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Stage-related surgery for cervical spine instability in rheumatoid arthritis

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Abstract Thirty-six consecutive patients with cervical spine instability due to rheumatoid arthritis (RA) were treated surgically according to a stage-related therapeutic concept. The aim of this study was to investigate the clinical results of these procedures. The initial change in RA of the cervical spine is atlanto-axial instability (AAI) due to incompetence of the cranio-cervical junction ligaments, followed by development of a peridontoid mass of granulation tissue. This results in inflammatory involvement of, and excessive dynamic forces on, the lateral masses of C1 and C2, leading to irreducible atlanto-axial kyphosis (AAK). Finally, cranial settling (CS) accompanied by subaxial subluxation (SAS) occurs. According to these three separate pathological and radiological lesions, the patients were divided into three therapeutic groups. Group I comprised 14 patients with isolated anterior AAI, who were treated by posterior wire fusion. Group II comprised 15 patients with irreducible AAK, who were treated by transoral odontoid resection. The fixation was done using anterior plating according to Harms in combination with posterior wire fusion according to Brooks. Group III comprised seven patients with CS and additional SAS, who were treated with occipito-cervical fusion. Pre- and postoperatively, evaluation was performed using the parameters pain (visual analog scale), range of motion (ROM), subjective improvement and Health Assessment Questionnaire (HAQ). The neurologic deficit was defined according to the classification proposed by Ranawat. Radiographs including lateral flexion and extension views, and MRI scans were obtained. The average clinical and radiographic follow-up of all patients was 50.7 ± 19.3 months (range 21–96 months). No perioperative fatality occurred. Postoperative pain was significantly relieved in all groups ($P < 0.001$). In group II a slight improvement in the HAQ was obtained. In groups I and II the ROM of all patients increased significantly (average gain of motion in group I: 11.3° ± 7.8° for rotation; 7.8° ± 5.6° for bending; average gain of motion in group II: 21.5° ± 14.0° for rotation; 17.2° ± 5.5° for bending), while it decreased significantly in group III (10.7° ± 18.1° for rotation; 6.7° ± 18.5° for bending). Preoperatively 27 patients had a manifest neurologic deficit. At follow-up four patients remained unchanged, all others improved by at least one Ranawat class. All patients, except one, showed solid bony fusion. According to the significantly improved postoperative subjective self-assessment and the clinical and radiological parameters, transoral plate fixation combined with posterior wire fixation after transoral odontoid resection represents an effective reliable and safe procedure for the treatment of irreducible AAK in rheumatoid arthritis.

Key words Atlanto-axial instability · Transoral approach · Decompression · Rheumatoid arthritis · C1/C2 fusion

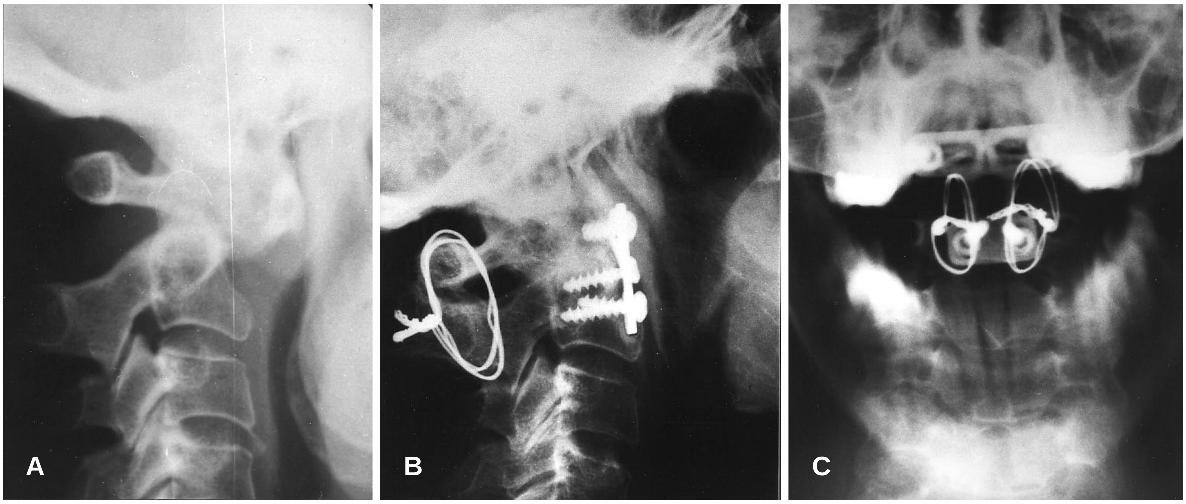

Fig. 1 A–C Patient 5, group II: a 48-year-old woman with a 24-year history of rheumatoid arthritis. **A** Preoperative lateral radiograph showing severe atlanto-axial kyphosis (AAK). Neurological deficit was Ranawat class III B and pain was 9.7 points on the visual analog scale. **B** Postoperative lateral radiograph 57 months after surgery, showing accurate reduction and realignment. Neurological deficit was Ranawat class II and pain was 3.9 points on the visual analog scale. **C** Postoperative anterior-posterior radiograph 57 months after surgery. MR image (*not shown*) showed a retrodental pannus compressing the spinal cord

Introduction

The cervical spine is a common focus of destruction in rheumatoid arthritis (RA). The resultant instability and neural compression are known severe complications.

The transverse ligament represents a structure crucial in preventing forward shifting of the atlas over the axis [18]. In RA this ligament can become edematous and lax due to direct involvement or secondary to spread of inflammation from its synovial articulation with the dens [15]. However, a complete discontinuity loss of this ligament allows a forward movement of the atlas over the axis of 4–5 mm only, further displacement being prevented by secondary stabilizers including the alar ligaments and the capsules of the occipito-atlanto-axial joints [18]. The peridontoid granulation tissue derives from the synovial lining of the articulations around the odontoid. It is most pronounced in patients with severe atlanto-axial instability (AAI) [46], but interestingly it is reduced when the atlanto-axial movement is restricted by surgical procedures [36] or superior migration of the odontoid [46]. Thus, it appears that the volume of the peridontoid soft tissue mass was influenced by the active motion due to the severity of the AAI and not by the underlying inflammatory process [46]. This soft tissue mass can contribute to compression of the upper cervical cord and involves the surrounding secondary stabilizers. In this first stadium of the disease the ligamentous insufficiency can produce severe AAI, the anterior column still being intact.

Dynamic forces generated by the weight of the skull, osteoporosis, and progressive inflammatory degradation of cartilaginous and bony structures of the lateral atlanto-axial joints [28] lead to a destruction of the anterior column at C2, resulting in progressive atlanto-axial kyphosis. During the second stage the atlas slips gradually over the axis in an anterior-caudal direction and produces a larger upward movement of the posterior arch of the atlas, resulting in a greater anteverted C1/C2 kyphosis angle (Fig. 1). The peridontoid synovial pannus, ossifications or scarring between the odontoid and the axis, and an atlanto-axial joint impaction may prevent the reduction by closed treatment such as skull traction.

Secondarily this feature is often accompanied by superior migration of the odontoid (SMO), resulting from collapse of the anterior column, especially involving the lateral masses of the axis and the atlas. This involvement of the occipito-atlantal articulation corresponds to the third stage and has also been described as cranial settling [14]. Subaxial subluxation, which also represents a late phenomenon of the disease, is due to erosions of the facet joints along with inflammatory destructive changes in the intervertebral disc. Deformity involving multiple subaxial levels is common.

Therefore, the progressive destruction of the occipito-atlanto-axial joints in RA seems to require a stage-related surgery. Our therapeutic concept of stage-related surgery depends on the status of the anterior column. The aim of this study was to analyze the clinical results achieved with this concept.

Materials and methods

Indications

General indications for surgery were intractable pain and/or a manifest neurologic deficit. Surgical management was also performed when the posterior atlanto-dental interval (PADI) during lateral standard radiography was 14 mm or less and MRI demonstrated a cord diameter in flexion of less than 6 mm [4].

Therapeutic concept

According to the aforesaid, three clinical and radiographic patterns of cervical spine instability in RA were observed.

Group I: In early stages of RA isolated anterior reducible atlanto-axial instability (AAI) with an intact anterior column was found.

Group II: In advanced stages irreducible atlanto-axial kyphosis (AAK), resulting from destruction of the anterior column, especially the lateral atlanto-axial joints, was registered.

Group III: In late stages additional involvement of the occipito-atlantal joints resulting in cranial settling (CS) often accompanied by subaxial subluxations (SAS) was found.

Corresponding to the different instability types, three different surgical procedures were performed.

Group I

In AAI, atlanto-axial fusion was performed using a standard wiring technique according to Gallie [21] (two patients) or Brooks [5] (12 patients).

Group II

In AAK, isolated anterior atlanto-axial plate fixation described by Harms et al. [30] was performed in the first three patients of this series. The other 12 patients underwent anterior plating in combination with posterior wire fusion according to Brooks [5] (Harms-Brooks procedure). This method involved an anterior transoral approach with exposure of the anterior aspect of the atlas and the odontoid process using a midline incision of the posterior wall of the pharynx. After vertical excision of a 10- to 14-mm-wide portion of the anterior arch of the atlas by micro-saw, the odontoid was removed by a high-speed diamond burr. The granulation tissue and the pannus around the odontoid were removed with rongeurs. While the atlantoaxial lateral joints were exposed, the articular cartilage was excised with a high-speed burr to unlock, reduce, and finally fuse the joints. Concerning restoration of the height of the atlanto-axial joint, if necessary, anterior iliac bone grafting was performed. After reduction, the Harms plate [30] was positioned and fixed with cancellous 2.0-mm screws. Posterior wire fusion according to Brooks [5] was done immediately after completion of the anterior procedure as a one-stage procedure.

Group III

In CS accompanied by SAS occipito-cervical fusion employing various techniques was performed. The extent of occipito-cervical fusion depended on the level of subaxial subluxation.

Postoperative treatment

A 24-h postoperative observation of neurologic function and airway control in the intensive care unit was standard. After transoral surgery intravenous antibiotics were given for 5 days after surgery, and nasogastric nutritional support was started on the morning of the 3rd postoperative day. Oral diet was usually started on the 7th postoperative day. Head immobilization was achieved with a cervical collar for at least 4 weeks (group I: average 7.9 ± 1.4; range 6–16 weeks; group II: average 6.4 ± 1.6; range 4–12 weeks; group III: average 10.8 ± 2.3; range 6–18 weeks), depending on intraoperative assessment of bone quality.

Complications

Group I

With the Gallie method one painfree pseudarthrosis was found, which did not require further surgical intervention. A further two cases of insufficient reduction (AADI > 3 mm) were registered. During the postoperative period one occipital decubitus related to the stiff collar immobilization and two cases of pneumonia were observed.

Group II

The first three patients had been treated with an isolated anterior plate fixation according to Harms. Surgical procedure was then changed and a posterior wire fusion according to Brooks was routinely added, because screw loosening in two out of these three initial patients had been observed. Additionally, one occipital decubitus related to stiff collar immobilization, one mediastinitis without the need for further surgical intervention, and two cases of pneumonia had been observed in the postoperative period.

Group III

Two cases of pneumonia and one occipital decubitus were registered. There were three patients with incomplete reductions (AADI > 3 mm).

Patients

Thirty-six consecutive patients with cervical spine instability were examined retrospectively at the Department for Arthritis Surgery at the J.W. Goethe University, Frankfurt/Main, Germany. In all patients rheumatoid arthritis (RA) had been diagnosed according to the revised criteria of the American College of Rheumatology.

Group I

Fourteen patients (9 female, 5 male) had isolated anterior reducible AAI. The average length of clinical and radiographic follow-up was 49.5 ± 20.8 months (range 21–86 months). Eleven patients were seropositive, three patients were seronegative. The average age at the time of surgery was 62.9 ± 6.6 years (range 53–75 years). The average duration of RA was 17.3 ± 2.6 years (range 14–23 years). All patients, except one, had a synovial pannus around the odontoid process that bulged into the spinal canal, compressing the upper cervical cord of six patients.

Group II

Fifteen patients (12 female, 3 male) had irreducible AAK. The average length of clinical and radiographic follow-up was 50.7 ± 15.6 months (range 26–77 months). Thirteen patients were seropositive and two patients were seronegative. The average age was 61.3 ± 14.9 years (range 31–82 years), with an average duration of RA that amounted to 25.4 ± 8.4 years (range 11–37 years). Eight patients had superior migration of the odontoid (SMO), while three patients had a non-traumatic fracture of the odontoid (NFO) (Fig. 2). All patients had a synovial pannus around the odontoid process that bulged into the spinal canal compressing the upper cervical cord of ten patients.

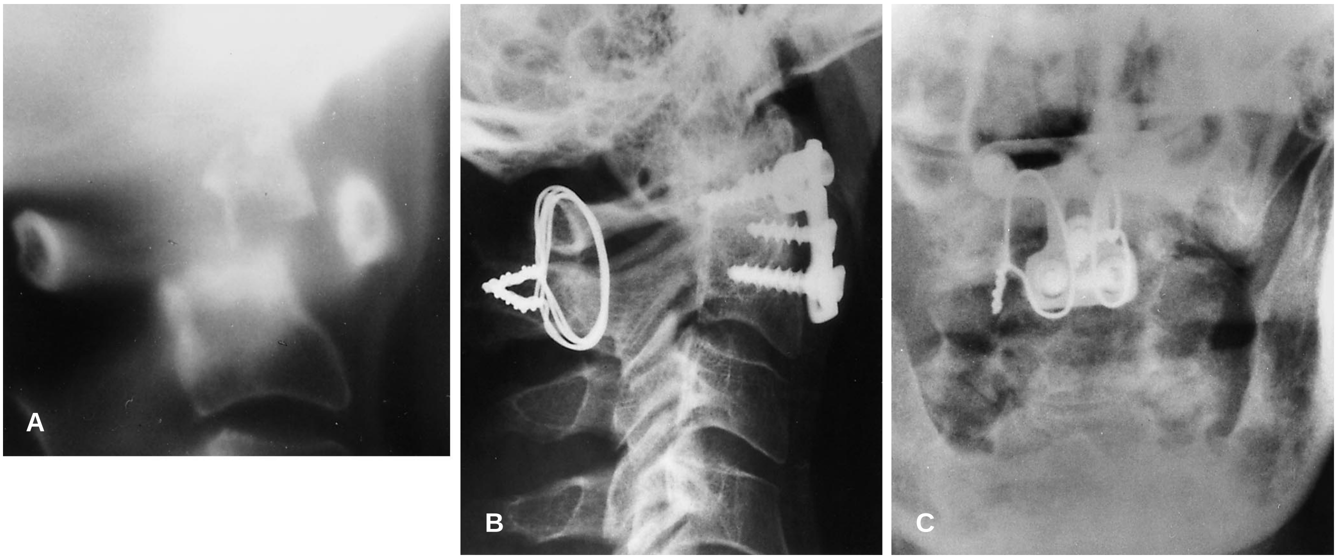

Fig. 2 A–C Patient 13, group II: a 61-year-old woman with a 31-year history of rheumatoid arthritis. **A** Preoperative lateral radiograph showing AAK with non-traumatic fracture of the odontoid. Neurological deficit was Ranawat class II and pain was 9.1 points on the visual analog scale. **B** Postoperative lateral radiograph 30 months after surgery showing accurate reduction and realignment. Neurological deficit was Ranawat class I and pain was 4.8 points on the visual analog scale. **C** Postoperative anterior-posterior radiograph 30 months after surgery

Group III

Seven patients (all female) had CS, accompanied by SAS in four cases. The average length of clinical and radiographic follow-up was 57.8 ± 24.8 months (range 26–96 months). All patients were seropositive. The average age was 66.2 ± 8.1 years (range 52–75 years) and the average duration of RA was 24.4 ± 6.6 years (range 18–36 years). All patients had SMO. Four patients had a synovial pannus around the odontoid process, but none was compressing the upper cervical cord.

Study protocol

Preoperatively and at the follow-up each patient had a careful and detailed rheumatologic, neurologic and orthopedic examination. Evaluation included the following parameters: pain (visual analog scale: 0 points means no pain; 10 points means severe pain) range of motion (ROM), subjective improvement, and Health Assessment Questionnaire (HAQ) [20]. The HAQ assigns a minimum of 0 points to persons performing daily activities at ease and a maximum of 3 points to persons who are completely unable to perform such activities. Range of motion for rotation and bending was measured clinically in degrees. Flexion/extension values were given as the difference between the maximum chin-jugulum distance in flexion and extension. A neurologic deficit was defined according to the classification proposed by Ranawat [51]. Plain radiographs of the cervical spine, including lateral flexion and extension views, were obtained on all patients. The anterior (AADI) and posterior atlanto-dental intervals (PADI) were measured. In patients who had substantial erosion of the odontoid process, measurement was made from the base of the remaining portion of the second cervical vertebra. An AADI of more than 3 mm was defined as abnormal [18, 19, 42]. Superior migration of the odontoid (SMO) was assessed using Redlund-Johnell [52] measurements and McGregor's line [6]. A projection of the odontoid of more than 4.5 mm above this line (dens-McGregor-interval, DMI) was considered as SMO [6, 51, 52]. To determine restoration of the height of the atlanto-axial joint, Redlund-Johnell [52] measurements were used. Before surgery patients were evaluated by MRI. The spinal cord diameter (SCD) and the synovial pannus around the odontoid process were assessed.

Statistical analysis

Intra-individual comparison of pre- and postoperative values was performed using Fishers least-square difference. Statistical significance was defined at a 95% confidence level ($P < 0.05$). The values were given as mean ± standard deviation. SPSS (release 7.0, SPSS, Chicago, Ill.) software supported statistical evaluation.

Results

Group I

At follow-up two patients (patients 2 and 11) were deceased 3 and 4.2 years after surgery due to causes unrelated to surgery. One patient (patient 7) was lost to follow-up (Table 1). Of the remaining 11 patients, HAQ increased in 5 patients, remained unchanged in 2 patients and decreased in 4 patients. The average HAQ increased from 1.47 ± 0.66 (range 0.375–2.75) points preoperatively to 1.59 ± 0.68 (range 0.75–3) points at follow-up (P = 0.78).

Values for the parameters pain, neurologic involvement, and ROM are shown in Table 1. Pain decreased significantly ($P < 0.001$) from an average of 6.9 ± 1.92 (range 3.6–9.2) points preoperatively to an average of 3.2 ± 1.24 (range 1.2–5.1) points, postoperatively. Preoperatively, one patient (patient 8) had objective signs of

Table 1 Results of the posterior wire fusion according to Gallie [21] (patients 1 and 2) and Brooks [5] (patients 3–14): values for Health Assessment Questionnaire (HAQ), pain, neurologic involvement, and range of motion (*RA* rheumatoid arthritis, + deceased, ++ lost to follow-up)

Patient	Age (years)	Sex	Follow-up (months)	RA (years)	HAQ pre (points)	HAQ post (points)	Pain pre (points)	Pain post (points)	Ranawat pre	Ranawat post	Chin-jugulum distance pre (cm)	Chin-jugulum distance post (cm)	Total right/left rotation pre (degrees)	Total right/left rotation post (degrees)	Total right/left bending pre (degrees)	Total right/left bending post (degrees)
1	60	f	86	15	1.5	1.825	9.2	3.9	II	II	6.1	8.8	72	84	32	38
2	72	m	+	16	2.75	+	4.3	+	II	+	10.3	+	76	+	44	+
3	68	f	76	17	1.25	1.25	8.8	3.3	I	I	11.2	13.7	82	88	40	62
4	56	f	64	19	0.75	1.25	6.9	2.8	I	I	9.8	14.7	88	92	46	58
5	56	m	58	20	1	0.75	7.1	2.1	I	I	6.3	9.1	86	86	44	48
6	57	m	58	20	2	1.5	9.0	5.1	I	I	8.4	13.6	64	76	50	54
7	68	f	++	16	0.375	++	3.7	++	II	++	6.5	++	72	++	46	++
8	68	f	47	20	0.75	1.75	3.6	1.8	III A	II	9.5	13.9	64	72	42	52
9	66	f	44	17	1	0.75	7.7	1.2	I	I	10.9	14.9	84	102	52	60
10	62	f	32	14	2.25	3	6.9	4.6	I	I	12.9	15.6	70	88	52	58
11	75	m	+	14	1.75	+	8.0	+	I	+	9.9	+	68	+	40	+
12	58	f	31	16	1.5	1.25	5.5	4.5	II	II	13.6	13.8	80	96	56	60
13	62	m	28	23	2	2.5	7.1	2.6	I	I	12.1	15.9	70	96	58	60
14	53	f	21	16	1.75	1.75	8.6	2.9	I	I	12.2	13.8	76	80	38	48

Table 2 Results of posterior wire fusion according to Gallie (patients 1 and 2) and Brooks (patients 3–14): radiographic and MRI parameters (*AADI* anterior atlanto-dental interval, *PADI* posterior atlanto-dental interval, *SCD* spinal cord diameter in maximum flexion on MRI)

Patient	AADI pre (mm)	AADI post (mm)	PADI pre (mm)	PADI post (mm)	SCD pre (mm)
1	5	6	19	18	8
2[a]	9	6	14	17	6
3	8	3	16	21	8
4	7	2	18	23	7
5	8	3	18	23	8
6	7	1	17	23	7
7[a]	6	1	16	21	6
8	9	3	13	19	9
9	6	2	15	19	8
10	8	1	17	24	7
11[a]	6	3	16	19	8
12	8	3	18	23	7
13	8	2	16	22	8
14	8	1	18	25	8

[a] Data from immediate postoperative radiographs

neurologic compromise (Ranawat class III A). Postoperatively, this patient improved one Ranawat class, the other patients remained unchanged. Postoperative chin-jugulum distance increased significantly ($P < 0.05$), by 3.2 ± 1.48 (range 0.2–5.2) cm. The average improvement for rotation was 11.3° ± 7.76° (range 0°–26°) ($P < 0.05$) and for lateral bending 7.8° ± 5.62° (range 2°–22°) ($P < 0.05$). All patients, except patient 6, were satisfied with the procedure and reported a subjective improvement.

Table 2 shows the radiographic results of group I. AADI decreased from an average of 7.3 ± 1.21 (range 5–9) mm, preoperatively, to an average of 2.6 ± 1.64 (range 1–6) mm, postoperatively ($P < 0.001$). PADI increased from an average of 16.5 ± 1.69 (range 13–19) mm, preoperatively, to an average of 21.2 ± 2.45 (range 17–25) mm, postoperatively ($P < 0.001$). With the Gallie method, one painfree pseudarthrosis occurred (patient 1), which did not require further surgery, and two insufficient reductions (patients 1 and 2). The mean SCD was 7.5 ± 0.85 (range 6–9) mm preoperatively. Redlund-Johnell (R-J) values remained unchanged. A non-union with the Brooks method was not observed.

Group II

At follow-up three patients (patients 3, 7, and 11) were deceased. These patients died at least 1 year after surgery (range 1.5–3.2 years) due to causes unrelated to surgery (Table 3). Of the remaining 12 patients, the HAQ increased in 3 patients, remained unchanged in 3, and de-

Table 3 Results of isolated anterior plating (patients 1–3) and the combined Harms-Brooks procedure (patients 4–15): values for HAQ, pain, neurologic involvement, and range of motion (+ deceased)

Patient	Age (years)	Sex	Follow-up (months)	RA (years)	HAQ pre (points)	HAQ post (points)	Pain pre (points)	Pain post (points)	Ranawat pre	Ranawat post	Chin-jugulum distance pre (cm)	Chin-jugulum distance post (cm)	Total right/left rotation pre (degrees)	Total right/left rotation post (degrees)	Total right/left bending pre (degrees)	Total right/left bending post (degrees)
1	55	f	77	35	2.75	3	4.4	3.2	II	I	8.1	9.8	55	94	22	38
2	82	f	74	17	3	2.25	9.8	6.1	III B	I	5.3	14.2	82	90	44	60
3	71	f	+	36	2.75	+	8.9	+	III A	+	6.2	+	68	+	34	+
4	70	f	64	16	3	3	8.8	4.2	III B	III A	7.1	14.0	40	82	40	56
5	48	f	57	24	3	2.875	9.7	3.9	III B	II	5.9	11.1	74	80	46	56
6	70	m	54	31	2.625	2.875	4.2	2.1	II	I	8.2	16.6	48	78	31	58
7	74	f	+	28	2.75	+	7.8	+	II	+	5.9	+	62	+	32	+
8	31	m	51	11	1.75	1.75	8.5	3.1	III A	I	11.9	14.3	74	78	40	52
9	62	f	48	21	2.875	2.25	9.5	4.6	III A	II	4.3	10.9	83	93	48	60
10	64	f	47	37	2.875	2.875	9.3	5.6	II	II	5.9	16.1	54	80	30	58
11	67	f	+	19	2.75	+	7.9	+	III A	+	6.6	+	54	+	36	+
12	34	m	41	18	2.75	2.25	9.1	2.6	II	I	8.1	9.8	62	94	24	44
13	61	f	36	30	1.75	1.25	9.1	4.8	II	I	9.9	14.9	60	84	46	64
14	55	f	34	35	2.75	2.25	5.3	4.1	II	I	7.2	9.6	56	65	26	41
15	79	f	26	23	1.125	1.5	7.6	2.3	II	I	7.8	16.0	64	82	38	54

Table 4 Results of isolated anterior plating (patients 1–3) and the combined Harms-Brooks procedure (patients 4–15): radiographic and MRI parameters (*DMI* dens-McGregor interval, *R-J* Redlund-Johnell measurement)

Patient	AADI pre (mm)	AADI post (mm)	PADI pre (mm)	PADI post (mm)	SCD pre (mm)	DMI pre (mm)	R-J pre (mm)	R-J post (mm)
1	11	1	14	24	8	3	30	30
2	9	1	13	21	5	8	24	27
3[a]	6	2	14	18	8	10	32	31
4	10	3	10	17	8	11	23	26
5	11	1	10	20	8	3	24	26
6	7	2	15	20	5	5	33	33
7[a]	7	2	15	20	6	7	27	29
8	9	2	12	19	6	7	32	33
9	9	1	13	21	6	7	26	26
10	8	1	17	24	6	2	33	32
11[a]	6	2	15	19	7	5	28	30
12	7	2	16	19	5	1	37	36
13	10	3	13	20	9	3	34	32
14	8	2	15	21	6	2	34	33
15	8	3	17	22	8	2	31	31

[a] Data from immediate postoperative radiographs

creased in 6 patients. The average HAQ was 2.51 ± 0.62 (range 1.125–3) points preoperatively and 2.34 ± 0.60 (range 1.25–3) points at follow-up ($P = 0.92$).

Values for the parameters pain, neurologic involvement, and ROM are shown in Table 3. Pain decreased significantly ($P < 0.001$) from an average of 7.9 ± 1.87 (range 4.2–9.8) points preoperatively to an average of 3.8 ± 1.27 (range 2.1–6.1) points postoperatively. All patients improved at least one Ranawat class after surgery, except a Ranawat class II patient (patient 10), who remained unchanged. The ROM increased significantly in all patients. Postoperative chin-jugulum distance increased by 3.8 ± 1.61 (range 1.7–6.6) cm ($P < 0.05$). The average improvement for rotation was 21.5° ± 14.0° (range 4°–42°) ($P < 0.05$) and for lateral bending 17.2° ± 5.54° (range 10°–28°) ($P < 0.05$). All patients were satisfied with the procedure and reported a subjective improvement.

Table 4 shows the radiographic results of group II. AADI decreased from an average of 8.4 ± 1.63 (range 6–11) mm, preoperatively, to an average of 1.9 ± 0.74 (range 1–3) mm, postoperatively ($P < 0.001$). PADI increased from an average of 13.9 ± 2.15 (range 10–17) mm, preoperatively, to an average of 20.3 ± 1.95 (range 17–24) mm, postoperatively ($P < 0.01$). The mean SCD was 6.7 ± 1.33 (range 5–9) mm, preoperatively. The eight patients (patients 2, 4–9, and 11) with SMO had a mean DMI of 7.5 ± 2.13 (range 5–11) mm and an average Redlund-Johnell (R-J) value of 27.1 ± 3.72 (range 24–33) mm. Due to reduction and bone grafting, height was restored in these patients to an average R-J value of 28.7 ±

Table 5 Results of occipito-cervical fusion: values for HAQ, pain, neurologic involvement, and range of motion (+ deceased)

Patient	Age (years)	Sex	Follow-up (months)	RA (years)	HAQ pre (points)	HAQ post (points)	Pain pre (points)	Pain post (points)	Ranawat pre	Ranawat post	Chin-jugulum distance pre (cm)	Chin-jugulum distance post (cm)	Total right/left rotation pre (degrees)	Total right/left rotation post (degrees)	Total right/left bending pre (degrees)	Total right/left bending post (degrees)
1	52	f	96	22	1.75	1.875	8.7	4.2	III A	III A	6.2	7.3	44	50	44	52
2	63	f	71	31	3	3	9.3	3.9	III B	III A	2.8	2.7	24	10	22	8
3	75	f	62	21	2.25	2.75	7.9	6.1	III B	III A	5.3	0	42	0	32	0
4	73	f	60	18	2.75	2.75	10.0	6.9	III B	II	2.8	3.7	18	38	26	32
5	61	f	32	36	1.5	1.75	8.8	3.7	III A	II	9.6	8.5	60	42	54	40
6	69	f	+	19	2.5	+	7.4	+	II	+	6.4	+	64	+	38	+
7	71	f	26	24	3	3	8.1	3.2	II	I	5.1	3.2	52	50	40	32

3.0 (range 26–33) mm, postoperatively ($P = 0.17$). A non-union was not observed.

Group III

At follow-up one patient (patient 6) was deceased. The patient died 1.6 years after surgery due to cardiac arrest (Table 5). Of the remaining six patients, the HAQ increased in three patients and remained unchanged in three. By HAQ patients were graded with an average of 2.39 ± 0.59 (range 1.5–3) points, preoperatively, and 2.52 ± 0.56 (range 1.75–3) points at follow-up ($P = 0.52$).

Values for the parameters pain, neurologic involvement, and ROM are depicted in Table 5. Pain decreased significantly ($P < 0.001$), from an average of 8.6 ± 0.88 (range 7.4–10.0) points, preoperatively, to an average of 4.7 ± 1.47 (range 3.2–6.9) points, postoperatively. All patients improved at least one Ranawat class after surgery, except a Ranawat class III A patient (patient 1), who remained unchanged. The ROM increased in two patients (patients 1 and 4) and decreased in the other patients. Postoperative chin-jugulum distance decreased by 1.1 ± 2.37 (range - 5.3 to 1.1) cm ($P = 0.12$). The average decrease for rotation was 10.7° ± 18.09° (range –32° to 8°) ($P < 0.05$), and for lateral bending 6.7° ± 18.53° (range –10° to 28°) ($P < 0.05$). All patients, except patient 5, were satisfied with the procedure. All patients reported a subjective improvement.

Table 6 shows the radiographic results of group III. AADI decreased from an average of 7.0 ± 1.15 (range 5–8) mm, preoperatively, to an average of 3.3 ± 1.49 (range 2–6) mm, postoperatively ($P < 0.01$). PADI increased from an average of 15.7 ± 2.62 (range 12–19) mm, preoperatively, to an average of 19.7 ± 3.49 (range 15–24) mm, postoperatively ($P < 0.05$). The mean SCD was 6.6 ± 1.13 (range 5–8) mm, preoperatively. Preoperatively, the mean DMI was 7.0 ± 0.81 (range 6–8) mm and the average Redlund-Johnell (R-J) value was 25.1 ± 1.67 (range 23–27) mm. Postoperatively, the average R-J value was unchanged, at 25.1 ± 2.03 (range 22–37) mm. A non-union was not observed.

Discussion

The progressive destruction of the occipito-atlanto-axial joints in RA requires stage-related surgery. The basis for the presented therapeutic concept of stage-related surgery was the status of the anterior column. It was the objective of this study to analyze in detail the clinical results with this concept.

Since 1909, when Mixter and Osgood [47] described the posterior approach for atlanto-axial joint stabilisation, various investigators have advocated different techniques to achieve C1/C2 spondylodesis [5, 18, 19, 21, 37, 46, 55,

Table 6 Results of occipito-cervical fusion: radiographic and MRI parameters

Patient	Fusion level	AADI pre (mm)	AADI post (mm)	PADI pre (mm)	PADI post (mm)	SCD pre (mm)	DMI pre (mm)	R-J pre (mm)	R-J post (mm)
1	C0–C2	5	4	18	19	6	5	27	27
2	C0–C5	7	2	15	20	7	5	26	27
3	C0–T1	8	3	19	24	6	7	25	25
4	C0–C4	8	2	14	20	7	6	27	27
5	C0–C2	8	6	14	16	7	8	23	23
6[a]	C0–C2	7	4	12	15	8	8	23	22
7	C0–C4	6	2	18	24	8	5	25	25

[a] Data from immediate postoperative radiographs

56, 58]. Although Gallie [21] never published a detailed technique of surgical correction of C1/C2 instability, one of the standard operations bears his name. His technique, which uses a modified H-shaped bone graft secured by a midline wiring, is described by Fielding et al. [19]. In 1987, Brooks and Jenkins [5] described atlanto-axial arthrodesis by the wedge compression method. In early stages of cervical spine instability due to RA, especially when the anterior column is intact, these methods have proven to be safe and effective. For both fixation techniques, results of several clinical studies with an average success rate of 90% are available [5, 18, 19, 21, 46, 56]. In the present study, patients of group I showed continued neurologic improvement, significant pain reduction, and an increased range of cervical motion. ROM increase in this group might mainly be due to a pain decrease. The radiographic follow-up showed a solid bony fusion in all patients, except in one patient with a Gallie fixation, without subsequent subaxial subluxation.

With increasing pathology of the underlying disease process alternative procedures are required. In particular, when chronic irreducible atlanto-axial kyphosis (AAK), superior migration of the odontoid, irreducible dislocation of the atlas, posterior atlanto-axial subluxation, or atraumatic fracture of the odontoid in combination with anterior compression of the cervical cord becomes manifest, anterior decompression and odontoidectomy is necessary. AAK corresponding to a destruction of the anterior column at C2 with cord compression is especially difficult to treat, because the cord is compressed both posteriorly by the posterior arch of the atlas and anteriorly by the odontoid or the retrodental pannus.

Historically, surgical treatment of irreducible ventral pathology at the craniovertebral junction was performed via a posterior approach only. In general, however, posterior surgical procedures for irreducible AAK with spinal cord compression were associated with high morbidity and mortality and often achieved incomplete reduction, only [1–3, 13, 24–26, 33, 40, 57]. Suboccipital decompression with atlanto-axial fusion can rarely satisfactorily decompress and particularly realign irreducible AAK [9, 27]. In addition, irreducible AAK increases the operative risk of the posterior approach because of the need to flex the head intraoperatively to achieve adequate exposure of C1/C2, thereby risking fatal anterior injury to the spinal cord by the odontoid or the pannus [25]. Moreover, the posterior approach may even be contra-indicated in cases where the dislocated posterior arch severely compresses the spinal cord [40, 48].

In contrast, the anterior transoral approach allows release of the anterior compression of the spinal cord, the direct loosening of the C1/C2 joints, and restoration of the anterior column height by bone grafting. Transoral odontoid resection is an effective and safe surgical procedure. Several authors have described good results obtained with this procedure [7, 8, 16, 17, 30, 39, 44, 45, 60–62], but very few studies have been published about the transoral approach including odontoid resection for irreducible AAK [22, 24, 35, 53, 59].

Apart from the value of this procedure with regard to decompression, postoperative instability after odontoid resection is an important issue. Biomechanically, the transoral odontoidectomy produces a significant antero-posterior translation, increased ranges of motion, and mobile unconstrained instantaneous axes of rotation of the atlanto-axial joint [11, 12]. Clinically, Menezes and Van Gilder [44] found that 72% (52 of 72) of their patients required additional posterior stabilization procedures after odontoidectomy. Di Lorenzo [13] also reported that 88% (22 of 25) of his patients required additional stabilization. Therefore, several authors have postulated internal fixation after transoral odontoidectomy [7, 12, 13, 44, 45].

The optimum fixation method to achieve fusion at the C1/C2 level after odontoid resection is still a matter of discussion. Several fixation methods have been described in the literature.

Atlanto-axial wire fusion after odontoid resection has proved to be ineffective for adequate stabilization [10]. If posterior wiring is used, a tendency for anterior instability persists, because the circular wires are not efficient in preventing translatory displacement. This phenomenon has been termed the “parallelogram effect” by Panjabi.

Fang and Ong [17] and Subin et al. [59] described a different therapeutic pathway. They performed transoral odontoid resection without secondary posterior fixation. This procedure requires a long-term external fixation in a cast or a halo jacket for at least 4 months and implies a high non-union rate (3 out of 10 patients [59]).

Although we are not aware of any study dealing with this subject, another alternative after odontoid resection might be atlanto-axial transarticular screw fixation according to Magerl [41]. But, this procedure would make it necessary to turn the patient around after the transoral procedure during the period of least stability, thereby risking fatal injury to the spinal cord.

The most common alternative procedure to achieve C1/C2 fusion after transoral odontoid resection is posterior occipito-cervical (C0-C2) fusion [7, 8, 23, 29, 38, 49, 63]. Yet, occipito-atlanto-axial fusion has several disadvantages in comparison to isolated atlanto-axial fusion. The major disadvantages are an 18–33% failure rate [23, 38, 54, 56], prolonged bed confinement with traction [23, 29, 49], a ROM decrease, and chronic overload of the subaxial segments [34, 43, 50, 64]. In this study, although we performed occipito-cervical fusion only in cases of occipito-atlantal joint involvement, we saw similar problems. In group III only in four out of seven patients was a sufficient reduction and realignment achieved. The ROM after occipito-cervical fusion was significantly decreased and the head immobilization lasted longer than in the other groups. Therefore we tried to avoid occipito-cervical fusion if at all possible.

If transoral odontoidectomy is necessary, then transoral plate fixation according to Harms [30] combined with posterior wire fixation according to Brooks [5] might represent a surgical technique avoiding the cited risks and providing adequate immediate stability. Experimental studies have shown high immediate postoperative stability values [31, 32], which allow a consequent postoperative mobilization of the patients in a stiff collar. Stresses to the subaxial spine are reduced, because the fusion is limited to the atlanto-axial joint and therefore nodding movements are not limited. An accurate reduction and restoration of normal C1/C2 height can be obtained by the option of unlocking C1/C2 joints and inserting bilateral anterior bone grafts.

Our clinical results of this combined Harms-Brooks procedure (group II) for irreducible atlanto-axial kyphosis in patients with rheumatoid arthritis are encouraging. The patients who had an anterior transoral decompression and stabilization have shown continued neurologic improvement, significant pain reduction, and an increased range of cervical motion. According to our observations, the ROM increase in these patients might mainly be due to a pain decrease. The radiographic follow-up showed a solid bony fusion in all patients without subaxial subluxation. The Redlund-Johnell measurements documented restoration of the height of the atlanto-axial joint. The anatomical restoration of the radiographic atlanto-axial joint parameters (see AADI, PADI, Table 4) mainly contributed to the good neurologic outcome of these patients. There were no perioperative deaths. This may not only reflect the success of anterior decompression, but also the effect of early mobilization, which reduces morbidity and mortality in this susceptible group.

Bearing in mind that posterior wiring gives excellent clinical results in early stages of atlanto-axial instability, this combined procedure is not advocated as a routine replacement of standard techniques. However, this combined procedure is strongly recommended in cases of irreducible atlanto-axial instability with anterior cervical cord compression.

Conclusion

A stage-related surgery in cervical spine instability due to RA appears to be necessary. Posterior wiring yields excellent clinical results only in early stages of atlanto-axial instability. If the anterior column is destroyed including a manifest cervical myelopathy where a contribution of anterior structures to the cervical-medullary compression has been proven, transoral surgery is recommended. Transoral plate fixation combined with posterior wire fixation after transoral odontoid resection is an apparently effective, reliable, and safe procedure for treatment of irreducible AAK in RA. Occipito-cervical fusion should be performed only if the occipito-atlantal joints are destroyed, since major functional deficits are to be expected.

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