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ORIGINAL ARTICLE

Health-related quality of life in patients undergoing cervico-thoracic osteotomies for fixed cervico-thoracic kyphosis in patients with ankylosing spondylitis

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Abstract

Purpose Ankylosing spondylitis (AS) can result in severe cervico-thoracic kyphotic deformity (CTKD). Few studies have addressed the relationship between cervico-thoracic osteotomies in AS and health-related quality of life scores. The aim of this study is to evaluate the impact of cervico-thoracic osteotomy (CTO) on improving quality of life for patients with fixed CTKD.

Methods A database of all patients who underwent a CTO for CTKD in patients with AS was created. Data entered into the database consisted of patients' demographics and comorbidities, as well as surgical, clinical and radiological data. The outcome measures used in our study were Neck Disability Index (NDI), EuroQol 5D-5L (EQ-5D-5L) and Visual Analogue Scale. We also measured the following radiological parameters: chin-brow to vertical angle (CBVA), C7-Slope, C2–7 angle, Regional Kyphosis Angle, C2–C7 sagittal vertical axis (SVA) and C7–S1 SVA.

Results A total of 13 male patients with AS were included in our study. The mean age was 57.5 years (40–74); and mean follow-up was 37.6 months (12–78). Following the C7–T1 osteotomy (10 Smith Peterson Osteotomies and 3 Pedicle Subtraction Osteotomies), NDI improved from a mean of 65.54 (SD 8.95) to a mean of 22.09 (SD 6.99). The EQ-5D-5L improved from a mean of 0.41 (SD 0.16) to 0.86 (SD 0.088). Pre-operative CBVA was on average 54° (40°–75°) and post-operative was 7° (2°–12°). There were no major complications, 1 superficial infection and 5 minor nerve root irritations.

Conclusions Cervical osteotomy for the management of fixed flexion deformity of cervical spine in ankylosing spondylitis is a safe procedure and can result in restoration of horizontal gaze and sagittal balance with significant improvement of the patient's health-related quality of life.

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.

Keywords Spinal osteotomy · Ankylosing spondylitis · Outcome · Complications · Quality of life

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Extended author information available on the last page of the article

Introduction

Ankylosing spondylitis (AS) or Strümpell–Marie–Bechterew disease [1, 2] is a chronic inflammatory seronegative arthropathy of unknown aetiology which primarily affects sacroiliac joints and the axial skeleton [3, 4] leading to a painful, stiff and deformed spine. Men are more often affected than women, with a ratio of approximately 3:1 [5]. Patients usually develop symptoms in early adulthood and the most frequent complaints are lower back pain and morning stiffness. In advanced forms, it can be associated with severe postural deformities such as cervico-thoracic kyphotic deformity (CTKD) also known as "chin on chest" deformity [6]. The kyphotic deformity of the cervico-thoracic spine often causes severe pain and loss of horizontal gaze, and interferes with daily activities such as chewing and swallowing. CKTD has a measurable impact on health-related quality of life (HRQOL) [7]. The aim of the cervico-thoracic osteotomy (CTO) is to correct chin on chest deformity, relieve patients' pain, restore forward gaze and improve the health-related quality of life [8]. Various techniques for correcting "chin on chest" deformity have been popularized, starting with the opening-wedge osteotomy in the awake and sitting patient introduced by Urist [9], to posterior column osteotomy [6] and culminating with the posterior-based closing three column osteotomies [6, 8, 10] such as pedicle subtraction osteotomy (PSO) or vertebral column resection (VCR). However, it is less clear from the available literature whether the patients who are subjected to such invasive procedures have a significant improvement in their quality of life as quantified by health-related quality of life measurements. Several papers have shown that ankylosing spondylitis has a significant impact upon an individual's health-related quality of life (HRQOL) [11, 12]. A study [13] investigating a group of 51 patients with ankylosing spondylitis found that 25.4% of the patients had severe functional disability, 60% had mild-to-moderate functional disability, while 5.8% had no functional loss. Corrective procedures for CTKD have the potential to improve the quality of life as measured by HRQOL [10]; however, to our knowledge there has been no study reporting exclusively the clinical and radiological results of CTO and instrumented fusion for patients with ankylosing spondylitis.

Materials and methods

We performed a retrospective study of prospectively collected data from 13 consecutive patients who underwent a CTO for fixed CTKD between 2006 and 2014.

Our department is a tertiary referral spinal centre, with a lot of experience (and history) with complex spinal pathologies. On average, our unit undertakes between 60 and 80 complex adult spinal operations (including osteotomies) per year. The majority are performed by the two senior authors (one of whom is now retired). The number of cervico-thoracic fixations is not greater than 6–8 per annum in non-AS patients.

All patients will have had standing whole spine X-rays, including skull, to assess chin-brow angle, MRI scans as well as a CT. They have a pre-operative work up with the anaesthetic department and the case is discussed in a MDT setting. If there is any concern about the vasculature, then an additional CT angiography is organised.

The information recorded for each patient included: *demographic data*—age, gender, Body Mass Index (BMI) *surgical data*—upper instrumented vertebra (UIV), lowest instrumented vertebra (LIV), previous surgeries, strategy and approach, bone grafts material, osteotomy grades according to Schwab et al. [14], total estimated blood loss (mL), neuromonitoring: loss of MEPs, details for neuromonitoring events, summed surgical time (min), intraoperative none/major/minor complication (acc. to Glassman et al. [15]), details for intra-operative complication (acc. to Glassman et al. [15]) *clinical outcome data*—Neck Disability Index (NDI), EuroQol 5D-5L (EQ-5D-5L) and Visual Analogue Scale (VAS) *radiological parameters*—C7–Slope, C2–C7 angle, C2–C7 Cobb angle, C0–C2 angle, Regional Kyphosis Angle (RKA), C2–C7 sagittal vertical axis (SVA), C2–S1 SVA, C7–S1 SVA. All measurements were performed on a full spine lateral standing radiographs. Research approval was not required, as this study was conducted for 'service evaluation' as per our Hospital's guidelines.

Statistical analysis

Statistical analysis was performed using the Analysis ToolPak for Microsoft Excel. Results are expressed as means, range and standard deviation. Comparison between pre- and postoperative data was performed using paired two-tailed Student *T* test. A p value less than 0.05 was considered statistically significant.

Results

A total of 13 male patients with a mean age of 57.5 years (range 40–74, SD 11.29) underwent C7–T1 osteotomy for ankylosing spondylitis related fixed cervico-thoracic kyphotic deformity. The mean length of follow up was 37.6 months (range 12-78, SD 26.73). The mean BMI was 25.017 kg/m^2 (range 20–31, SD 3.38).

Table 1 Clinical outcomes after CTO for fixed CTKD

	HRQOL scores		*p* value
	Pre-op mean (range) SD	Post-op mean (range) SD	
NDI	65.54 (55–80) 8.95	22.09 (10–25) 6.99	< 0.001
EQ-5D-5L	0.41 (0.15–0.65) 0.16	0.86 (0.8–0.95) 0.088	0.014
VAS	8 (6–9) 0.8	1.7 (0–4) 1.44	< 0.001

NDI Neck Disability Index, *EQ-5D-5L* EuroQol 5D-5L, *VAS* Visual Analogue Scale

Surgical data

Out of the total of 13 cervicothoracic osteotomies, 10 were Smith Peterson type osteotomies and 3 were Pedicle Subtraction Osteotomies. Postoperatively, all patients were immobilised in a halo-vest for a period of 3 months. Prior to the correction of cervicothoracic kyphosis, one patient had a previous L4 PSO and one patient had a previous L3 PSO. Nine patients were instrumented proximally to C3, 3 had the UIV to C4 and one to C2. The LIV was T5 in ten patients and T4 in three patients. Only one patient required anterior support in a form of interbody cage plus plate. The average blood loss was 1200 mL (range 400–3600, SD 920). The average operative time was 450 min (range 300–540, SD 68). All surgical interventions were performed under neuromonitoring. Loss of motor evoked potentials (MEPs) was noted in two cases. In both cases, raising the blood pressure and reducing the correction resulted in restoration of MEPs.

Clinical outcome

Following the CTO for fixed CTKD, all HRQOL scores were significantly improved (Table 1 and Figs. 1, 2).

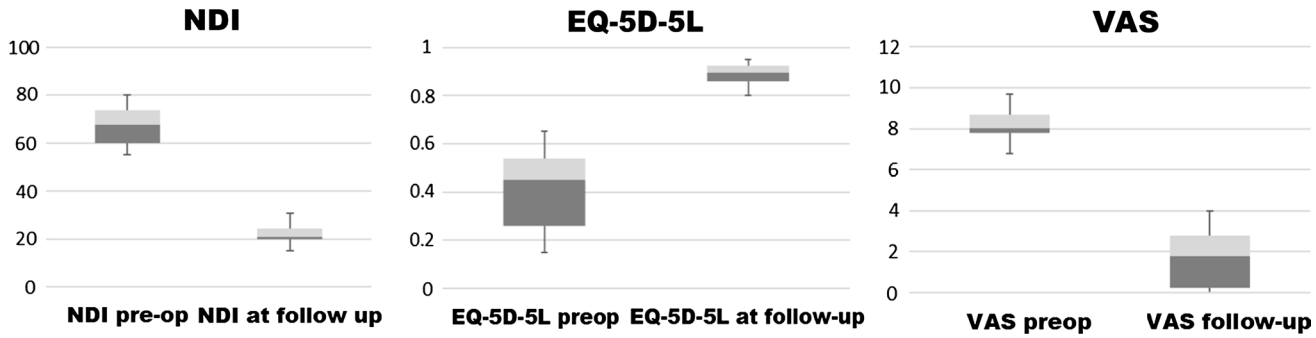

Fig. 1 Comparison of preoperative and postoperative HRQoL scores

Fig. 2 Preoperative and postoperative clinical photography, showing significant improvement in CBVA

Table 2 Pre-operative and post-operative radiographic parameters after CTO for fixed cervico-thoracic kyphosis

	Pre-op mean (range) SD	Post-op mean (range) SD	Follow-up mean (range) SD	*p* value
CBVA	54° (40°–75°) 11.14	7° (2°–12°) 5.04	7° (2°–15°) 5.46	< 0.01
C2–C7 angle (°)	4.92 (1–7.9) 2.24	25.53 (16.8–32.5) 4.77	27.34 (16.8–35.5) 4.77	< 0.01
C0–C2 angle (°)	38.88 (35.9–42.2) 1.93	37.37 (29.5–41.5) 4.17	37.57 (28.5–41.5) 4.17	0.16
RKA angle (°)	14.4 (10.3–16.8) 2.04	− 31.39 (− 21.6; − 35.2) 3.99	− 32.37 (− 21.4; − 35.8) 3.87	< 0.01
C7 slope (°)	67.86 (59.8–80.9) 6.82	40.96 (26.4–55.8) 8.60	41.86 (26.4–54.7) 8.64	< 0.01
C2–C7 SVA (mm)	93.66 (75.8–110) 13.43	39.18 (19–50.4) 11.21	39.98 (19.9–51) 11.82	< 0.01
C2–S1 SVA (mm)	158 (135.8–192.9) 17.11	103.96 (44.8–168) 40.23	112.96 (47.8–172) 41.31	< 0.01
C7–S1 SVA (mm)	64.87 (43.8–95.7) 17.35	66.25 (29–116.1) 28.16	66.41 (28.9–118.1) 30.16	0.44

RKA Regional Kyphosis Angle, *SVA* sagittal vertical axis

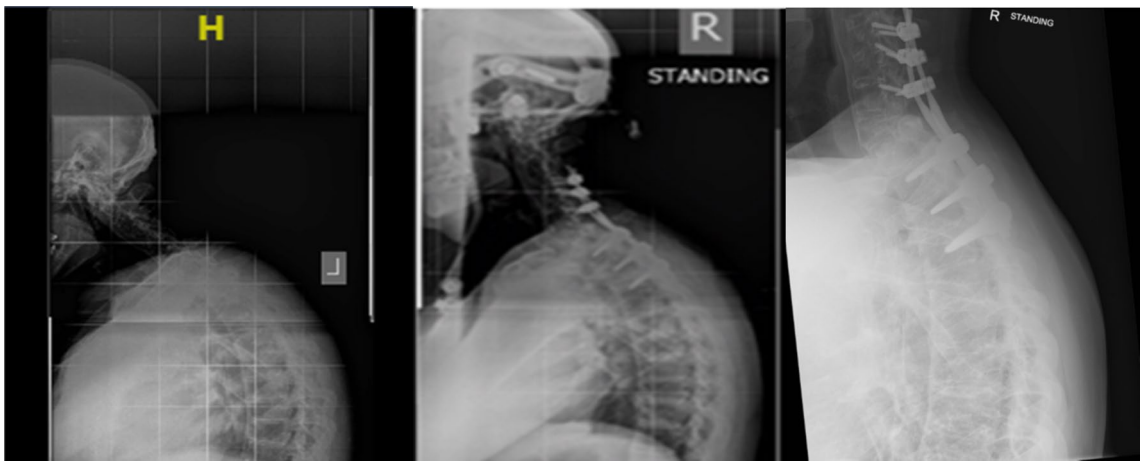

Fig. 3 Preoperative and postoperative lateral X-ray showing improvement in cervico-thoracic sagittal parameters

Radiological parameters

Post-operative radiographic results (Table 2) show significant improvement in C7–Slope, C2–C7 angle, RKA, C2–C7 SVA, and C2–S1 SVA. The chin-brow to vertical angle was on average 54° (range 26 40°–75°, SD 11.14) pre-operatively, and improved to an average of 7° (range 2°–12°, 27 SD 5.04) post-operatively (Fig. 3).

Complications

As expected, the most common complications were post-operative neurological deficit, wound infection and delayed wound healing. No major neurologic loss was encountered in this series, however, 5 out of 13 patients (38%) developed post-operative monoradicular motor deficit grade 3–4/5 on the MRC (Medical Research Council) scale. All patients fully recovered by the 6 months follow up, and no patient required revision surgery during the follow-up period (Table 3).

Table 3 Post-operative complications after CTO

Total 8 (61%)	
Infection	1 (7.7%)
Delayed wound healing	1 (7.7%)
Neurological deficit	5 (38%)
Medical	2 (15.3%)

Discussion

Our study analysed the effect of cervico-thoracic osteotomy on health-related quality of life in patients with ankylosing spondylitis. We performed a retrospective study of prospectively collected data from 13 patients who underwent a CTO for fixed cervico-thoracic kyphotic deformity. All patients exhibited significant improvements in HRQOL scores as well as pain and radiological parameters. Severe cervical thoracic deformity deeply impacts on the health-related quality of life of a patient with AS.

A direct correlation between cervicothoracic kyphosis and HRQOL was recently demonstrated [7]. The main indication for correction of fixed cervical kyphosis in patients with AS is severe deformity causing functional impairment and intractable pain [16]. The inability to look forward not only restricts mouth opening leading to difficulty in chewing and swallowing, but also creates psychological problems, causing anxiety and difficult interpersonal communication. Spinal osteotomies have been shown to be effective in restoring sagittal balance and improving health-related quality of life in patients with severe thoraco-lumbar and cervico-thoracic deformities [17, 18]. However, the available literature on cervico-thoracic osteotomies either lacks validated outcome scores or reports on heterogeneous groups of patients [10], rather than concentrating on a specific pathology. A PubMed search among English literature published in the last 10 years, with the following keywords: "ankylosing spondylitis; spine; osteotomy; cervical", produced only 24 papers. Excluding case reports and papers dealing with cervical kyphosis related to other aetiologies, the remaining subgroup is made up of only 17 papers. It is, therefore, evident that the number of patients affected by severe cervical thoracic kyphosis requiring surgery is relatively small and reporting on the clinical or radiological outcomes of surgical intervention based on a large series of patients is a difficult undertaking (Table 4).

Two main posterior techniques are used to address the problem of the CTKD: the Smith Petersen osteotomy, also known as open wedge osteotomy (OWO) [6], and the pedicle subtraction osteotomy [18] or closed wedge osteotomy. The OWO was first described by Smith-Petersen in 1945. Subsequently, variations have been introduced and then applied to treat CTKD in AS patients. In the majority of cases, the osteotomy was performed at the cervicothoracic junction. This location was selected because of the large size of the spinal canal at C7–T1, the mobility of the spinal cord, the position of the vertebral artery in front of the transverse process of C7 and the presence of the eighth cervical nerve in this region. The Smith-Petersen procedure begins with a laminectomy of C6, C7, and T1. Next, a wide lateral decompression of the C8 nerve roots is performed by removing both C7–T1 facet joints and most or all of the C7 pedicles. The correction is based upon the creation of an opening wedge by breaking the ankylosed spine throw the anterior column. As such, all three spinal columns are disrupted during the Smith-Peterson osteotomy creating inherent spinal instability. Arthrodesis is dependent upon the posterior bony elements as the anterior column is lengthened [19, 20]. Recently, pedicle subtraction procedures are being used more commonly because of increased stability at the osteotomy site, which has been biomechanically demonstrated [21]. PSO is performed by creating a closing wedge through the vertebral body, which is more stable than the opening wedge produced by the Smith-Petersen procedure. Furthermore, the anterior column remains intact and is not lengthened, which further enhances the stability of the spine following the procedure [16].

Weatherley et al. [22], reported on two cases of vascular injury associated with SPO in AS finding that the aorta lengthened by 2 cm for a 45° correction. Indeed, others too have reported that kyphosis correction with a sharp lordotic angle, L1/L2 levels and L2/3 levels [23], as well as correction greater than 40 degrees predisposed to a higher risk of aortic injury [23–25]. Fortunately, no vascular injuries were observed in our study.

There are other techniques described for rigid thoracolumbar kyphosis in AS such as vertebral column decancellation [26] and V–Y osteotomy [27]. These open the anterior column (rather than pivoting on this), and thus could potentially involve greater risk to the aorta.

Over the last few years, in such complex patients with abnormal anatomy, we have relied on navigated guidance systems to allow for better accuracy of pedicle screw insertion, as well as shortening operative time and potential morbidity. With greater experience and numbers, we hope to publish our results. One case report dealt with simultaneous navigated cervico-thoracic and thoraco-lumbar fixation for a multiple segment fractures with a good outcome [28].

The optimal chin-brow angle is also interesting. Suk et al. [29] had stated that the patients with CBVA between − 10° and 10° had better horizontal gaze. In their study of 25 AS thoracolumbar kyphotic patients with cervical ankyloses, Song et al. [30], found that a CBVA of 10°–20° was optimal for a better quality of life, especially for household chores as well as working at a desk. In our series, this improved to a mean of 7°.

In our series, improvement in cervicothoracic alignment resulted in significant improvement in patients' quality of life as demonstrated by the measured HRQOL scores—NDI, EQ-5D-5L and VAS scores all significantly improving at follow up. Spinal cord injury and paralysis are the most feared complications of cervicothoracic osteotomy. Although this complication was not encountered in our series, we did notice C7 and C8 palsies in five cases. Nevertheless, all

Table 4 Details of neurological complication

	Neurological complication	Grade
Patient 2	Bilateral C8 motor deficit	MRC 4/5
Patient 3	Left C8 motor deficit	MRC 3/5
Patient 4	Right C7 motor deficit	MRC 4/5
Patient 5	Bilateral C7 motor deficit	MRC 3/5
Patient 11	Left C7 motor deficit	MRC 4/5

MRC Medical Research Council, *ASIA AIS* American Spinal Injury Association Impairment Scale

patients fully recovered following conservative treatment. The avoidance of post-operative dysphagia, and the need for tracheostomy with posterior approaches as compared with the anterior approach, was highlighted by Theologis et al. [10]. In our series, none of the patients developed post-operative dysphagia nor did they require tracheostomy. Our study has several limitations. Our database did not include disease activity scores such as Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) or functional limitations scores such as Bath Ankylosing Spondylitis Functional Index (BASFI). The retrospective nature of the study plus the small number of patients included may have affected the results. However, as previously stated, CTOs are rarely encountered, even in a specialist centre for spinal deformities. Nevertheless, the improvement in the radiological parameters and HRQOL scores was statistically significant in our series. Our patients' data continue to be entered into a multicentre European database as a collaboration with the Cervical Spine Research Society (CSRS).

Conclusions

Cervical-thoracic osteotomy for the management of fixed flexion deformity in patients with ankylosing spondylitis results in significant improvements in health-related quality of life scores as well as radiological outcome. There were no major complications in our series and five minor neuropraxia which fully recovered.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

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