ACTIVE CONSERVATIVE TREATMENT OF ATLANTOAXIAL SUBLUXATION IN RHEUMATOID ARTHRITIS

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SUMMARY
Twenty patients with rheumatoid atlantoaxial subluxation (AAS) underwent an active conservative treatment programme, aiming at relieving the symptoms and influencing the natural course of AAS. The treatment consisted of intensive multiprofessional intervention in the hospital for a fortnight, and control and motivational visits 6 and 12 months later. The patients self-evaluated the symptoms several times, and the cervical spine radiographs were taken at the first and last visit. Seventeen of the 20 patients adopted the instructions well and undertook the exercise programme at least weekly, even after a 12 month follow-up. Cervical pain was significantly reduced \((P < 0.001)\) during the first intervention, and the favourable effect continued at least 12 months. Promising changes were also seen in some cervical radiographs. It is possible to educate and motivate AAS patients to take active care of their neck, and to relieve their chronic neck pain significantly. Atlantoaxial instability may also be reduced during active conservative treatment.

KEY WORDS: Rheumatoid arthritis, Atlantoaxial subluxation, Cervical spine, Rehabilitation, Treatment.

It is characteristic of rheumatoid arthritis (RA) that the cervical spine becomes affected [1–4]. The most common rheumatoid abnormality in the area is atlantoaxial subluxation (AAS), which has been reported to occur in 29–70% of RA patients, depending on the patient group studied [3]. AAS was found in every third RA patient in a population-based study in Finland, and cervical spine involvement was present in every second case [4]. AAS and other cervical spine disorders may cause severe complications, e.g. tetraparesis and sudden death [1–3]. It has been suggested that these complications can be avoided by early operative treatment [5]; however, rheumatoid cervical spine involvement is so common that early surgery cannot be the only solution to the problem [4, 6].

Most AAS patients do not undergo surgery, but active conservative treatment is rarely given [4, 6, 7] and the effect of passive conservative treatment has been reported to be poor [8–10]. We have developed a programme of active conservative treatment of AAS, based on our clinical and scientific experience, the first results of which are reported.

PATIENTS AND METHODS
The first author (MK) proposed to the Social Insurance Institution of Finland that a new type of active conservative treatment (educational rehabilitation) be initiated for patients with rheumatoid cervical spine problems. The Institution decided to finance treatment for 20 patients during 1995, and an information letter was sent to every Finnish rheumatologist, inviting patients with rheumatoid AAS with or without cervical symptoms to attend.

Our series consisted of the first 20 patients (19 women, one man) who were admitted to treatment; no patient was excluded. Their mean age was 47.9 (s.d. 9.7) yr (range 26–67 yr). All had RA, and the mean duration of the disease was 19.9 (s.d. 8.6) yr (range 3–32 yr). Six had secondary amyloidosis. The disease remained active in most [mean erythrocyte sedimentation rate on admission 32.8 (s.d. 21.6) mm/h]; 19 were on disease-modifying anti-rheumatic drugs, 19 used low-dose systemic corticosteroids and all needed non-steroidal anti-inflammatory drugs. The mean latency period from the physician’s referral to the beginning of the treatment programme was 8.9 (s.d. 4.0) weeks.

The treatment consisted of two in-patient periods (14 days at early treatment and 3 days after 6 months), followed by a control visit 12 months later. The patients were studied clinically during each visit by a rheumatologist and a physiotherapist, and basic laboratory tests were carried out.

The intensity of the cervical and/or occipital pain was studied by the visual analogue scale (VAS). The patients evaluated the intensity of their cervical pain during the last day in the morning, afternoon and night at the beginning of the treatment and after 2 weeks, 6 months and 12 months, and during the last month when they came to each visit; detailed patient histories were also taken. Lateral-view cervical spine radiographs (using a 150 cm tube-to-plane distance) were taken in full flexion, full extension and neutral positions at admission and at the 12 month follow-up. All the radiographs were evaluated by the first author (MK). The shortest perpendicular distance between the anterior arch of the atlas and the dens of the axis was measured and is referred to here as the atlantoaxial (AA) distance. The AA instability is the difference between the AA distance values in flexion and extension radiographs. The AA impaction (AAI = vertical AA subluxation) was evaluated by the Sakaguchi–Kauppi method [11]. Friedman’s two-way analysis of variance and Bonferroni-adjusted paired sample confidence interval were used in the statistical analysis.
The Spearman correlations were counted of the associations between the pain values (VAS) at different times of the day. The probability of rejecting the null hypothesis when it is false was estimated by the power analysis in the study. The criterion for significance ($\alpha$) was set at 0.05 (two-tailed).

**ACTIVE CONSERVATIVE TREATMENT**

Active conservative treatment of the rheumatoid cervical spine has two aims: (1) to relieve the symptoms and (2) to halt or retard the progression of abnormalities seen in the radiographs. Ankylosis between subluxated vertebrae may develop spontaneously and it is probably encouraged by conservative treatment [12]. The treatment consists of six elements: patient education, physiotherapy, collars, practical aids, symptomatic treatments and active disease-modifying medication [6–7].

Patient education is the basis of conservative treatment [7, 13–14], and much attention was devoted to it. The patients were taught individually and in small groups about cervical spine anatomy, rheumatoid changes and treatment methods by a rheumatologist, physiotherapists and occupational therapists; the guidelines for patient information have been published earlier [7]. The main point is that the flexion position is harmful in cases with unstable AAS and patients should avoid it. Ergonomics was taught in addition to traditional methods, but video was also used. Videofilms were taken of the patients' everyday activities and evaluated by the patients and staff.

Physiotherapy was given individually, and each patient received individual instructions for home exercises. Cervical pains in AAS cases are mostly of muscular origin and thus the treatment aimed at reducing muscular tension and inducing their fitness [7]. We suppose that the six small muscles of the occipital triangle are especially important in AAS cases. They interconnect the occipital bone, atlas and axis, and extend and rotate the head, but are also postural muscles responsible for fine movements of the head and they cooperate with the eye muscles [15, 16]. Much effort was aimed at the strength and endurance of these and their antagonists, the deep flexors. We taught the patients to do isometric or only slightly dynamic exercises (flexion, extension, rotation) against compliant resistance, such as a finger, etc. The exercises were performed, using a pumping method, with a tension followed by a rest before the next tension. Short and longer (~8 s) tensions were used in successive series to activate all types of muscle cells [17–18]. The number and frequency of series carried out were planned individually. The patients were intensively taught to feel and use the small deep muscles of the upper neck in the exercises. Exercises of the larger neck and shoulder muscles were also performed, but dynamic flexion and maximal rotation were avoided. One aim was the maintenance of good posture, and exercises were performed to improve the posture of the entire body, as well as of the neck. Muscle relaxation methods were also taught to the patients.

A stiff custom-made collar was made for each patient; the instructions for use have been published earlier [7]. Practical aids, such as book rests, were shown and recommended to the patients as part of their ergonomic education. Symptomatic medication was optimized, and in several cases local corticosteroids were injected into the painful muscle insertions in the occipital and neck areas, while muscular tension was also usually reduced by gentle massage. RA inflammatory activity was evaluated, and in four cases the disease-modifying medication was changed to more active therapy. No changes were made to the systemic corticosteroid treatments. Corticosteroid injections were given to inflamed extremity joints in several cases. The patients also participated in more general rehabilitation exercises, e.g. water gymnastics, etc.

The first part of the treatment (14 days) was very intensive, and we attempted to motivate the patients to follow our instructions. Their treatment compliance was self-evaluated using questionnaires after 6 and 12 months. The patients kept a treatment diary during the first 6 months, and in the first control they evaluated by VAS how well they had been able to carry out the programme. The individual treatment instructions were re-evaluated and sometimes slightly improved during the 6 month follow-up visit. The patients were also instructed to continue treatment after the end of follow-up.

**RESULTS**

Eight of the patients had no collar, and only nine had used a collar at least once weekly, when they attended the active conservative treatment programme. After the first part of the treatment, most patients used the stiff collar at least weekly during the follow-up (Table 1). We did not insist on daily use if the patients were aware of the importance of good posture and ergonomics. Most had followed our exercise programme at least weekly (Table 1), and many said that they increased the frequency if they felt neck pains. The patients undertook self-evaluation by VAS of how well they had been able to carry out the whole treatment programme (0 mm = not at all; 100 mm = totally). The mean VAS was 51.5 (s.d. 23.2) mm (range 7–84 mm), but in three cases the VAS was < 15 mm. There were no drop-outs.

All the six aspects of pain (morning, afternoon, night; during the last day and last month) had a statistically significant correlation with each other. Individual means of the three VAS values of pain during the last day were used in the evaluation of the pain relief. The mean of the individual means of VAS was 41.5 mm (s.d. 24.9) at the early treatment and was reduced by 21.6 mm [95% confidence interval (CI) –36.8 to –6.4] after 2 weeks; the reduction after 6 and 12 months was 16.8 mm (95% CI –30.6 to 2.9) and 23.9 mm (95% CI –38.1 to –9.7), respectively. The study (the 12 month pain reduction) will have the power of 99% to yield a statistically significant result. The pain was significantly reduced during the first 2 weeks of the treatment and the good effect continued at least 12 months ($P < 0.001$) (Fig. 1). One patient was practically pain free (severity of pain by VAS < 5 mm in all six questions) early on and during follow-
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TABLE I
Treatment compliance during follow-up. The frequency indicates how often the patients used custom-made stiff collars and performed muscle exercises.

<table>
<thead>
<tr>
<th>Use of the custom-made stiff collar</th>
<th>Muscle exercises</th>
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<tbody>
<tr>
<td>Before 6 months</td>
<td>12 months</td>
</tr>
<tr>
<td>Daily</td>
<td>5</td>
</tr>
<tr>
<td>Every 2 days</td>
<td>3</td>
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<td>Once weekly</td>
<td>1</td>
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<tr>
<td>Sometimes</td>
<td>1</td>
</tr>
<tr>
<td>Not at all</td>
<td>10*</td>
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</table>

*Eight patients had not had collars before treatment.

The mean AA distance in the flexion radiographs was 7.0 (s.d. 2.1) mm (range 3–10 mm) and the mean AA instability was 5.0 (s.d. 2.9) mm at the beginning. The AAS was stable in three cases, all of whom also had AAI; AAI was also seen in two additional cases. In neutral-position radiographs, the AAS was absent in nine cases. Two patients had a subaxial subluxation of 3 mm and several had some degenerative changes in their cervical spine. In follow-up flexion radiographs (12 months later), the AAS had disappeared in two cases (one of whom had AAI). In these and in one additional case (with AAI), the instability had been reduced 3 mm since the beginning. Minimal reduction of the instability was seen in some other cases. No significant radiological change occurred in the subaxial area during the follow-up.

**DISCUSSION**

No previous reports are available of active conservative treatment of patients with rheumatoid cervical spine disorders. This treatment aims at relieving the symptoms and influencing the natural course of AAS. Cervical pain is a common complaint among rheumatoid patients. It is mostly caused by muscular tension, which may be combined with tissue inflammation, cartilage damage and compression of neural structures [1–3, 7, 19]. Effective pain relief would reduce the need for cervical spine operations, since pain is their frequent indication [7, 20–22]. Optimal conservative treatment would also stop or retard radiological progression of the abnormalities, or even possibly reduce the instability, which would reduce the risk of complications [6–7, 12]. Thus, the treatment has to be multidisciplinary: the basis is patient education, but collars, ergonomics, practical aids, muscular exercises, massage, anti-inflammatory and analgesic drugs, local injections and active disease-modifying medication together form the combination, which may be able to fulfil the aims of active conservative treatment.

All our patients showed permanent rheumatoid changes in the AA area and some also subaxially, and most experienced neck pains. Although the patients were primarily treated by rheumatologists, 10 did not use a stiff collar when they began the treatment, and none had earlier participated in special neck exercise programmes for rheumatoid cervical spine. The rheumatoid inflammation was active in several patients and all had permanent changes in their peripheral joints. Thus, most of them also complained of other problems, in addition to those of the cervical spine.

Our basic result is that with intensive education and treatment it is possible to change the habits of patients with rheumatoid cervical spine disorders. The treatment team was multiprofessional and very interested in cervical problems, which helped to motivate patients to participate actively in the treatment. Most of our patients followed the instructions rather well, continuing as long as 12 months after intervention (Table I), and thus the changes in symptoms and radiographs were probably induced by the active conservative treatment.

Most patients had long-lasting cervical pain before treatment; none had only acute pain, and thus spontaneous pain relief was not probable. The pain was significantly (statistically and clinically) relieved during the first 14 days of treatment in most patients, and the
effect continued for at least 12 months. This probably occurred for several reasons, but the patients were especially content with the means they had of controlling their cervical pain.

Atlantoaxial ankylosis may develop spontaneously, especially in cases with AAI and rough, eroded surfaces of the AA facet joints [12]. The treatment aims to teach patients to avoid situations with maximal subluxation, and to induce fitness of the stabilizing muscles of the area, and thus to encourage the development of spontaneous ankylosis (or reduction in the AA instability). Three patients had a stable AAS at early treatment, and in these cases no radiographical improvement was possible [12]. In three cases, the AA instability had been reduced by 3 mm during the follow-up. In two of them, the reduction in AAS was associated with prevalence of AAI, and in at least these cases the change was probably permanent.

The natural course of AAS is a multifactorial phenomenon that cannot be precisely predicted in small series such as the present one. Longer follow-up of these patients would be needed to evaluate the true prognosis in the AA area, but the present results are promising. This was a pilot study, and further research is needed to confirm the findings and also to improve the treatment methods continuously. A controlled trial would be beneficial; however, it will be difficult to find sufficient controls matched for pain, inflammation activity, radiology, age, etc. Those AAS patients in our hospital who do not attend our special cervical spine programme will also receive instructions for active treatment. Simple instructions may be given in a few hours, but more time is needed to teach them properly and to motivate the patients to adapt to changes in their behaviour.

We conclude that it is possible to educate and motivate AAS patients to take active care of their neck, relieve their chronic cervical pain and probably influence the natural course of the rheumatoid cervical spine. Some kind of active conservative treatment should be available, as a part of clinical routine, for all patients with AAS, preferably in early cases to prevent the development of severe AAS with the risk of complications and to reduce the need for operative treatment.

Acknowledgements

We are grateful to our skilful occupational therapists, Raija Lind, Johanna Rontti and Hanna Tallgren, for their enthusiastic work in rehabilitation. The treatment programme was funded by the Social Insurance Institution of Finland.

References