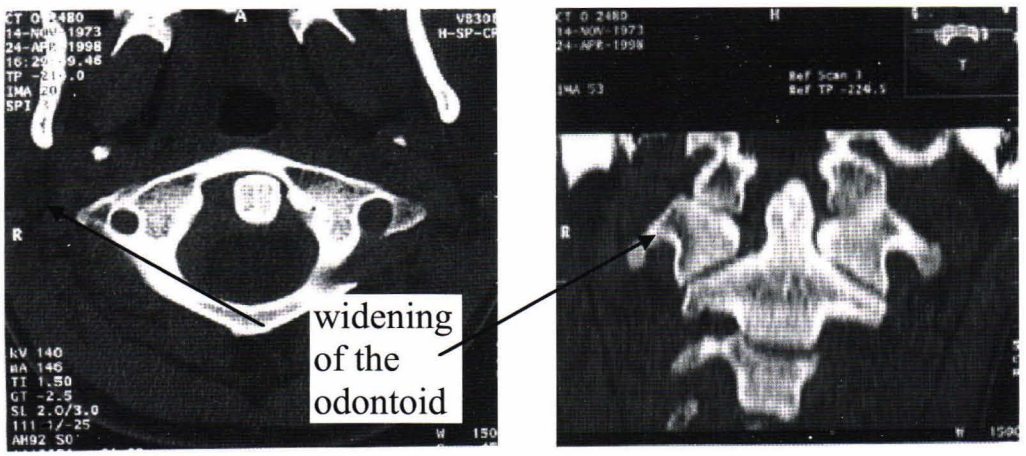


**Figure 5.** CT Scan 5A - Axial image,widening of the odontoid.
5B - Coronal image, widening of the odontoid.

CT scanning can be useful to confirm rotatory fixation and exclude fracture of the atlas and axis. (Figure 5)

Classification for the atlantoaxial rotatory displacement by Fieldings and Hawkins as shown in figure 3 was used for management decision. Most of atlantoaxial rotatory subluxation /displacement presented with torticolis resolves spontaneously but some may need treatments.[1,11-13]

If the patients have this problem less than one week, the immobilization by the soft collar on the neck and taking rest for about 1 week may be helpful. If the patients do not have spontaneous recovery, hospitalization, Halter traction, muscle relaxants and analgesia medcations are considered.

If the patients have the symptoms between 1 week to 1 month, hospitalization for cervical traction, muscle relaxants, and analgesia medications must be concerned. Halo vest should also be considered. When there is no anterior displacement after reduction, the patient should wear cervical support as long as symptoms persist. If the patient still has the anterior displacement after reduction, immobilization of the cervical spine must be continued for 6 weeks.

If the symptoms persist more than one month, the patient should be hospitalized for halo skeletal traction. If the patient still has resubluxation or fixed rotatory subluxation, the operation should be considered.

The indications for operative treatment are [14] neurological involvement/ neurological deficit, anterior displacement, failure to achieve and maintain correction if deformity exist for longer than 3 months and recurrence of defomity after adequate conservative treatment consisting of at least 6 week of immobilization.

The prognosis of the atlantoaxial instability (AAI) is good for those who have symptomatic AAI in whom posterior spinal fusion is successful and function returns. Surgery has been demonstrated to relieve pain in 95 % of the patients and decrease myelopathy in 74 % of the patients, depending on the severity of the symptoms and the cause of the instability.The treatments of atlantoaxial instability in

pediatric patients according to the clinical review of 23 pediatric cases of atlantoaxial instability treated from March 1990 to October 2002 showed that 60.9 % were treated without surgical intervention and resulted in excellent outcomes and 21.7 % of cases were treated with a cervical halo (mean patient age 72.6 months) alone for 3 months.

## Conclusion

Atlantoaxial displacement is one of the most common causes of childhood torticollis. Although the atlantoaxial displacement is common but rotatory displacement cases are less found. Early diagnosis, investigation and treatment have to be done urgently for the better outcome. Some patients received the wrong diagnosis such as cervical muscle spasm and received inadequate treatment which could be harmful delay treatment resulting in poor outcomes.

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