ATLANTO-AXIAL ROTATORY FIXATION AND FRACTURE OF THE CLAVICLE
AN ASSOCIATION AND A CLASSIFICATION

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Five children with atlanto-axial rotatory fixation (AARF) in association with fractures of the clavicle are described. It is postulated that the rotary fixation is a direct result of the trauma which produces the fracture. The importance of early diagnosis is stressed, since delayed diagnosis may lead to chronic deformity. Early diagnosis depends on awareness of the possibility of AARF, and either fluoroscoping the patient in order to take appropriate spot films or imaging the atlanto-axial joint by CT. A simple classification of AARF is proposed based on distinct radiological features which differentiate subluxation from dislocation.

Atlanto-axial rotary fixation (AARF) is a well-documented but uncommon cause of childhood torticollis. It is usually associated with major trauma or upper respiratory tract infection. We report a series of five children with AARF associated with clavicular fractures. We are unaware of any previous reports of this association.

CASE REPORTS

Case 1. A nine-year-old girl presented in November 1978 with a greenstick fracture of the left clavicle sustained when she fell while playing in the garden. When reviewed two days later she was noted to have a torticollis with her head turned to the left and tilted to the right. The plain radiographs were unhelpful and she was treated with a soft collar. In February 1979 repeat films, followed a week later by fluoroscopy, confirmed an atlanto-axial dislocation, with the right lateral mass of the atlas rotated anteriorly on the axis (Figs 1, 2 and 3).

She was admitted for skull traction; this was unsuccessful in reducing the dislocation and was discontinued. In May 1979, at surgical exploration of the cervical spine, the dislocation was confirmed. It proved irreducible so a Gallie fusion was performed with the atlas fixed in the rotated position.

By February 1980 she was holding her head straight and had no facial asymmetry. She was discharged from follow-up.

Case 2. An eight-year-old girl fell off her bicycle in June 1981, sustaining a fracture of the medial end of her left clavicle. She was noted to have a torticollis with her head turned to the right. Cervical spine radiographs were thought to be normal; the torticollis was considered to be the result of a protective mechanism relaxing the sternal insertion of the sternomastoid.

Her torticollis persisted, and in May 1982 the diagnosis of AARF was made after screening and CT scanning, which showed $35^\circ$ rotation to the right of C1 on C2 with locked facets on the left (Figs 4 and 5). In June 1982 fusion was performed in situ for an irreducible

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<th>Table I. Differentiation of types of atlanto-axial rotatory fixation</th>
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<tr>
<td><strong>Type I</strong></td>
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<td>C1/2 joints are fixed in partial rotation but not dislocated</td>
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<td>Children and adults</td>
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<td>On AP radiography of C2 the anteriorly-rotated lateral mass of C1 appears:</td>
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<tr>
<td>wide</td>
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<tr>
<td>closer to the odontoid</td>
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<td>slightly elevated</td>
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dislocation. At review in June 1986 her head was held straight and she had no facial asymmetry, but there was some limitation of both flexion and rotation to the left.

Case 3. A six-year-old girl fell from her bunk in October 1981, sustaining a greenstick fracture of her right clavicle. When reviewed three weeks later she was noted to have a torticollis, with her head turned to the left and tilted to the right. In addition she had an otitis media with cervical lymphadenopathy.

In January 1982, the lymphadenopathy had resolved but the torticollis persisted and she was therefore admitted for physiotherapy and hydrotherapy for three weeks, but with no improvement. The diagnosis of AARF was finally made in April 1982 by radiographic screening (Fig. 6).

In May 1982 she had developed left-sided long-tract signs with sustained ankle clonus and an up-going plantar response. She was admitted to hospital for preliminary
skull traction, followed by an unsuccessful manipulation under anaesthetic. At operation in June 1982 an irreducible dislocation was found and in situ fusion performed. Six years later she has a minimal head tilt with slight restriction of rotation to the right, but there was no neurological deficit.

**Case 4.** A seven-year-old girl fell onto her right shoulder whilst playing at school in July 1984, sustaining a greenstick fracture of the mid-shaft of her right clavicle. The following day she was noted to have a torticollis with her head turned to the left, and plain radiographs confirmed the diagnosis of AARF with locked facets on the right.

She was admitted immediately for halter traction; the dislocation reduced over the following 24 hours. Three years later she was clinically normal.

**Case 5.** A nine-year-old girl fell off her bicycle in September 1987 sustaining a fracture of the mid-shaft of her left clavicle and an associated torticollis, with her head turned to the right. Three weeks later her torticollis was still present; a CT scan confirmed the diagnosis of AARF with locked facets on the left.

She was admitted for halter traction, and the dislocation reduced over the next four days. This was followed by six weeks protection in a 'Neo-fract' collar, when repeat radiography and CT scanning confirmed the reduction. One year later she was clinically normal with a full range of movement.

**DISCUSSION**

The diagnosis of clavicular fracture rarely presents a problem, but the recognition of atlanto-axial rotatory fixation frequently does; it is a well-known but uncommon cause of childhood torticollis, usually associated with rotational injury or an upper respiratory tract infection. The association of AARF and clavicular fracture has not been previously recognised.

All of the patients we report were children between the ages of six and nine years, with a history of a fall onto the shoulder sufficient to result in a clavicular fracture. Interestingly, all five were girls. This female preponderance has previously been noted by Ono et al (1985) whose series of 13 children included 10 girls. All but one (case 3) of our patients were noted to have a torticollis on their first attendance at the fracture clinic, but the significance of this was not always immediately realised. In our first three patients the diagnosis was delayed by three to 11 months; they all eventually needed atlanto-axial fusion. The later two cases were recognised within two weeks, because of our increased awareness of the association. In these cases, a complete and lasting reduction was achieved by simple traction.

Atlanto-axial rotatory fixation was first described in detail byCorner in 1907 who reviewed 20 cases. There have subsequently been many case reports especially in children (Watson-Jones 1932: Coutts 1934: Fielding and Hawkins 1977; Burkus and Deponte 1986). In those cases related to trauma (El-Khoury, Clark and Gravett 1984) it is likely that the injury resulted from forced rotation combined with an element of flexion; this is consistent with a fall onto the shoulder and side of the head. The flat facet joints in the child, whose ligaments are relatively lax, allow excessive rotation (beyond 45°), so permitting anterior subluxation or dislocation of C1 on C2 (Jacobsen and Adler 1956; Wortzman and Dewar 1968). This mechanism would certainly explain the co-existence of the clavicular fracture, although a different mechanism must have operated in case 1 as the fracture was on the opposite side.

The natural history of the condition is uncertain, since there are few reports of late, untreated cases. There are, however, reports of a compensatory counter atlanto-occipital subluxation leading to a so-called 'pseudo-reduction' (Ono et al 1985; Clark et al 1986), with the occiput and axis pointing in the same direction, leaving the atlas dislocated at both levels. This leads one to question the rationale of in situ fusion, as recommended...
by Fielding and Hawkins (1977). This would appear merely to perpetuate an already stable situation, especially as all our patients who required operation were found to have an irreducible dislocation. There are of course valid indications for fusion where there are signs of neurological involvement (as in our case 3).

**Radiology.** The diagnosis of AARF may be difficult or even impossible on routine anteroposterior and lateral radiographs, because the torticollis makes it impossible for the radiographer to judge the correct positioning. The diagnosis can be established by fluoroscopy or ideally by CT. During fluoroscopy the key to diagnosing the dislocation lies in obtaining a true anteroposterior view of C2. This is best done by screening the patient supine on a table with an overcouch tube. With an open mouth, the patient turns until C2 is projected symmetrically; some angulation of the tube is usually necessary to project it trans-orally. A spot view is taken and studied for the way it demonstrates C1 in relation to C2.

CT scanning can be used either instead of, or in addition to, the above technique (Fielding et al 1978). A series of 5 mm slices is taken through C1 and C2, and are supplemented by 2 mm slices through the anteriorly-rotated inferior facet of C1 if difficulty is experienced in assessing its relationship to the superior facet of C2. CT also enables the rotation to be quantified by measuring the angle between the axes of C1 and C2 on successive slices (Ono et al 1985).

**Classification.** Rotation at the atlanto-axial joint results in two distinct radiographic and clinical presentations which we shall designate type I and type II.

In type I AARF there is anterior rotation of one side of C1 on C2 without dislocation. The joint is fixed at, or near, the extremitry of its normal range of movement by forces which are not fully understood. Type I fixation is seen in adults as well as children and the radiological features have been well described by Wortzman and Dewar (1968). The anteriorly-rotated lateral mass of C1 appears to be closer to the odontoid and to be wider than its fellow on a true anteroposterior view of C2 (Fig. 7). Because the facet joints slope upwards anteriorly, the anteriorly-rotated lateral mass tilts up slightly and the joint space appears slightly wider than that on the other side.

In type II AARF there is anterior dislocation of one lateral mass of C1 on C2 with interlocking of the facets. All of our cases are examples of this type. It has been reported only in children, and is generally associated with trauma. There must be a potent soft tissue element in the dislocation because reduction by traction is often followed by recurrence of the dislocation. The anteroposterior view of C2 shows a very different relationship to C1 from that of type I AARF. The anteriorly-rotated lateral mass of C1 still looks wide but instead of tilting up it drops down, overlapping the lateral mass of C2. It moves away from the odontoid peg whilst the opposite lateral mass moves close to it. Several papers have described this type of dislocation (El-Khoury et al 1984; Ono et al 1985) but its differentiation from type I AARF, and in particular its distinct radiological features, have not been described. Table I summarises the difference between the two types.

**Conclusions.** The diagnosis is relatively easy, but only when the possibility of the condition and its association with a fractured clavicle has been considered, and the appropriate fluoroscopic or CT views have been taken. However, if one is not aware of the possible association of the two injuries, undue reliance may be placed on conventional radiographs and the AARF missed. It is therefore important to explain the presence of a torticollis and to be absolutely satisfied that the radiographs are indeed normal, and not to assume that it is due to protective sternomastoid spasm related to the clavicular fracture. If there is any lingering doubt, then CT scanning must be used to exclude AARF.

The incidence of these cases, about one every two years in two health districts with a combined population of 650 000, is undoubtedly low but if this is true nationwide then a minimum incidence of 35 cases annually is probable, many of which may be undiagnosed. Our experience suggests that prompt diagnosis and treatment leads to a permanent resolution of the deformity.

We wish to express our thanks to Mr J. A. M. Philpison, Mr J. K. Tucker, Dr J. F. B. Dossetor, Mr C. Shaw and Mr M. H. Matthewson, under whose care cases 1 to 4 were admitted.

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**REFERENCES**


