Case report

Spontaneous atlantoaxial subluxation: an unusual presenting manifestation of Reiter's syndrome

R D MELSOM,¹ J C BENJAMIN,² AND C G BARNES¹

From the Departments of ¹Rheumatology and ²Neurosurgery, The London Hospital, Whitechapel, London

SUMMARY A male Sikh presented with spontaneous neck pain. Plain cervical radiography showed gross atlantoaxial subluxation (AAS). Computed tomography showed soft tissue anterior to a subluxed odontoid peg. A diagnosis of atlantoaxial tuberculosis was considered. Confirmation of this would have required a trans-oral anterior cervical biopsy. Despite the paucity of other rheumatic symptoms a diagnosis of Reiter's syndrome with spondylitis was made, and the only surgical procedure required was posterior fusion of C1 and C2, with resultant spinal stability and pain relief. This is the first report of Reiter's syndrome presenting with AAS.

Key words: seronegative spondyloarthropathy.

Spontaneous atlantoaxial subluxation (AAS), a serious complication of rheumatic disease, is seen most frequently in rheumatoid arthritis, psoriatic arthritis, and ankylosing spondylitis. In one study AAS was reported with a prevalence of 189 in 1000 patients with rheumatoid arthritis admitted to hospital.¹ In another study nine cases of AAS were found in 21 patients with psoriatic spondylitis, who were themselves identified from 87 patients with psoriatic arthritis.² When about 100 patients with ankylosing spondylitis were reviewed 17 cases of AAS were noted.¹ In addition, there are three isolated reports of AAS in Reiter's syndrome,^{1 3 4} and one study in which two patients with AAS were identified from 145 consecutive cases of Reiter's syndrome.⁵ Atlantoaxial subluxation has not previously been reported as a presenting manifestation of Reiter's syndrome.

Case report

A 30 year old Sikh, born in India but resident in the UK for 13 years, presented with a three week history of acute continuous neck pain without

preceding trauma. There were no neurological symptoms and no history of psoriasis. Initially he denied any other relevant information but subsequently recalled an episode of urethritis occurring six years previously while revisiting India, and requiring parenteral antibiotics. Immediately afterwards he had experienced an episode of neck, low back, and left ankle pain. The neck pain disappeared completely over a few weeks as did the ankle pain, leaving only persistent mild low back pain. Until the recent episode of neck pain he had neither sought medical advice nor had treatment for any of these rheumatological symptoms. On examination, neck movements were reduced and painful. Lumbosacral and thoracic spine movement was reduced but pain free. Chest expansion was 10 cm. Neurological examination was normal.

His erythrocyte sedimentation rate was 60 mm/h (Westergren). Haematology, biochemistry, serology, urine culture for tuberculosis, chest radiography, and Mantoux test were negative or normal. He was B27 positive. Plain cervical radiographs showed 10 mm of AAS on extension, increasing to 15 mm on flexion. Computed tomography (CT) of the cervical spine showed AAS, an eroded odontoid peg compressing the theca and soft tissue anterior to the peg (Figs 1a and b). Cervical myelography showed a distorted column of contrast at C1 to C2.

Accepted for publication 14 July 1988.

Correspondence to Dr R D Melsom, Department of Rheumatology, The London Hospital, Whitechapel, London E1 1BB.

Fig. 1a

Fig. 1b

Fig. 1 (a) Computed tomogram at C1 without contrast; soft tissue is seen around the subluxed odontoid peg. The latter is compressing the theca. (b) Computed myelotomogram at C1; again the subluxed odontoid peg is seen to compress the dura which is now outlined by radio-opaque contrast medium.

(Fig. 2). Plain radiography demonstrated asymmetrical lumbar syndesmophytes and bilateral sacroiliitis. Joint space loss and erosions were evident at the left subtalar, mid-tarsal, and first toe interphalangeal joints.

He was treated initially with a collar. Subsequently, a posterior cervical fusion was performed; two laterally placed Harrington rods were attached by hooks superiorly to the atlas and inferiorly to C2. Iliac crest cancellous bone was then

Fig. 2 Lateral cervical myelogram showing a column of radio-opaque contrast distorted at the level of C1 to C2 by atlantoaxial subluxation.

packed between the two vertebrae. After the operation his neck pain resolved.

Discussion

Lifeso, in 1987, described 12 patients from Saudi Arabia with atlantoaxial tuberculosis.⁶ Fifteen additional sites of tuberculous involvement were

identified in these patients; seven were musculoskeletal, three soft tissue, and five spinal. Features shared by those 12 patients and the patient reported here included neck pain and AAS at presentation, Asian origin, musculoskeletal and spinal involvement elsewhere, a raised erythrocyte sedimentation rate, and absence of features of tuberculosis on chest radiograph. Also, in the patient reported here CT showed material which could have been tuberculous around the subluxed odontoid. Thus, initially, tuberculous AAS was considered to be the most likely diagnosis in the patient described here. Confirmation of this diagnosis would have required a trans-oral approach to the odontoid peg to obtain involved tissue. Indeed, the report from Saudi Arabia advocated a trans-oral approach for both diagnosis and decompression of the spinal cord, followed by halo traction and, later, a posterior fusion of C1 and C2 for patients who had the degree of AAS caused by tuberculosis seen in the patient described here.

The patient reported here, however, differed in two respects from those described by Lifeso: he was negative on Mantoux testing and CT showed no evidence of a retropharyngeal abscess, whereas all 12 of the Saudi Arabian patients were Mantoux positive and 10 had retropharyngeal abscesses. Furthermore, in this patient there was much in favour of a diagnosis of Reiter's syndrome despite the paucity of recent rheumatological symptoms other than neck pain. Six years previously there had been an episode of urethritis and arthritis. Conjunctivitis had not been noticed, but eye involvement may be absent or subclinical in Reiter's syndrome.^{4 7} There had been persistent, if mild, lumbosacral pain with reduced, albeit pain free, spinal movement.⁷ There was radiological evidence of asymmetrical erosive joint disease in the legs but not the arms,⁷ asymmetrical lumbosacral syndesmophyte formation,³ and bilateral sacroiliitis.^{7 8} Furthermore, the patient was found to be positive for the HLA-B27

antigen, which is found in only 5% of the northern Indian population.⁹

The diagnosis of Reiter's syndrome thus seemed secure. The possibility remained that this patient had Reiter's syndrome and tuberculous AAS. Although AAS is a rare occurrence in Reiter's syndrome compared with rheumatoid arthritis and psoriatic arthritis,^{1 2} it has been reported on five separate occasions previously.^{1 3 4 5} Therefore it was felt that the AAS was secondary to Reiter's syndrome and that complex surgery merely to exclude tuberculosis was not justified. The indications and possible advantages of the anterior and posterior surgical approaches to C1 and C2 in the treatment of AAS in rheumatoid arthritis have been reviewed elsewhere.¹⁰ In this case anterior decompression was not considered necessary as there were no signs of spinal cord compression. A posterior fusion of C1 and C2 was performed with complete relief of neck pain.

References

- 1 Sharp J, Purser D W. Spontaneous atlanto-axial dislocation in ankylosing spondylitis and rheumatoid arthritis. *Ann Rheum Dis* 1961; **20**: 47-77.
- 2 Killebrew K, Gold R H, Sholkoff S D. Psoriatic spondylitis. *Radiology* 1973; **108**: 9-16.
- 3 Santavirta S, Slati P, Sandelin J, Lindqvist C, Konttinen L. Atlanto-axial subluxation in patients with seronegative spondylarthritis. *Rheumatol Int* 1987; **7**: 43-6.
- 4 Latchaw R E, Meyer G W. Reiter's disease with atlanto-axial subluxation. *Radiology* 1978; **126**: 303-4.
- 5 Moilanen A, Yli-Kerrtula U, Vilppula A. Cervical spine involvement in Reiter's syndrome. *ROFO* 1984; **141**: 84-7.
- 6 Lifeso R. Atlanto-axial tuberculosis in adults. *J Bone Joint Surg [Am]* 1987; **69**: 183-7.
- 7 Ford D K. Reiter's syndrome. *Bull Rheum Dis* 1970; **20**: 588-91.
- 8 Dirheimer Y. *The craniovertebral region in chronic inflammatory rheumatic diseases*. Berlin, Heidelberg, and New York: Springer, 1977: 132.
- 9 Aizawa M. HLA in Asia-Oceania 1986. *Proceedings of the third Asia-Oceania histocompatibility workshop conference*. Japan: Hokkaido University Press, 1986: 1086.
- 10 King T T. Rheumatoid subluxations of the cervical spine. *Ann Rheum Dis* 1985; **44**: 807-8.