Atlanto-axial subluxation

A six-year follow-up report

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Atlanto-axial subluxation has been shown to be a common radiological feature of cases of rheumatoid arthritis, particularly those with other destructive features. There is poor correlation with neurological abnormality, even for those cases with gross degrees of subluxation. It has been postulated, however, that complications and sudden death could be more common than is generally believed (Sweetnam, 1970), but no definitive long-term studies have yet been reported.

In 1963 a prospective study of 333 consecutive hospital admissions for rheumatoid arthritis was performed by the present authors (Conlon, Isdale, and Rose, 1966); of these, 23 per cent. showed atlanto-axial subluxation, which was greater than 5 mm. displacement in a quarter of the cases. A follow-up survey of these patients is reported to give information about the progress of the condition and its significance.

Material and methods

In 1969 an attempt was made to contact all patients included in the 1963 study. A further x-ray study was made of those contacted, using the same technique of lateral flexion centring on C2. Where possible patients were examined clinically, but, because of distances often involved, radiological assessment was all that could be achieved in 56 patients.

The follow-up survey was done 5½ to 6½ years after the original assessment (mean 6 years); 59 patients (17·8 per cent.) had died in the interim and, of those with subluxation at the initial examination, the cause of death was ascertained from the doctors in attendance at the time of death. 88 patients could not be contacted, most because of change of domicile, some because of a change of name, and a few because of emigration. Ten patients were too ill to be x-rayed and five would not co-operate, making a total of 103 not followed.

171 patients were contacted and satisfactory films obtained, this being 62 per cent. of those not known to be dead.

As expected, the patients who had died between the two radiological surveys included an increased number

of cases with subluxation and severe disease. There was no significant difference in the radiological features of the follow-up cases and those not followed, as shown in Table I. The 1969 films of the 171 cases followed up were read separately by the two authors, with particular regard to the changes previously noted to be characteristic of rheumatoid involvement of the cervical spine, and subsequently agreement reached by excluding doubtful changes.

Table I *Follow-up of 333 RA cases seen in 1963*

<i>Follow-up</i>	<i>Deaths</i>	<i>No follow-up possible</i>	<i>Follow-up done</i>	<i>Total</i>
No. of cases	59	103	171	333
AAS Minor (2½-5 mm.)	10	24	31	65
AAS Major (over 5mm.)	7	5	7	19
	17	29	38	84
Other RA features	18	18	47	83
No RA features	24	56	86	166

Results

(a) Deceased patients

Seventeen of the 59 who died were known to have had atlanto-axial subluxation, but in no case did this contribute to mortality. The causes of death, ascertained in all, covered a wide range, including uraemia (2), myocardial infarction (2), ruptured abdominal aortic aneurysm (1), cancer (3), gastrointestinal haemorrhage (2), cerebral haemorrhage (2), cerebral thrombosis (1), cor pulmonale and heart failure (2), aortic stenosis and heart failure (1), and suicide (1). The ages of the patients ranged from 49 to 83. Among those patients in whom subluxation was not found in 1963, only incomplete mortality data could be obtained, but in none was it thought that death had been due to rheumatoid involvement of the neck.

(b) *The 171 patients followed-up*

These cases showed an increase in the rheumatoid changes and an increase in osteoarthritic abnormalities with a diminution in the cases showing no definite change (see Figure).

The nature of the rheumatoid change is shown in Table II. The increase in atlanto-axial subluxation (AAS) over the 6-year period was marked. Other features, such as serial subluxation, odontoid erosion, and involvement of vertebral end-plates, also showed considerable increase. The incidence of osteoarthritis, particularly in those cases also showing rheumatoid features, showed a moderate increase, while the cases showing no radiological abnormality diminished.

Table II *Progression of x-ray changes in 171 RA cases from 1963 to 1969*

X-ray feature	1963	1969
Cases showing RA changes	85	138
AAS	38	79
Serial subluxation	13	26
Local osteoporosis	37	53
Odontoid erosion	5	18
Vertebral end-plate erosion	24	33
Apophyseal joint erosion	62	95
Disc space loss	26	49
Cases showing osteoarthrosis only	53	16
Cases showing no abnormality	33	17

Of the 31 cases showing AAS in 1963, 21 showed no progression, and 27 patients formerly without subluxation developed minor subluxation. Of the 31 who in 1969 showed major subluxation (greater than 5 mm.), there were seven who were involved in about the same degree in 1963, ten who showed minor subluxation, and fourteen who showed no AAS then (see Table III).

Table III *Progression of AAS in 171 RA cases*

1963	1969			Total 1963
	No AAS	'Minor' AAS	'Major' AAS	
No AAS	92	27	14	133
'Minor' AAS	—	21	10	31
'Major' AAS	—	—	7	7
Total 1969	92	48	31	171

Analysing the data, the age of the patient was not related to the development of AAS but the duration of disease activity was important (Table IV). In the 1963 sample, the maximum incidence was achieved in those patients with active disease, of between 5 and 10 years' duration or more. In the 1969 sample, the maximum incidence was found in patients with 11 to 16 years' duration, remaining constant after that time. The incidence in the patients of less than 11 years' duration was higher than the comparable group in the 1963 series, suggesting that the 171 patients surveyed included a high number of chronically active and progressive cases, and that in any sample the incidence figure will continue to rise for at least 15 years from disease onset.

Table IV *Incidence of AAS with increasing duration RA*

Year of onset of RA	Before 1953	1953-1958	1959-1963	Total
No. of patients	62	42	67	171
Per cent. AAS 1963	23	28	18	22
1969	50	54	37	46

Of the 171 patients, 92, including 49 of the 79 cases with AAS, were seen between 3 and 6 years of the original assessment and a further 23 (four cases of subluxation) in the first 3 years of follow-up. None showed evidence of gross neurological change. The

remaining 56 patients were not clinically assessed at this hospital after 1963.

Discussion

In cases of atlanto-axial subluxation with spinal cord or vertebral artery compression, surgical stabilization is obviously important. In our experience such complications have been uncommon and surgery rarely needed. This limited radiological survey shows that in many patients the subluxation stabilizes, and that late major complications of the condition are uncommon. Some of the patients with neck symptoms in this series have been helped by simple immobilization by a collar, but most have had no specific treatment and none has had surgery. Some patients showed progression of subluxation and a surprising number developed subluxation in the 6 years following the original study. The final figure (approximately 50 per cent. of cases showing atlanto-axial subluxation and 80 per cent. showing rheumatoid radiological features) is comparable to the figures of Martel (1961), and Serre, Simon, Janicot, and Lévy (1964).

Many of the questions raised by Sweetnam and others (Sweetnam, 1970) cannot be answered by this limited study, for not all of the survivors from the original study were x-rayed, and not all of those x-rayed were examined clinically. Nevertheless, positive evidence of serious complications was not obtained. The majority of cases with subluxation

have a progressive and destructive type of rheumatoid arthritis, and their prognosis has depended less on the neck abnormality but more on the peripheral joint disease. In none of those who have died has death been directly related to the subluxation.

For these reasons, surgical intervention in the absence of progressive neurological or vascular changes seems unwarranted.

Summary

After approximately 6 years, 171 of 333 rheumatoid arthritic patients previously studied were available for further radiological investigation of the neck. These x rays showed a marked increase in incidence of rheumatoid features, especially atlanto-axial subluxation. No cases of serious neurological complications were found even in those with marked subluxation.

Of the original series of 333 patients, 59 had died; seventeen were known to have had atlanto-axial subluxation, but in no case was death due to this abnormality.

In the absence of neurological or vascular complications surgery seems unwarranted in the management of rheumatoid atlanto-axial subluxation.

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