Subaxial cervical spine subluxation in rheumatoid arthritis

A retrospective analysis of 16 operated patients after 1–5 years

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We evaluated the clinical and radiological outcomes in 16 patients (15 women) having rheumatoid arthritis with a mean age of 66 (55–77) years, on average 2 (1–5) years after decompression and stabilization, for subaxial subluxation of the cervical spine. The duration of rheumatoid arthritis averaged 30 (10–67) years and the duration of neck symptoms averaged 15 (1–60) months. Preoperatively, 11 of the patients had pain in the neck, all 16 suffered from arm rhizopathy and varying degrees of myelopathy.

4/5 patients with severe myelopathy died within 3 months of surgery. Fixation failure occurred in 7 patients, but had no clinical significance in 5. There were 1 deep infection and 1 nerve root lesion resulting in deltoid weakness. Other complications were dysphagia and donor-site pain. 4 reoperations were performed, 2 extension of fusion, 1 revision of infection, and 1 foraminotomy.

Neck pain was reliably relieved, while arm rhizopathy was less positively affected. Myelopathy carried a poor prognosis for relief and its occurrence correlated with death. Early treatment, before significant myelopathy has developed, is recommended. Decompression, both via realignment and bone resection, followed by fusion of the entire cervical spine, is advocated. Due to the poor bone quality and with the presently available implant systems, simultaneous anterior and posterior fixation is beneficial.

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Subaxial subluxation of the cervical spine (SAS) in rheumatoid arthritis presents as local and radiating pain, arm rhizopathy, and/or myelopathy. The condition may progress towards tetraplegia. Cervical spinal cord compression may be one of the major causes of premature death in this population (Mikulowski et al. 1975, Lipson 1989, Boden 1994).

The destruction of joints and discs results in multiple olistheses, stepladder deformity, sometimes interrupted by spontaneously ankylosed segments. Hypermobility may occur in one or a few segments in-between the stiff areas. Neural compression can occur anteriorly by the upper corner of the vertebral body distal to a slip, posteriorly by the neural arch of the slipped vertebra or by rheumatoid pannus, especially around hypermobile areas (Kataoka et al. 1979, Kudo and Iwano 1991) (Figure 1).

The cornerstones of treatment are decompression by reduction of deformity and/or resection of compressing tissue, and stabilization. By achieving bony fusion, we reduced the risk of recurrence of SAS due to fatigue failure of the implants.

We report our clinical and radiographic results of treating SAS according to the above described principles.

Patients and methods

16 consecutive patients (15 women) were treated for SAS at our department between 1990 and 1994 (Table 1). At operation, the patients had a mean age of 66 (55–77) years, and the duration of rheumatoid arthritis was 30 (10–67) years. They had suffered from neck symptoms for 15 (1–60) months.

6 patients had previously had an operation on the cervical spine because of instability: 1 had an occipito-cervical fusion, 3 had fusions from C1 to C2 , 1 had a fusion from C4 to C7, and 1 patient had a pseudarthrosis at C3 combined with instability at lower cervical levels, after an attempted fusion from occiput to C5.

Preoperatively, 11 patients suffered from local pain in the neck. 6 patients complained of radiating arm pain. All 16 patients had various degrees of neurological symptoms from the arms (e.g., numbness or weakness caused by neurological dysfunction). All 16 patients had various degrees of myelopathy (Table 1).

The operation was performed with a 3 kg skull traction to help reduce the deformity. Decompression was achieved by spinal realignment and by resection of soft tissue and bone compressing the neural struc-

Figure 1. Case 13.

Subaxial subluxation of the cervical spine. Note the stepladder deformity and the ankylosed areas.

Lateral T1-weighted MRI. The spinal cord is pinched between the posterior upper corner of the vertebra under the slip and the neural arch of the slipped vertebra.

tures. 2 of the patients were operated on posteriorly, 5 anteriorly, and 9 both posteriorly and anteriorly. AO reconstruction plates and 4 mm screws were used for posterior fixation and AO Orozco plates and 4 mm screws were used for the anterior fixation. Autologous iliac bone grafts were used in all patients.

Postoperatively, all patients were treated in a custom-made soft collar (Milbrink and Wigren 1989) for 6–12 weeks.

Follow-up. All available pre- and postoperative cervical spine radiographs, CT scans, and MRI scans were examined by a radiologist for preoperative pathology, and postoperative results: reduction of slips, spinal alignment, failure of fixation, and union of fusion (Table 1). For all 16 patients, the records of pre-, per-, and postoperative details were reviewed with special emphasis on pain, neurological symptoms, complications, and reoperations.

Follow-up was performed 2 (1–5) years postoperatively by an independent observer. 9 of the 16 patients who were still alive at the time of follow-up were examined for local neck pain, radiating arm pain, neurological dysfunction of the arms, signs of myelopathy, and the patients' own opinion of their functional performance. Pain was classified according to the following arbitrary scale: 0 no pain, 1 mild pain (generally well tolerated by the patient, requiring little, if any, analgesics), 2 moderate pain (significant problem to the patient, requiring regular use of oral analgesics), 3 severe pain (intractable pain, not responding to oral analgesics).

Neurological involvement of the arms was classified as follows: 0 normal neurology, 1 mild impairment (e.g., mostly subjective complaints of numbness and motor weakness), 2 moderate impairment (e.g., discrete objective findings of sensory disturbance and motor weakness), 3 severe impairment (e.g., significant objective sensory disturbance and paresis).

Myelopathy was classified according to Ranawat et al. (1979): I) normal function, II) subjective weakness and numbness, increased reflexes, IIIA) objective signs of myelopathy, ambulating, and IIIB) objective signs of myelopathy, not ambulating.

Information regarding the outcome of the 7 patients who had died at the time of follow-up was gathered from the records and by interviewing relatives.

Table 1. General table concerning 16 patients with rheumatoid arthritis operated for subaxial subluxation of the cervical spine

A	B	C	D	E	F	G	H	I	J	K	L	M	N	O	P	Q	R	S	T	U	V	W	X	Y	Z	AA	AB
1	F	55	16	0		0	1	2	3	4.10	3-5		0	0	1	0			1	55	0	0	1	3	3	1	2
2	M	66	25	0-2	60	2	0	2	3		3-7		0	0	1	1		1	0	21	1	0	1	3		0	1
3	F	72	40	0	2	2	0	3	4	6.30	4-7	4-7	0	1	2	1		2	0	46	1	0	2	3		1	2
4	F	59	10	0	48	1	0	2	2	1.55	4-5		0	0	1	1			2	45	0	0	0	1	1	1	2
5	F	74	61	0	1	2	2	3	4	3.25	4-5	1-2	0	0	1	0		1	0	1						1	
6	F	70	25	0		0	0	2	3	4.55	5-7		0	0	2	1		3	0	25	0	0	0	1		1	2
7	F	58	11	1-2	2	2	2	3	3	5.15	3-6	1-8	1	0	1	1	1,2		3	26	1	0	3	3	5	1	2
8	F	63	30	0		0	0	3	3	4.10	4-8	4-8	0	1	2	0			0	20	0	0	3	4	5	1	2
9	F	77	67	0		0	0	3	4			2-7	1	0	1	0		1	0	3						1	
10	F	61	40	0-5[a]	2	3	2	2	3	3.15	5-9		0	0	1	1	1		4	18	0	0	0	1	2	1	2
11	F	69	21	0		0	0	2	3	7.05	3-6	3-6	0	1	1	0	3		0	15	0	0	1	2	2	1	2
12	F	67	32	4-7	8	2	0	3	4	2.20		1-9	1	0	2	1		1	0	3						1	
13	F	66	22	1-2	30	2	2	1	2	5.00	3-8	1-9	1	0	2	0			0	12	0	0	1	2	2	0	1
14	F	63	27	1-2	2	1	1	3	4	4.30	2-8	1-8	1	1	1	0		2	0	2						1	
15	F	73	38	0	12	1	0	2	3	6.20	2-8	1-8	1	1	1	0	4		0	12	0	0	0	3	3	1	2
16	F	56	22	0	1	2	0	1	2	5.30	2-8	1-8	1	1	1	0	4,5		0	14	0	0	0	1	1	1	2

A Patient number
B Sex
C Age
D Duration of rheumatoid arthritis, years
E Previous cervical spine fusions (count from C1)
  [a] indicates non-union
F Duration of neck symptoms, years
G Neck pain
  0 no pain
  1 mild pain
  2 moderate pain
  3 severe pain
H Radiating arm pain
  0 no pain
  1 mild pain
  2 moderate pain
  3 severe pain
I Neurological involvement of arms
  0 normal neurology
  1 mild impairment
  2 moderate impairment
  3 severe impairment
J Myelopathy, Ranawat class
  1 I-normal function
  2 II-subjective weakness, hyperreflexia, dysesthesia
  3 IIIa-objective weakness, long tract symptoms, able to walk
  4 IIIb-objective weakness, long tract symptoms, not walking
K Operation time, hours, minutes
L Level of anterior fusion (count from C1)
M Level of posterior fusion (count from C1)
N Entire cervical spine fused
  0 no
  1 yes
O Same anterior and posterior levels fixed
  0 no
  1 yes
P Residual anterior displacement
  1 0–2 mm
  2 3–6 mm
Q Loss of fixation
  0 no
  1 yes
R Complications
  1 loss of fixation requiring revision
  2 infection
  3 C4 root lesion
  4 transient dysphagia
  5 transient donor site pain
S Cause of death
  1 general weakness
  2 cervical spinal cord compression
  3 myocardial infarction
T Reoperations
  1 foraminotomy
  2 fusion extension C3–C6
  3 revision C1-Th1
  4 fusion extension C1–Th2
U Follow-up time or time to death, months
V Neck pain at follow-up
  0 no pain
  1 mild pain
  2 moderate pain
  3 severe pain
W Radiating arm pain at follow-up
  0 no pain
  1 mild pain
  2 moderate pain
  3 severe pain
X Neurological involvement of arms at follow-up
  0 normal neurology
  1 mild impairment
  2 moderate impairment
  3 severe impairment
Y Myelopathy at follow-up, Ranawat class
  1 I-normal function
  2 II-subjective weakness, hyperreflexia, dysesthesia
  3 IIIa-objective weakness, long tract symptoms, able to walk
  4 IIIb-objective weakness, long tract symptoms, not walking
Z Patients' own evaluation at follow-up
  1 fully recovered
  2 major improvement
  3 minor improvement
  4 unchanged
  5 deterioration
AA Spinal alignment
  0 unsatisfactory
  1 satisfactory
AB Fusion healing
  0 definitely not healed
  1 no evident healing disturbance
  2 definitely healed

Results

Mortality. All 4 patients who died within 3 months of the operation had myelopathy grade IIIB preoperatively: 1 patient died suddenly 2 months after the operation, due to tetraplegia. Autopsy suggested that the cause of death was cervical spinal cord compression secondary to failure of fixation. The other 3 patients died of general weakness, without any further specification. Of the 3 patients who died later than 3 months after the operation, 1 died of myocardial infarction, 1 of general weakness and 1 of a spinal cord compression, probably at a previously nonfused cervical level.

Failure of fixation occurred in 7/16 patients. 1 of the 7 was the above-described patient who died of tetraplegia 2 months postoperatively (case 14). 1 of the 7 had a concomitant deep wound infection and required revision (case 7). The failure of fixation was of no consequence in 5/7: One or a few screws had backed out. No major increase in deformity was noted and the fusions healed, in spite of the loose implants.

Other complications. 1 patient developed a C4 nerve root lesion with deltoid muscle weakness. 2 patients experienced transient dysphagia, and 1 suffered from transient pain at the donor site.

Reoperations. 4 patients in the series were reoperated. In 2 patients initially treated with short fixations over a few segments only, fusion extensions had to be performed due to progress of the SAS to adjacent levels; in patient no. 4 from C3 to C6 and in patient no. 10 from C1 to Th2. The subsequent fusion-healing was uneventful for both patients. The above-described patient (case 7) with failure of fixation and deep wound infection was successfully revised and restabilized. She was also treated with adjuvant halo-vest fixation for 4 months. 1 patient (case 1) had a posterior foraminal decompression due to persistent numbness of 1 hand—however, without clinical improvement.

Radiological review. The radiological review of the pre- and postoperative radiographs of all the 16 patients revealed that in 11 the forward slips had been reduced to a great extent (0–2 mm residual slip), whereas a small slip (3–6 mm) was still present in 5. In no case did the slip exceed 6 mm.

The overall spinal alignment was judged on an arbitrary scale as good in 14 patients and poor in 2. In the poor cases, the spine was fused in kyphosis.

The fusions were regarded as solid in 10 patients (e.g., continuation of bone trabecles extending from one vertebra to the next or mature fusion mass) and showed no signs of healing disturbance in 2 (e.g., no motion and no change in position of internal fixation devices). No patient developed pseudarthrosis. 3 of the 4 patients who died within 3 months showed no signs of healing disturbances, whereas 1 patient died because of cervical spinal cord compression as a result of fixation failure.

Pain, neurology and functional performance. Neck pain was relieved to a great extent. The 4 patients without neck pain remained pain-free. 5 patients experienced 1 arbitrary unit improvement, 2 patients experienced 2 arbitrary units improvement, and 1 patient experienced 3 arbitrary units improvement. Arm pain disappeared in all patients. Arm neurology was relieved to a lesser degree than neck pain. 3 patients did not improve. 5 patients experienced 1 arbitrary unit improvement, and 4 patients experienced 2 arbitrary units improvement. No patient improved more than 2 arbitrary units.

The patients recovered poorly from myelopathy. As described above, 4/5 patients with severe myelopathy (Ranawat class IIIB) died within 3 months of the operation. In the 1 surviving patient (case 3), fusion healed. She initially improved to class IIIA, but deteriorated later to class IIIB and died at 46 months from spinal cord compression. No radiographic or autopsy documentation is available regarding the exact level of spinal cord compression in the second episode of deterioration.

Of the 12 patients available for outcome evaluation, 1 patient had deteriorated 1 Ranawat class, 5 patients remained unchanged, 4 patients improved 1 class, and 2 improved 2 classes. No patient recovered 3 classes.

The patients' own evaluation of the outcome was available for 9 patients. 2 of the 9 patients claimed that they had recovered fully, 3 had made a major improvement, 2 had made a minor improvement, none was unchanged and 2 reported that they had deteriorated.

Discussion

It is generally agreed that the prognosis for recovery of severe myelopathy is poor; that is why early operation, before irreversible neurological damage has developed, is advocated (Peppelman et al. 1993, Boden 1994). This is fully supported by our study, since recovery from severe myelopathy was poor, with a high mortality rate due not only to neural damage but also to general weakness as reported by others (Zygmunt et al. 1988, Grob 1993).

Extent of fusion. Although some authors state that progression of subluxation seldom occurs at levels adjacent to fusions (Krieg et al. 1993), most authors consider subaxial subluxation in rheumatoid arthritis to be a progressive condition. Especially segments distal to fusions seem to deteriorate rapidly. Agarwal et al. (1992) reported development of SAS in 8 of 22 patients fused from the occiput to C3 (95% confidence interval of 20–57%). Stirrat and Fyfe (1993) reported that additional surgery was required in 4 of 12 patients with subaxial fusions. Similar experience was reported by others (Conaty and Mongan 1981, Bryan et al. 1982, Zoma et al. 1987, Zygmunt et al. 1988, Santavirta et al. 1990, Kraus et al. 1991).

Nevertheless, most authors advocate fusion of affected segments only (Conaty and Mongan 1981, Grob et al. 1993). In our series, short fusions were not very successful. Taking this into account, we now prefer to fuse the entire cervical spine in patients with rheumatic subaxial subluxation, especially as our experience is that a totally fused cervical spine in good

Table 2. Relationship between type of treatment and failure of fixation. Data on patients with ankylosing spondylitis and cervical spine fractures from a previous series (Olerud et al. 1996) are pooled with data from the present series

Type of treatment	Loss of fixation [a]	No loss of fixation [a]
One side only	(3+5) 8	(7+2) 9
Double fixation [b]	(0+1) 1	(7+5) 12

[a] (data from Olerud et al. (1996) + data from present series). The 3 patients who died before 3 months from causes not related to the present operation are omitted; the patient who died at 2 months from failure of fixation is included
[b] Double fixation means that the same levels of the spine had been fixed both anteriorly and posteriorly
p = 0.024, Fisher's exact test

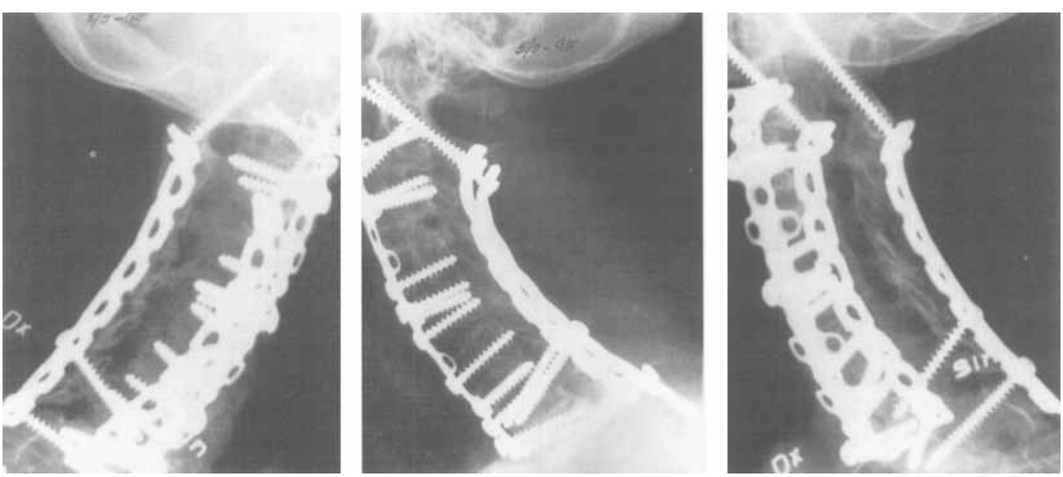
balance is well tolerated by the patient. Similar views have also been presented by Kraus et al. (1991).

Fixation technique. In contrast to the report by Grob et al. (1993), with only 1 patient having a loose implant in a series of 23, a high proportion of the patients in our series had loose implants. Apparently, the same screw technique and implants were used in both studies and we have no explanation of this difference. However, just as in other chronic inflammatory diseases, a major problem with the rheumatoid spine is poor bone quality caused by inflammation, steroid medication (Spector et al. 1993), and possibly atrophy from inactivity. The osteoporosis makes internal fixation problematic, whatever technique is selected. Screws, as used in the present series, may loosen, but also sublaminar wires (Glynn and Sheehan 1983) and hooks fail by cutting out.

In a previous series of spine fractures in patients with ankylosing spondylitis, 17 patients with cervical fractures were presented (Olerud et al. 1996). 3 fixation failures occurred among 10 patients fixed either anteriorly or posteriorly, whereas none of 7 patients fixed simultaneously from both sides failed. As in the present study, this series was small. However, if the data from both series are pooled, it is evident that the incidence of fixation failure is reduced when the spine is fixed from both sides (Table 2). Thus, in contrast to others (Conaty and Mongan 1981, Zoma et al. 1987, Heywood et al. 1988, Grob et al. 1993, Stirrat and Fyfe 1993), we recommend simultaneous anterior and posterior fixation in patients with rheumatic conditions. Posteriorly, the fusion is extended from C1 (or the occiput in the presence of occipitocervical instability) to Th1 or Th2. We prefer the C2-C1 trans-facet joint screw described by Magerl as the upper anchorage (Magerl and Seeman 1986, Jeanneret and Magerl 1992). In the lower cervical and upper thoracic spine, pedicle screws (Heywood et al. 1988, Abumi et al. 1994) at 2 levels have proved effective. Additional grip can be achieved with short screws in-between, either into the facets or through the facet joints. Anteriorly the fusion is extended from C2 to the same level as the posterior fixation (Figure 2).

We used AO pelvic reconstruction plates posteriorly to connect the anchors. This is sufficient, but not ideal, as the loose connection between the screws and the plate predisposes to mechanical failure. Another drawback is that the holes in the plate determine the placement of the screws. We are presently developing a rod-based fixation system, which will allow screws to be inserted in different directions, and with which hooks and sublaminar wires also can be added (Whitaker 1995) (Figure 3).

Figure 2. Case 15.



Postoperatively. The cervical spine has been fused from C1 to Th1 posteriorly and from C2 to Th1 anteriorly. Note the normal alignment of the spine.

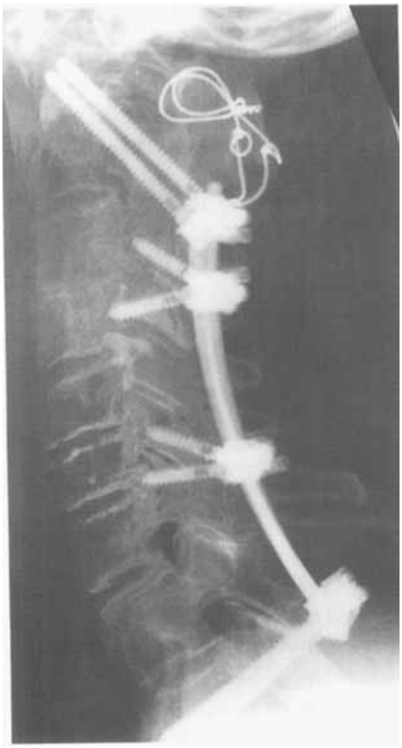


Figure 3. The newly developed spinal fixation device applied to a patient with rheumatoid arthritis and subaxial subluxation.

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