THREE-DIMENSIONAL COMPUTED TOMOGRAPHY IN INFANTILE ATLANTOAXIAL ROTATORY FIXATION

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Three-dimensional CT was used to examine the atlantoaxial joint of a nine-year-old girl who presented with fixed rotation of the head 3.5 months after an apparently spontaneous acute torticollis. The method provided clear, anatomical images to show the site, extent and direction of the type-I rotatory fixation (Fielding and Hawkins 1977).

The 3-D reconstructions helped to guide manipulation which was successful in this late case.

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Rotatory subluxation or dislocation of the atlantoaxial joint is not uncommon in children and adolescents. It is usually caused by minor, sometimes unnoticed trauma during games or by sudden rotation and lateral tilting of the head. Laxity of the joints’ capsule and ligaments, and the orientation of the flat articular facets may even allow C1-C2 dislocation to occur spontaneously in sleep (Torklus and Gehle 1975). Children are sometimes referred for treatment after the spontaneous reduction of such an acute torticollis.

Rotatory displacement should be suspected in every child who presents with acute, painful torticollis (Sullivan 1949; Jackson 1950; Scapinelli 1971-2; Fielding and Hawkins 1977; El-Khoury, Clark and Gravett 1984), sometimes in association with upper respiratory infection (Grisel 1930).

The characteristic posture is with the head rotated in one direction and tilted in the opposite direction. The pathognomonic limitation of rotatory movement is towards the convex, painful side of the torticollis. The clinical diagnosis should be made as early as possible and confirmed by radiography (Dankmeijer and Rethmeier 1943; Jacobson and Adler 1953; Wortzman and Dewar 1968) to allow prompt reduction. An open-mouth view of C1-C2 shows a persistent displacement of the dens between the lateral masses of the atlas, asymmetry of the atlantoaxial joint spaces caused by reciprocal gliding or overriding, and displacement of the spinous process of the axis from the midline, usually towards the same side as the chin (Sudeck’s sign; Sullivan 1949). On a lateral view the atlas may appear to be tilted on the axis, with different forward displacement of each of the lateral masses. Less commonly, the lateral view may show an increase in the anterior space between atlas and dens, which in normal children never exceeds 4 to 5 mm (Jackson 1950; Oberthaler and Schwarz 1984).

The abnormal position of the head and the stiffness make conventional radiology difficult and even on plain tomography the C1-C2 relationship is not always easy to appreciate. Better definition can now be offered by static or functional CT (Fielding et al 1978; Post 1984; Kowalski et al 1987; Ellis 1991; Grøgaard, Dullerud and Magnaes 1993).

When the condition is longstanding, the deformity may become fixed (Wortzman and Dewar 1968; Fielding and Hawkins 1977). Adaptation between growing vertebrae may occur, and make reduction progressively more difficult or even impossible (Oberthaler and Schwarz 1984).

We report a case of type-I rotatory fixation (Fielding and Hawkins 1977) diagnosed late and studied by spatial reconstruction using a third-generation CT scanner (Tomoscan LX, Philips, Germany).

CASE REPORT

All but one of the author’s series of 45 children with rotatory atlantoaxial subluxation or dislocation were successfully reduced by simple conservative methods, with no recurrences or late sequelae. The diagnosis was usually made within 24 to 48 hours, up to a maximum of ten days. Eighteen of these cases have been reported in detail (Scapinelli 1971-2).

In the patient reported here, the diagnosis was considerably delayed. A healthy nine-year-old girl woke one morning with painful torticollis, her head rotated to the left and tilted to the right. On the previous afternoon, she had been diving from the edge of a swimming-pool, with no immediate symptoms. She was seen by several doctors and diagnosed as having acute muscular torticollis, local inflammatory disease having been excluded.

Ten days after the onset anteroposterior, lateral and
Open-mouth (a) and lateral (b) radiographs taken ten days after the onset of torticollis. The diagnosis of rotatory atlantoaxial dislocation was not made, despite clear evidence of overriding of the articular facets, displacement of the spinous process of C2 and of rotation of the atlas on the axis.

Fig. 1a  Fig. 1b

Appearance of the patient 3.5 months after the onset of torticollis (a). Rotation to the left is free (b), to the right it is severely limited (c).

open-mouth radiographs were taken (Fig. 1), but the signs of incongruity between the articular facets of the two upper cervical vertebrae were overlooked by a number of specialists and she was treated by exercises, gentle manipulation, massage, local heat and a few days of traction and a collar without improvement. The persistence of the abnormal head position led to referral to a neurologist and a psychiatrist. She was referred to us 3.5 months after the initial appearance of the torticollis.

**Diagnosis.** On admission she showed the characteristic 'cock-robin' position with prominence of the elongated left sternomastoid (Fig. 2a). There was local tenderness behind the left mastoid process. She could turn her head freely to the left (Fig. 2b), but rotation to the right was blocked and painful (Fig. 2c). Flexion and extension were nearly normal and there were no signs of neurological or upper-respiratory involvement. She showed mild signs of general joint laxity.

CT was performed and three-dimensional reconstructions were made from contiguous 1.5 mm sections (Fig. 3). These showed complete C1-C2 locking caused by rotatory dislocation of the atlas on the axis with anterior luxation of the right C1 articular facet and posterior displacement of the left. The atlas was tilted anteriorly to the right. The odontoid process was in an asymmetrical position (Figs 3a to f), but still in contact with the anterior arch of the atlas indicating that the transverse ligament was intact. No congenital abnormalities or dysplasia were present, and the diagnosis of type-I rotatory fixation was made.

**Treatment.** Five days after admission, with the patient under general anaesthesia and in a supine position, careful manipulation was performed by traction on the neck in extension and rotation to the right, assisted by the use of an image intensifier and guided by the three-dimensional CT images. Reduction was finally achieved and confirmed by an anteroposterior open-mouth radiograph. A Minerva cast was then applied with the head tilted and rotated in an overcorrected position, and retained for two months. The maintenance of reduction was confirmed by open-mouth views before and after removal of the cast. Full and painless rotatory movements were gained in a few weeks, but at the six-month review, there was still some reluctance to go to the extremes of active rotation. At one year the patient had no symptoms and a full free range of movement. The radiographs showed normal atlantoaxial
Three-dimensional CT images showing complete rotatory dislocation on anterior (a), oblique (b), lateral (c), posterior (d), from above (e) and below (f) views. The inferior articular facets of the atlas are locked anteriorly on the right side and posteriorly on the left. In all the reconstructions and mainly in (e) the dens is seen to be eccentric but with good apposition to the anterior arch of the atlas. The spinous process of the axis is displaced.
DISCUSSION

Atlantoaxial rotatory dislocations are said to become irreducible after one month. In our case, three-dimensional CT showing the dislocated and locked facets of the atlas on the axis (Fig. 3) helped us to reduce the displacement 3.5 months after the initial signs.

The previous attempts to correct the torticollis seem to have changed the anatomy. The C1-C2 dislocation seen on three-dimensional CT was different from that initially shown on the standard radiographs (Fig. 1).

The C1-C2 rotatory fixation was clearly due to mechanical locking of the articular facets (Fig. 3), with no apparent lesion of the transverse ligament. The main predisposing factor to dislocation was probably physiological joint laxity. If the manipulation had been unsuccessful residual fixed deformity would have resulted.

CT scans and three-dimensional reconstructions are rarely needed: only doubtful or difficult cases or delayed diagnoses can justify the local exposure to rather high doses of radiation, and the expense. With early diagnosis, reduction is usually possible by simple traction in extension in a Glisson’s sling, use of a plastic collar for about two weeks and physiotherapy thereafter (Stimson and Swenson 1935; Scapinelli 1971-2; Sherk 1975; Oberthaler and Schwartz 1984). The use of skull traction in children is controversial, and only indicated in cases of longstanding fixation. When there is neural involvement, or rarely when conservative management has failed, posterior fusion may be considered after a period of traction (Fielding and Hawkins 1977).

Increased awareness of the simple, sometimes spontaneous onset of this form of torticollis in children and adolescents and recognition of the associated radiological signs are essential if missed diagnoses, fixation, permanent deformity or instability requiring surgery are to be avoided.

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REFERENCES


