

Original Article

The surgical management of the rheumatoid spine: Has the evolution of surgical intervention changed outcomes?

Robin Bhatia^{1,2}, Nikolas Haliasos², Pierluigi Vergara², Caroline Anderson³, Adrian Casey²

¹Department of Orthopaedic Surgery, The Great Western Hospital, Swindon, ²Department of Neurosurgery, The National Hospital for Neurology and Neurosurgery, London, ³Norwich Medical School, Norwich, UK

Corresponding author: Mr. Robin Bhatia, Department of Orthopaedic Surgery, The Great Western Hospital, Swindon, UK. E-mail: robbhatia@yahoo.com

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Abstract

Context: Surgery for the rheumatoid cervical spine has been shown to stabilize the unstable spine; arrest/improve the progression of neurological deficit, help neck pain, and possibly decelerate the degenerative disease process. Operative intervention for the rheumatoid spine has significantly changed over the last 30 years. **Aims:** The purpose of this study was to review all cases of cervical rheumatoid spine requiring surgical intervention in a single unit over the last 30 years. **Materials and Methods:** A prospectively-maintained spine database was retrospectively searched for all cases of rheumatoid spine, leading to a review of indications, imaging, Ranawat and Myelopathy Disability Index measures, surgical morbidity, and survival curve analysis. **Results:** A total of 224 cases were identified between 1981 and 2011. Dividing the data into three time-epochs, there has been a significant increase in the ratio of segment-saving Goel-Harms C1-C2: Occipitocervical fixation (OCF) surgery and survival has increased between 1981 and 2011 from 30% to 51%. Patients undergoing C1-C2 fixation were comparatively less myelopathic and in a better Ranawat class preoperatively, but postoperative outcome measures were well-preserved with favorable mortality rates over mean 39.6 months of follow-up. However, 11% of cases required OCF at mean 28 months post-C1-C2 fixation, largely due to instrumentation failure (80%). **Conclusion:** We present the largest series of surgically managed rheumatoid spines, revealing comparative data on OCF and C1-C2 fixation. Although survival has improved over the last 30 years, there have been changes in medical, surgical and perioperative management over that period of time too confounding the interpretation; however, the analysis presented suggests that rheumatoid patients presenting early in the disease process may benefit from C1 to C2 fixation, albeit with a proportion requiring OCF at a later time.

Key words: Atlantoaxial subluxation, basilar invagination, C1-C2 fixation, occipitocervical fixation, rheumatoid cervical spine

INTRODUCTION

Rheumatoid arthritis commonly affects the cervical spine, and if left untreated, can result in progressive debilitating neurological morbidity and increased mortality.^[1-5] There are several, overlapping pathological processes occurring in the cervical spine of the symptomatic rheumatoid patient. These include: atlanto-axial (horizontal) subluxation, pannus-related stenosis, subaxial instability/stenosis, and vertical translocation otherwise referred to as basilar invagination.^[2,6] To what extent does one pathological process exacerbate or result in another?

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There is some evidence to suggest that untreated atlanto-axial subluxation can lead to vertical translocation and brainstem compression.^[2,7-9] The corollary of this is that early fixation of C1-C2 may reduce the incidence of late-onset vertical translocation, although definitive evidence for this is lacking at present. There is evidence that early C1-C2 fixation can improve pannus-related stenosis whilst sparing the atlanto-occipital segment;^[10-13] however, this may be offset by the potential for exacerbation of subaxial instability.^[14-16] Furthermore, there is the school of thought that C1-C2 fusion merely delays the inevitable and definitive operation of occipitocervical fixation (OCF). Two camps have thus arisen: The “segment spacers” and “occipitocervical fusionists”.

How has the evolution of instrumented craniocervical surgery reflected our understanding of the natural history/pathophysiology of rheumatoid disease? Broadly, we may define three interventions in the surgical management of rheumatoid spine disease at the craniocervical junction: C1-C2 fixation, which had its origins in the original Gallie^[17] and Brooks^[18] type fusion, superseded by transarticular C1-C2 fixation,^[19] and later the Goel-Harms C1-C2 fixation;^[20] OCF: Using onlay graft, sublaminar wire/loop fixation^[21] and rigid OCF;^[22,23] and the transoral anterior odontoid decompression popularized by Casey and Crockard.^[24,25]

In previous publications from this group, we have demonstrated the safety and efficacy of C1-C2 fixation^[26] and rigid OCF^[23] for the treatment of various rheumatoid and nonrheumatoid cervical pathologies. It is the purpose of this study to review, in totality, the entire surgical output for instrumented rheumatoid cervical spine management in a single unit over the last 30 years. There are three major questions we would wish to address:

1. How have trends in instrumentation changed over that time period?
2. How have changes in surgical intervention influenced functional improvements and mortality rates associated with rheumatoid arthritis of the cervical spine?
3. What proportion of patients undergoing (early) C1-C2 fixation require OCF at a later date?

MATERIALS AND METHODS

This was a retrospective review of three prospectively-maintained databases of posterior instrumented cervical spine surgery for patients with rheumatoid arthritis in a single unit, spanning from 1982 to 2012. The operative procedures comprised: Ransford loop/sublaminar wire OCF, rigid (lateral mass/pedicle screw and occipital plate) OCF, and C1-C2 fixation (transarticular screw fixation and Goel-Harms posterior fusion). We did not include noninstrumented, or Brookes-Gallie wire, craniocervical fusions since these procedures largely comprised the preceding era and data collection was not sufficiently reliable before 1982.

This study was carried out at a tertiary referral center for spinal surgery; the operating surgeons were all consultant/attending neurosurgeons, assisted by spinal fellows from both neurosurgical and orthopedic backgrounds.

Baseline patient demographics included age, sex, radiology, pre-operative Ranawat grade, and Myelopathy Disability Index (MDI).

Operative procedures were categorized as above. C1-C2 fixation was subcategorized into transarticular screw insertion as originally described by Magerl,^[19] or Goel posterior C1-C2 fusion.^[20] A review of the operative steps for these two procedures has been described in a previous publication by Vergara *et al.*^[26] Ransford loop/sublaminar wire OCF has been described by Moskovich *et al.*^[21] The operative technique for rigid OCF has been described by Bhatia *et al.*^[23]

It was not possible to accurately define when patients developed rheumatoid arthritis, and insufficient data were retrievable to determine when in the disease process cervical symptoms developed; however, baseline preoperative MDI allowed comparisons to be made between operative cohorts.

Patients with rheumatoid arthritis were administered methotrexate from 2000 onwards, which was continued into the perioperative period. Disease-modifying antirheumatoid drugs (DMARDs) such as Infliximab were discontinued 4 weeks prior to surgery and recommenced at a similar period after.

The primary outcome measure was all-cause mortality. Mortality rates were updated for all patients in 2012 by accessing the National Health Service (NHS) portal to births and deaths; patients were thereby categorized as alive, dead or lost in 2012. The secondary outcome measures were the MDI as described by Casey *et al.*,^[27] and Ranawat grade^[28] at 12 months postoperatively as a measure of change in functional status. This was obtained either by written questionnaire submitted at the time of the outpatient appointment, or by telephone questionnaire conducted by the spinal nurse specialist. For those patients undergoing C1-C2 fixation, the presence of neck pain in the presenting symptomology, and whether this had improved/deteriorated or remained static at 12 months follow-up was also recorded.

Perioperative (defined as within 30 days) surgical complications including mortality were recorded. Instrumentation failure, or requirement for posterior cervical revision surgery, was recorded at any time in the postoperative period within the limit of follow-up.

Statistical analysis

In order to address the question of whether outcomes had changed over the given time period, we subanalyzed and compared survival curves for surgeries occurring between 1981 and 1990 (termed the “early era”) and 2000 and 2010 (termed the “late era”). A statistical comparison was made using log-rank test. A similar comparison was made for change in MDI at 1-year compared to preoperative status between the early and late epochs, using paired Student’s *t*-test analysis. For the rest of comparisons, Student’s *t*-tests were used. In all statistical analysis a cut-off of $P < 0.05$ was deemed significant. The statistical software of SPSS 17.0 (IBM SPSS) and Sigmaplot 12.0 (IBM SPSS) and Excel 2010 (Microsoft) were used for processing and graphical representation.

RESULTS

We identified 224 rheumatoid patients who underwent instrumented posterior cervical/craniocervical spine surgery between 1981 and 2011. The overall mean age (\pm standard deviation) at the time of surgery was 61 (\pm 11) years; the female:male proportion was 180:46 in the entire series. There were 42 cases of rigid OCF; 116 Ransford loop OCFs; 20 cases of Goel-Harms and 46 cases of Magerl transarticular C1-C2 fusion in this series. The mean length of follow-up for all patients was 42 ± 36 months. Table 1 summarizes the basic demographic data for each type of surgical intervention and the preoperative and (1 year) postoperative Ranawat and MDI scores. The mean age of patients undergoing C1-C2 fixation (combining transarticular and Goel-Harms fusions) was 60 ± 12 years, and the mean age for OCF patients (combining Ransford loop and rigid OCF cases) was 62 ± 10 y ($P = 0.2$, t -test). The mode level of inferior fixation for the OCF patients was C5.

Figure 1 displays the trend in instrumentation over the 30 years period, directly comparing the proportion of occipitocervical to C1-C2 fusions; the use of mono-segmental C1-C2 surgery has steadily increased to surpass OCF in the present decade in the surgical management of the rheumatoid spine.

Survival curve analysis

Twenty-five patients were “lost” to data retrieval from the NHS portal for births and deaths in 2012. Of the remaining 199 patients,

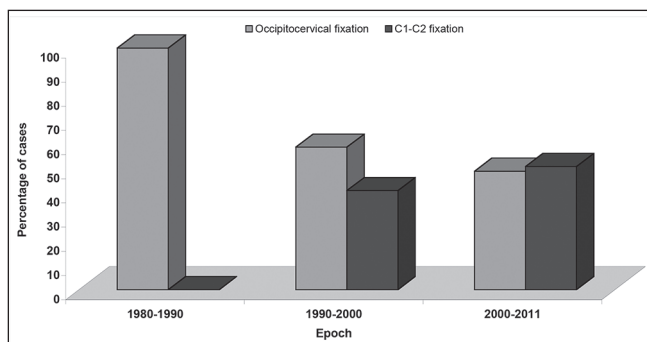


Figure 1: Bar chart depicting the trend in instrumented surgery in the management of rheumatoid craniocervical disease from 1980 to 2011. Occipitocervical fixation has proportionately lost ground, and been superseded by C1-C2 fixation as the primary operative procedure over the last 30 years

in the modern era (between 2000 and 2010) there is a 55% 10-year survival after posterior instrumented surgery for rheumatoid disease. This shows significant improvement compared to the earlier epoch of surgeries performed between 1981 and 1990 ($P = 0.001$, log-rank) [Figure 2]. Patients undergoing C1-C2 fixation have a significantly improved survival compared to those undergoing OCF ($P = 0.01$, log-rank) [Figure 3]. In order to adjust our comparison of the survival curves between the earlier and later epochs of surgery based on possible covariates, we fitted a Cox proportional hazards regression model. None of the covariates tested reached statistical significance; thus, we report in our graphs the unadjusted survival curves with log-rank comparisons.

Functional outcome data

Myelopathy Disability Index data was available for 190/224 (85%), and Ranawat scores for 176/224 (79%) patients at 1-year. There was a statistically significant difference in the baseline (preoperative) degree of myelopathy as measured by the MDI between patients undergoing C1-C2 fixation who had a mean MDI \pm standard deviation of $28.7 \pm 11.4\%$ compared to OCF patients presenting with a mean MDI of $62.6 \pm 15.4\%$ ($P = 0.001$, t -test), as shown in Figure 4. However, both groups

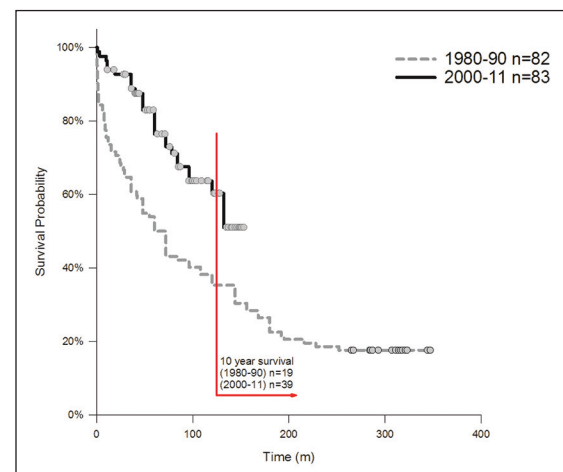


Figure 2: Survival probability has significantly improved in the modern era between 2000 and 2011 compared to the historical era between 1980 and 1990 for rheumatoid patients undergoing craniocervical surgery ($P = 0.001$, log-rank). This improvement is likely to be multifactorial

Table 1: Demographic information, and preoperative and postoperative functional outcome measures for all patients undergoing surgical fixation for rheumatoid cervical spine disease

Surgery	Type of instrumentation	n	Mean age (\pm SD)	Female:male	Mode preoperative Ranawat	Mode postoperative (1 year) Ranawat	Mean preoperative MDI (\pm SD)	Mean postoperative (1 year) MDI (\pm SD)
OCF	Ransford loop	116	63 \pm 9	94:22	IIIa	II	62 \pm 15	57 \pm 15
	Rigid screw-plate	42	61 \pm 13	32:10	IIIa	II	66 \pm 16	47 \pm 17
C1-C2 fixation	Magerl transarticular	46	58 \pm 13	40:6	II	I	30 \pm 13	24 \pm 10
	Goel-Harms fixation	20	65 \pm 10	17:3	I	I	25 \pm 4	15 \pm 4

OCF: Occipitocervical fixation; MDI: Myelopathy disability index (%); SD: Standard deviation

showed improved MDI at 1-year postoperatively [Table 1] ($P = 0.001$ for C1-C2 fusion, and $P = 0.001$ for occipitocervical fusion, paired samples t -test). The mode Ranawat preoperative score for all patients undergoing rheumatoid spine surgery was IIIa, compared to mode postoperative score at 1-year of II.

Taken altogether, functional improvements as measured by preoperative and 1-year postoperative MDI have significantly increased comparing the modern (2000-2011) with historical (1980-1990) eras of surgical intervention ($P = 0.019$, t -test) [Figure 5].

58/66 (88%) of patients who underwent C1-C2 fixation presented with neck pain (mixed axial neck pain and occipital neuralgia). 28/58 (48%) reported improvement in the neck pain at 1-year postoperatively.

Complications

Complications are listed in Table 2. Documentation of complication types varied over the 30-year period, resulting in some discrepancy in the reporting of respiratory and cardiovascular complications between the subgroups. In those undergoing C1-C2 fusion, there was a 9% instrumentation failure rate; five of the seven of the screw breakages were transarticular screws, resulting in a revision to OCF. All-in-all, 7/66 (11%) of patients undergoing C1-C2 fixation for rheumatoid disease required revision to OCF at a mean time of 28 months.

DISCUSSION

In 1987, Zoma and Hamblen wrote that it was difficult to define a clear strategy for the surgical management of symptomatic

Table 2:Thirty days mortality and complication rates for all types of surgical intervention for cervical rheumatoid disease

Instrumentation	Complication (%)
C1-C2 fixation (n=66)	1.5 mortality 9 instrumentation failure (most in first 5 years) 4 occipital neuralgia (worse/new-onset postoperative) 4 vertebral artery injury 3 wound infection
Loop OCF (n=116)	4 mortality 20 respiratory 10 cardiovascular 4 pressure sores 3 instrumentation failure 3 meningitis 1 wound infection
Rigid OCF (n=42)	5 wound infection 5 instrumentation failure 2 vertebral artery injury 2 neurological deficit

OCF: Occipitocervical fixation

instability in the rheumatoid cervical spine because “the results of established procedures were unsatisfactory.”^[29] A year later,

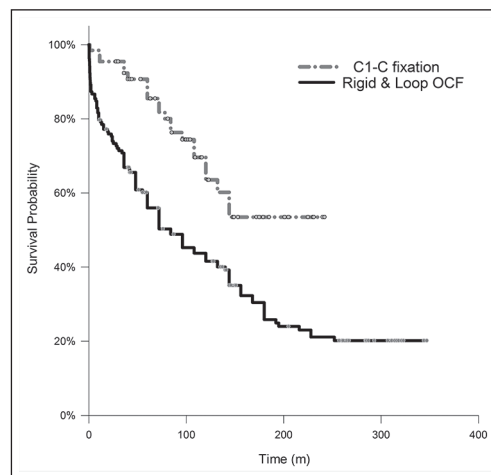


Figure 3: Patients undergoing C1-C2 fixation have a significantly improved overall survival compared to those undergoing occipitocervical fixation (OCF) (OCF, both rigid and loop fixation) ($P = 0.01$, log-rank)

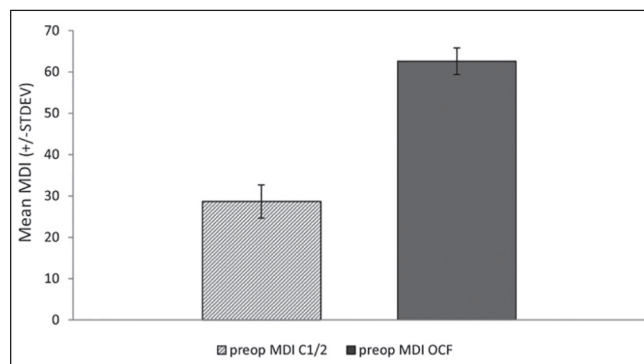


Figure 4: Patients undergoing occipitocervical fixation are significantly more impaired with a higher mean Myelopathy Disability Index at baseline compared to those undergoing C1-C2 fixation ($P = 0.001$). This may reflect intervention earlier in the disease process for the latter procedure

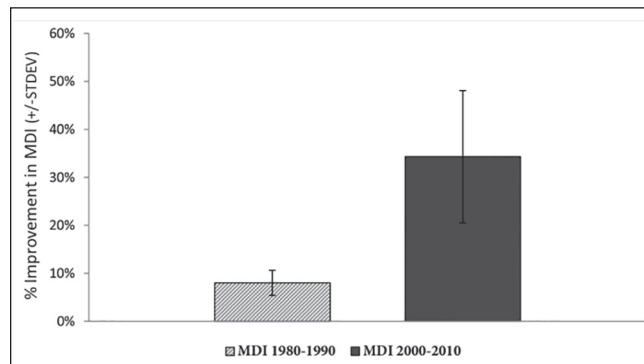


Figure 5: The degree of functional improvement as judged by % change in Myelopathy Disability Index postoperatively is greater in the modern compared to the historical era. This change is likely to be multifactorial, but may reflect the change in surgical technique as shown in Figure 1

Heywood *et al.* posed three questions in relation to cervical instability in rheumatoid arthritis:^[30]

1. What are the indications for fusing an unstable rheumatoid cervical spine?
2. When there is cord compression, is surgical decompression really necessary in addition to fusion?
3. What should the morbidity and mortality figures be? 25 years later, are we any closer to answering these questions?

We present the largest series to date, and our data suggests that not only have the surgical procedures for the rheumatoid spine changed, but so too have the instrumentation and outcomes over the last 30 years. Consistent with the publication of new techniques for posterior cervical fixation, notably C1-C2 transarticular fixation (1987), and Goel-Harms fixation (1994 and 2001), our data reveals that OCF has gradually been superseded as the operative procedure of choice by C1-C2 fixation. Moreover, we have shown that the mean preoperative MDI is significantly lower in those undergoing C1-C2 fixation compared to OCF. This suggests that increasingly, we are operating on rheumatoid patients earlier in the disease process with segment-sparing C1-C2 surgery.

The mortality of rheumatoid patients undergoing cervical spine surgery has significantly improved over the last 30 years. Between 1980 and 1990, the 10-year survival rate was 35%, compared to 55% in the decade of 2000-2010 ($P = 0.001$, LogRank). Our Cox proportional hazards regression analysis failed to identify any covariates that would be influencing statistically the patients' survival. On the other hand, there are confounding factors such as the improvements of perioperative patient care and neuroanesthesia; and the effects of DMARDs as regards the optimization of preoperative general systemic status that we would not be able to test reliably within our dataset. However, this survival improvement could also reflect technological advances in instrumentation materials and techniques, as well as intervention earlier in the disease process. C1-C2 fixation rheumatoid patients were of an equivalent age to patients undergoing OCF at the time of operative intervention ($P = 0.2$, *t*-test), but survived longer on average compared to the latter.

Have there been similar improvements in functionality? This database was limited in some respects: MDI and Ranawat data were reliably collected in a prospective manner for all types of operation; however, we lacked data for a small proportion of patients at follow-up, and lacked sufficient data across the board on VAS or Neck Disability Index to report on neck pain. In the C1-C2 fixation group only, the database documented neck pain and change in this symptom at 1-year, revealing that the commonest presenting symptom in this patient category was neck pain (88%) rather than myelopathy.

A further weakness with this study was the variability of the length of the disease process prior to referral to the spinal unit and definitive surgical treatment. Over the course of the study time frame, referral patterns are likely to have changed, and this is reflected in the baseline MDI [Figure 4].

The trend toward operating earlier in the disease process may represent a mixture of earlier referrals to spine surgeons from rheumatologists (possibly secondary to the publications such as "Surgery on the rheumatoid cervical spine for the nonambulant myelopathic patient — too much, too late" in 1996 by Casey *et al.*^[41]); earlier detection of cranicervical pathology by the increasingly widespread use of various imaging modalities; as well as the development of operative techniques at the C1-C2 junction. Martel reported a 34% incidence of atlantoaxial subluxation (AAS) in unselected patients attending the Arthritis Center;^[31] and Sharp reported that 20% of patients in hospital with rheumatoid arthritis have AAS;^[32] therefore, clearly a risk versus benefit analysis of intervention in the asymptomatic rheumatoid patient with AAS is required, and a watch-and-wait policy in this completely asymptomatic subgroup is reasonable.

In this series, we did not report on the fusion status in follow-up radiography. There are problems in the assessment of whether fusion has occurred after cervical instrumentation particularly with regards to the imaging modality of choice; whether fusion necessarily needs to be osseous or sufficiently fibro-osseous or even fibrous to prevent instrumentation failure; and the time point at which pseudoarthrosis can be determined to have occurred. Moskovich and Apostolides *et al.* described "stable fibrous unions" in a proportion of rheumatoid patients who did not progress to instability or instrumentation failure, but avoided the donor-site morbidity and the requirement for postoperative halo placement.^[21,33] In this series, we have 5/46 (11%) rate of transarticular screw breakage. This may be reflected in the trend in the unit toward Goel-Harms type fusions in the last decade, and the reasons for this high instrumentation failure may be related to the type of screw being inserted at that time and/or the efficacy of additional posterior C1-C2 wiring (Gallie-type fusion) in preventing sagittal plane movement. Nagaria *et al.* did not report instrumentation failure in their series of 37 rheumatoid patients undergoing C1-C2 transarticular fixation^[34] without the addition of Gallie fusion; however, Gluf *et al.* have reported a screw fracture rate of 6.5% in their series of 191 patients (63 rheumatoids). In the latter publication, it was remarked that thicker-core screws had no breakages.^[35]

We did not assess the incidence of subaxial instability after C1-C2 fixation as reported by Clarke *et al.*: 39% after C1-C2 interlaminar wiring,^[15] and Ito and Mukai *et al.*: 18% and 24%, respectively after C1-C2 transarticular screw fixation.^[16,36] Specifically, this may be related to the atlantoaxial kyphosis angle.^[37] This is not necessarily a strong argument for OCF over C1-C2 fusion; however, since OCF, stopping too short of the thoracic spine according to Hirano *et al.*,^[38] can also result in significant subaxial disease progression.^[22,39]

A proportion of patients underwent transoral decompression at some point; but, we have found that we are performing this procedure less commonly compared to over 10 years ago. This might reflect the data presented herein showing intervention earlier in the disease process halting progression to the later stage of vertical translocation and/or pannus development; for example, Landi *et al.* have reported improvement of pannus and anterior cervical compression after posterior fixation.^[11] Even intervention

late in the disease process that is, Ranawat IIIB patients (subclassified into patients who can sit or who are bedridden) can make functional improvements despite increased complications as evidenced by Nannapaneni and Tanouchi *et al.*^[40,41]

CONCLUSION

We have presented both historical and modern reviews of surgery in a single unit for rheumatoid spine disease, showing clear changes in the type of surgery, and stage of intervention. Moreover, we present improvements in mortality and functional outcomes over that time too, although causality underlying this is multifactorial.

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