An Observational Study of Patient-rated Outcome After Atlantoaxial Fusion in Patients With Rheumatoid Arthritis and Osteoarthritis

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Abstract

Background Fusion is used to address several types of abnormality of the atlantoaxial segment. Traditionally, outcome has been assessed by achieving solid bony union. Recently, however, patient-rated outcome instruments have been increasingly used, although these may be influenced by concomitant comorbidity.

Questions/purposes We therefore asked whether patients with rheumatoid arthritis (RA), with its associated comorbidity, had worse self-rated outcomes after C1-2 fusion than patients with osteoarthritis (OA).

Patients and Methods We retrospectively reviewed all 30 (23 OA and seven RA) prospectively followed patients in our local Spine Registry (part of the Spine Society of Europe Spine Tango Registry) who had undergone C1-2 fusion. Before surgery and 3 and 12 months later, patients completed the multidimensional Core Outcome Measures Index (COMI) questionnaire. Global outcome and satisfaction with treatment were also assessed.

Results We found no group differences for duration of operation, blood loss, or perioperative surgical or general complications. Compared with the OA group, the RA group showed a better baseline COMI score and less

improvement in the COMI from preoperatively to 12 months followup. However, the proportion of “good” global scores at 12 months followup was similarly high in both groups (87% OA and 86% RA) as was satisfaction (96% for OA versus 86% for RA).

Conclusions Symptoms and impairment were less severe in the RA group at baseline and showed less improvement after surgery, but the proportion of “good global outcomes” was similar in both groups, and the great majority of patients in both groups were satisfied with their treatment.

Level of Evidence Level III, therapeutic study. See Guidelines for Authors for a complete description of levels of evidence.

Introduction

The wide range of motion and the specific anatomic configuration of the occipitocervical junction, in particular the atlantoaxial joint, render it susceptible to the development of pathologic structural changes. Consequently, osteoarthritis (OA) of the C1-2 joint may develop in elderly people. In patients with rheumatoid arthritis (RA) or other rheumatic conditions, disease-related insufficiency of the transverse ligament and bony erosion can lead to instability of the C1-2 joint. In each of these scenarios, the therapeutic strategy from a surgical point of view, regardless of the specific etiology or pathology, comprises immobilization and stabilization.

Various techniques of fixation have been advocated over the years. Instrumentation is commonly used to improve stability and enhance bony union of the bone graft. In the late 1950s, wiring was the most popular fixation technique; however, the fusion rates were only moderate and

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pseudarthrosis rates of 10% to 15% were not uncommon [6], especially when transverse instability was present, as commonly seen in RA. In addition, a reliable reduction of the segmental subluxation was seldom achieved [10]. These deficiencies were improved by the introduction of transarticular screw fixation, which was first described by Magerl in the 1980s [14]. With this technique, the fusion rates reached as high as 100% [19]. This, and the improved anatomic reduction, led to generally more favorable outcomes and widened the indication for atlantoaxial fusion. Further development of instrumentation, to include rod/screw systems, allowed additional use of this type of surgery in specific conditions such as fixed deformities or anatomic anomalies [9, 12]. The latter method involved the introduction of screws bilaterally in the lateral masses of the atlas and the isthmus of C2 allowing more mechanical impact for reduction. Both techniques (transarticular screws and C1 lateral mass screws-C2 pedicle screws) carry a certain risk of iatrogenic injury to the vertebral artery in the area of the C2 isthmus. More recently, the method of translaminar fixation, with screws placed in the lamina of C2, was introduced to obviate this potential risk and now offers an additional method of atlantoaxial fixation [22].

Whatever the technique used, we consider radiographically confirmed solid fusion the intended goal of the surgery. Accordingly, the presence of bony union is generally considered to be the objective sign of a favorable outcome. Recently, however, such outcomes have been superseded by subjective patient-rated outcomes, which are considered to be more relevant to the patient and society alike [5]. Comorbidities, among other factors, can influence patient-rated outcome [15], and it might therefore be expected that self-rated outcomes after surgery would be less good in patients with RA, with their typically greater comorbidity [8], than in those with OA.

We therefore asked whether the patient-rated Core Outcome Measures Index (1) at baseline and (2) at 12 months followup would be worse in patients with RA than in patients with OA undergoing the same posterior transarticular fusion surgery.

Patients and Methods

We retrospectively reviewed all 30 patients who had undergone C1-2 fusion with at least 12 months followup recorded in our local Spine Surgical Registry. This registry was first initiated in 2005 in connection with the Spine Society of Europe [SSE] Spine Tango Registry and includes all patients operated on within our Spine Center [18]. Two groups of patients were involved. In the first group (RA), the indication was atlantoaxial instability of

more than 8 mm or progressive instability with or without pain but reducible in extension views, resulting from rheumatoid changes of the atlantoaxial segment; there were no neurologic deficits as indications. In the second group (OA), the indication was radiographic findings of unilateral degenerative changes of the atlantoaxial segment that were concordant with the clinical symptoms of unilateral high cervical intense pain with restriction of motion and pain provocation in head rotation and confirmed pain relief on intraarticular injection.

We excluded patients from the study if they had undergone fusion with inclusion of the occiput and/or the lower cervical spine and those with irreducible C1-2 instability. There were 23 patients in the OA group (19 female, four male; 70.8 ± 10.2 years) and seven patients in the RA group (five female, two male; mean age 55.0 ± 14.1 years). The patients with OA were older ($p = 0.03$) than the patients with RA, but there was no difference between the two groups in gender distribution. There was a nonsignificant tendency for patients to have greater comorbidity in the RA group than in the OA group (71% versus 52%, respectively, with American Society of Anesthesiologists Score III). Two patients (one OA and one RA) did not return a followup questionnaire at 12 months; each was contacted and had promised to return it but had not done so by the time of the data analysis. Each has since returned a questionnaire for their subsequent followup at 2 years postoperative (RA patient, operation helped a lot and was very satisfied; OA patient, operation helped but was not satisfied with the treatment).

All patients underwent the same surgical procedure with transarticular atlantoaxial screw fixation according to Magerl [14]. Through a posterior approach, two screws were inserted bilaterally in the sagittal plane, each crossing the C1-2 facet. To enhance bony fusion, a Gallie-type bone graft was fixed between the posterior arch of the atlas and the spinous process of C2. In all patients, the bone graft was harvested from the left posterior iliac crest using the same operative technique. The duration of the operation was comparable in the two groups as was the extent of blood loss (Table 1).

The postoperative regimen was identical for both groups: mobilization on the first postoperative day, no external fixation, and physiotherapy for muscle strengthening after 6 weeks.

Before and 3 and 12 months after surgery, patients were requested to complete the multidimensional Core Outcome Measures Index (COMI) questionnaire [7, 16, 17], the primary outcome variable. On each occasion, the questionnaires were sent to the patients to complete at home to ensure the information given was free of care-provider influence. The COMI is a multidimensional index consisting of validated questions covering five domains: pain

Table 1. Medical and surgical details for the two groups

Variable	Degeneration (N = 23)	Inflammation (N = 7)	p Value
Comorbidity, ASA score (%)			
I (no disturbance)	13.0%	0.0%	0.52
II (mild/moderate)	34.8%	28.6%	
III (severe)	52.2%	71.4%	
Operation duration (%)			
Less than 1 hour	34.8%	28.6%	0.93
1–2 hours	47.8%	57.1%	
2–3 hours	13.0%	14.3%	
3–4 hours	4.4%	0.0%	
Blood loss (%) [†]			
Less than 500 mL	87.0%	100.0%	0.99
500–1000 mL	13.0%	0.0%	
General complications (intra-/perioperative)	0%	0%	—
Surgical complications (intra-/perioperative)	0%	0%	—
Surgeon-rated complications at 6-week followup [‡]	5.6%	0%	0.99*
Patient-rated complications at 12-month followup	4.3%	14.3%	0.42*

* For complications, Fisher's exact test was used to determine p values; [†]data missing for one patient in degeneration, two patients in the RA group; [‡]data documented with the Spine Society of Europe Surgeon Followup Form available for just 18 of 23 patients in the OA group and 3 of 7 patients in the RA group; insufficient surgeon followups at any later time points; ASA = American Society of Anesthesiologists; OA = osteoarthritis; RA = rheumatoid arthritis.

(neck and arm pain intensity, each measured separately on a 0–10 graphic rating scale), function, symptom-specific well-being, general quality of life, and disability (social and work). The score includes a composite index using: the highest of the two pain scores, the average score for social and work disability, and the scores for function, symptom-specific well-being, and general quality of life. A high score reflects worse status and a low score, better status. The COMI was originally developed based on the recommendations for a short series of core outcome questions by an expert group in the field of spine outcome measurement [5] and subsequently validated as an outcome instrument by three research groups [7, 16, 17]. In addition to the COMI questions, at the 3- and 12-month followups there were further questions with 5-point Likert scales inquiring about satisfaction (“over the course of treatment for your neck problem, how satisfied were you with the medical care in our hospital?”; response categories from “very satisfied” to “very dissatisfied”) and the global outcome of surgery (“overall, how much did the operation help your neck problem?”; response categories from “helped a lot” to “made things worse”) [17]. For further analysis,

these measures were dichotomized into two groups for global outcome (“good outcome” [helped/helpful a lot] and “poor outcome” [helped only little/did not help/made things worse]) and satisfaction (“satisfied” [satisfied/very satisfied] and “not satisfied” [neither satisfied nor dissatisfied, dissatisfied, very dissatisfied]). The questionnaire also contained questions as to whether the patient had experienced any complications as a result of the surgery [11].

We used SSE Spine Tango Surgery forms to document information regarding the medical history (eg, main pathology with further indication of the specific type of abnormalities, most severely affected segment, extent of lesion, etc), operation duration (10 categories, from less than 1 hour to greater than 10 hours), blood loss (five categories: none, less than 500 mL, 500–1000 mL, 1000–2000 mL, greater than 2000 mL), comorbidity (assessed with the American Society of Anesthesiologists Physical Status Score from 1 [no disturbance] to 5 [moribund]), surgical details, surgical complications, and general complications.

We determined differences between the two groups (RA and OA) for continuous variables (baseline status for COMI item/composite scores and the change in COMI item/composite scores from preoperatively to 12-month followup) using Mann-Whitney U tests. As a result of the small group sizes, nonparametric analyses were used. Differences between RA and OA groups for nominal data (ie, surgical details [see previously] and the proportions of good outcomes, satisfied patients, and self-rated complications) were examined using chi square contingency analysis.

The data were analyzed using StatView 5.0 (SAS Institute Inc, San Francisco, CA).

Results

At baseline, preoperatively, the RA group had less severe ($p = 0.02$) neck pain than the OA group. The RA group tended to have better ($p = 0.10$) status in terms of function and greater ($p = 0.07$) symptom-specific well-being than the OA group (Fig. 1).

Improvements (ie, the reduction in scores from preoperatively to 12 months followup) were overall less marked in the RA group than the OA group (ie, lower [$p = 0.03$] change scores for neck pain, [$p = 0.04$] for symptom-specific well-being, and [$p = 0.02$] for social disability, and a tendency for lower [$p = 0.06$] change scores for function). The improvement in COMI score from preoperatively to followup showed a tendency to be less ($p = 0.06$) in the RA group than the OA group (Fig. 2); the difference between groups in their improvement at

Fig. 1 Individual Core Outcome Measures Index (COMI) domain scores and COMI composite score before surgery and at the 12-month followup in the two groups. 0 = best status; 10 = worst status for each domain. Blue = patients with osteoarthritis (OA); pink = patients with rheumatoid arthritis (RA). Solid lines with square markers are presurgery; dashed lines with circular markers are 12-month followup. Compared with the patients with OA, at baseline, the patients with RA had overall better status (ie, lower [$p = 0.02$]

scores for neck pain and a tendency for lower [$p = 0.10$] scores for function and [$p = 0.07$] for symptom specific well-being). Improvements (reductions in scores from preoperatively to 12 months followup) were overall less marked in the RA group than the OA group (ie, lower [$p = 0.03$] change scores for neck pain, [$p = 0.04$] for symptom-specific well-being, [$p = 0.02$] for social disability, [$p = 0.06$] for function, [$p = 0.06$] for COMI composite score).

Fig. 2 Change in mean Core Outcome Measures Index (COMI) composite score from before surgery to 3 months and 12 months after C1-2 fusion in the two diagnostic groups. The reduction in COMI score from preoperatively to 12 months later was less ($p = 0.06$) pronounced in the rheumatoid arthritis (RA) group than the osteoarthritis (OA) group, ie, the RA group showed less improvement in multidimensional outcome at followup. The mean difference between groups was 2.9 (90% C.I. 0.9–4.8).

12 months postoperatively was 2.9 (90% C.I. 0.9–4.8) points. The global outcome at both 3 months and 12 months followup was similarly high in both the OA and RA patient groups (87% and 86% good outcomes, respectively) (Table 2). Satisfaction with treatment at the

Table 2. Global outcome of surgery and satisfaction with treatment in the two groups

Followup	Osteoarthritis (N = 23)	Rheumatoid arthritis (N = 7)	Significance of group difference p value
3-month global outcome	87.0%	85.7%	0.99
3-month satisfaction	100.0%	85.7%	0.23
12-month global outcome	87.0%	85.7%	0.99
12-month satisfaction	95.7%	85.7%	0.42

3-month and 12-month followups was 96%–100% for the OA group and 86% for the RA group (Table 2).

No complications, of either a surgical or general nature, were reported in either of the two groups before discharge from the hospital (Table 1). In the 12-month questionnaire, the patients themselves reported complications in connection with their surgery at a similar rate ($p = 0.42$): 4.3% in the OA group and 14.3% in the RA group (Table 1).

Discussion

Positive subjective outcomes, with satisfied patients, should be the main goal of every surgical intervention. However, comorbidities can greatly influence patients' ratings of quality of life [2] and the effectiveness of treatment [15]. Because patients with RA typically have more comorbidities than those with other rheumatologic diseases [8], we questioned whether these patients might

also show worse self-rated status at baseline and poorer subjective outcomes after C1-2 fusion than patients with OA.

We bring several limitations to the reader's attention. First, owing to the small sample sizes, the power of the study was limited and caution must be exercised in interpreting the results given the chance that we may have failed to observe a difference when in truth there was one (ie, a Type II error). This most strongly influences interpretations of findings with borderline statistical significance, although in the present study, achieving significance for these would not have changed the overall conclusions. Our study is nonetheless best considered as an initial pilot study that will hopefully encourage larger, systematic followup studies; such studies should also allow more elaborate statistical analyses to take account of the possible group biases of age, comorbidity, and baseline symptoms. Second, we deliberately restricted our inclusion criteria to isolated atlantoaxial fusion to achieve homogeneity of the groups and an optimal basis for comparison, and the results are therefore applicable only to this select group. Third, factors such as race and culture as well as the focus of the outcome measure itself (pain, function, work loss, or quality of life) might influence the proportion of "good outcomes" reported [15]. Nonetheless, this is not a limitation unique to the present study; it applies to all outcome studies relying on self-ratings. Fourth, the short multidimensional COMI questionnaire was developed [7, 16, 17, 21] to obviate the need for many long questionnaires to address all the relevant outcome domains [5]. With its single question per key outcome domain, it represents a compromise between brevity and comprehensiveness; however, it does not provide an elaborate assessment of psychological status or quality of life. Nonetheless, its simplicity makes it ideal for completion at home, unaided, allowing for an optimum of independency of the outcome rating.

Patients with RA have various different pain sources, and one might therefore assume they would demonstrate a worse preoperative status regarding pain and function and quality of life. Our data, however, demonstrated the opposite, with patients with RA generally showing better status than patients with OA preoperatively. Painful OA of the atlantoaxial joint typically affects patients older than 60 years of age, and the pain is located at the occipito-cervical junction, often strictly unilaterally. Despite its relative frequency, the source of pain is often diagnosed with delay and after a considerable time of suffering, because the subaxial cervical spine is commonly suspected to be responsible for the symptoms. This may explain the rather high pain levels and associated impairment of function and quality of life (giving an overall higher COMI score) in the OA patient group. Patients generally demonstrate no additional pathologies other than age-related

degenerative changes and the surgery for OA usually closes the case, as corroborated in the present study. In contrast, the instability encountered in the patient with RA may or may not be painful. Neck pain is not normally their biggest problem and the cervical problem is just one of many others; the majority of patients have other osteoligamentous affections of the disease for several years and 50% of patients will undergo other orthopaedic interventions, mainly the replacement of large joints [13]. Accordingly, the regional pain may be lower and the tolerance for suffering and pain perhaps higher in patients with RA than in patients with degenerative pain sources [20]. These factors may all go toward explaining the lower preoperative COMI scores in the patients with RA.

We presumed the subjective outcome of surgery in the two groups would also differ as a result of their different medical history. With their multiple pathology, repeat surgery, etc, it is often supposed that patients with RA will report worse subjective treatment outcomes, although support for this in the literature appears to be lacking. We observed less marked improvement in the COMI score in the patients with RA and they also had lower postoperative quality of life. Therefore, although the resolution of the cervical problem contributed to a general improvement in scores, it was less dramatic than in the OA group, likely because of ongoing restrictions of the systemic disease and lesser pain and impairment to start with. In contrast, the patients with OA are more or less "back to normal" after fusion. Interestingly, the overall global outcome of the surgery ("how much did the operation help?") and satisfaction were similar for both groups, suggesting the patients in each diagnostic group felt the operation helped their own particular situation to a similar extent, even if the absolute change scores for the different domains differed between them and the residual deficits in function were more marked in RA. Comparison of our data with those in the literature is difficult, because few have concentrated on subjective outcomes and satisfaction. Using the data of 3617 total elbow arthroplasty cases from the Nationwide Inpatient Sample, Cook et al. [4] compared the hospital disposition factors and the rates of various perioperative complications in patients with RA and patients without RA and reported they were mostly quite comparable. Angst et al. [1] examined the subjective outcomes of total elbow arthroplasty in patients with RA and trauma patients, but the focus was on function compared with "normal" rather than satisfaction or the global outcome of surgery; postoperative function was lower in patients with RA than patients without RA, although pain and psychological well-being were comparable in the two groups. In patients undergoing TKA, satisfaction with the knee prosthesis, measured on a visual analog scale, was higher in patients with RA than in patients with OA, but there were no

differences in the physician's assessment of the overall clinical outcome as measured with the Knee Society Clinical Rating System score [3]. This finding of "greater satisfaction per unit change in absolute score for symptoms and function" for patients with RA concurs with that of the present study after C1-2 fusion.

In conclusion, the scores for symptoms and impairment were lower in the RA group than the OA group at baseline and showed less improvement change after surgery. However, the proportion of "good global outcomes" (ie, how much the operation helped the patient) was similar in both groups, and the majority of patients in both groups were satisfied with their treatment.

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