Adjacent-level failures after occipito-thoracic fusion for rheumatoid cervical disorders

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Received: 11 January 2013 / Revised: 1 December 2013 / Accepted: 1 December 2013 / Published online: 13 December 2013
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Abstract

Introduction The natural history of cervical spine lesions in rheumatoid arthritis (RA) is variable. We have actively performed occipito-thoracic fusion for severe destructive rheumatoid cervical disorders and reported its clinical results and complications. In our previous study, the most frequent complication was the adjacent-level failures caused by the fragile spine. The objective of this study was to determine risk factors for adjacent-level failures after occipito-thoracic fusion.

Materials and Methods Subjects were 35 RA patients (31 females and 4 males) who underwent occipito-thoracic fusion using RRS Loop Spinal System® (Robert Reid Inc. Tokyo, Japan), and the incidence and characteristics of adjacent-level failures were investigated. Furthermore, the adjacent-level failures were divided into two types according to their levels, fracture at the lowest level of the fusion area and that at the level inferior to the fusion area, and the characteristics of each type were evaluated.

Results and Conclusion Nine (26 %) of 35 patients suffered adjacent-level failures (10 vertebral fractures). Adjacent-level failures occurred when the distance of fixation was “O–T4” or longer. The long fusion might cause adjacent-level failures due to greater mechanical stress. Seven fractures occurred at the lowest level of the fusion area, and all of them were cured without symptoms by conservative treatment. Three fractures occurred at the level inferior to the fusion area, and one of them needed additional surgery due to

sudden paraplegia resulting from collapse of the adjacent vertebra. After occipito-thoracic fusion, burst fractures at the level inferior to the fusion area might cause sudden paraplegia, and therefore a careful observation should be required for patients with these fractures.

Keywords Rheumatoid arthritis · Adjacent instability · Cervical spine · Occipito-thoracic fusion · Distal junctional disease

Introduction

The cervical spine is frequently affected in patients with rheumatoid arthritis (RA), and its instability and deformity cause clinically severe occipito-cervical pain and neurological deficits. Especially, mutilating-type RA (MUD), the most aggressive form of RA, is frequently associated with severe cervical involvement at both the cranio-vertebral junction (CVJ) and the subaxial levels, and it often requires entire cervical fusion surgery [1–3]. In addition, even in patients with the more erosive subset (MES) and with the less erosive subset (LES), additional surgery is sometimes necessary due to new cervical and/or upper thoracic involvements after atlanto-axial or occipito-cervical fusion [4]. In these cases, we had actively performed occipito-thoracic fusion since 1991 [5]. In this surgery, the occipito-cervical motion was completely lost. In our previous report [5], adjacent-level failures were the most frequent complications after occipito-thoracic fusion. Since we had started to apply occipito-thoracic fusion, other complications had decreased, contrasting with increased adjacent-level failures [6]. In this study, we therefore evaluated the incidence and risk factors of adjacent-level failures after occipito-thoracic fusion for rheumatoid cervical disorders.

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Materials and methods

Patients

Thirty-five patients (31 females and 4 males) with rheumatoid arthritis (28 MUD and 7 MES) having severe cervical disorders underwent occipito-thoracic fusion between 2002 and 2010. The mean age at the time of surgery was 64.5 years (range 47–78 years). The duration of RA was 23.2 years on average (range 3–51 years). The main clinical finding was either neurological deficit due to spinal cord compression or intractable neck-occipital pain. The neurological status before surgery was Ranawat class II in 1 patient, IIIA in 10, and IIIB in 24. Two-thirds of patients could not walk preoperatively. Mean postoperative follow-up period was 42.9 months (range 1–108 months). The patient characteristics are summarized in Table 1. Cervical involvements were diagnosed by plain cervical radiographs, computed tomographic myelograms (CTM), and magnetic resonance images (MRI). Cervical involvements develop in the atlanto-axial joint, begin with anterior subluxation (AAS) and may progress to vertical subluxation (VS) and/or subaxial subluxation (SAS). In this series, AAS + VS was observed in 5 patients, AAS + SAS in 1, AAS + VS + SAS in 20, SAS + ankylosis at CVJ in 3, and SAS + anomaly at CVJ in 2 patients. Four patients had received previous surgery of the cervical spine (three had atlanto-axial fusion and one occipito-cervical fusion). In these patients, SAS and/or VS occurred, although solid fusion had once been achieved after previous fusion. The cervical involvements are summarized in Table 2.

Surgical procedures

During operation, segmental posterior decompression was performed in cases with local stenosis. This procedure was then supplemented with posterior spinal fixation using RRS Loop Spinal System[®] (Robert Reid Inc. Tokyo, Japan).

Table 2 Cervical involvements ($n = 35$)

	MES ($n = 7$)	MUD ($n = 28$)	Total ($n = 35$)
AAS + VS	0	5	5
AAS + SAS	1	0	1
AAS + VS + SAS	2	18	20
SAS + ankylosis at CVJ	0	3	3
SAS + anomaly at CVJ	2	0	2
Salvage case	2	2	4

AAS atlanto-axial subluxation, VS vertical subluxation, SAS subaxial subluxation, CVJ cranio-vertebral junction

Table 1 Patient profile ($n = 35$)

Gender	4 males 31 females
Age at surgery	64.5 years (37–78 years)
RA disease duration	23.2 years (3–51 years)
Type of RA	MES 7, MUD 28
Medication	PSL in 32 patients (avr. 5.66 mg), MTX in 13 (avr. 4.92 mg), DMARD in 11, BAs in 2 patients
No. of artificial joints	1.34
Neurological status (Ranawat classification)	Class II in 1, Class IIIA in 10, Class IIIB in 24
Follow-up	42.9 months (1–108 months)

MES more erosive subset, MUD mutilating-type disease, PSL prednisolone, MTX methotrexate, BAs biological agents

Basically, the fusion area was from the occipital bone to T5 in MUD patients, because most of these cases had multi-segmental destructive whole cervical spine instability with the fragile bone, and we could not find a reliable anchoring place at the lower cervical and upper thoracic spine.

We selected end vertebrae based on the evaluation of upper thoracic involvements using multiple reconstruction views on CTM and MRI. If the patients had upper thoracic involvements (fracture, listhesis, instability), we included these vertebrae to the fusion area. In fact, occipito-thoracic fusion of O–T1 was performed in one patient, O–T2 in 3, O–T3 in 1, O–T4 in 2, O–T5 in 24, O–T6 in 2, and O–T7 in 1.

As anchors of the caudal end vertebrae, pedicle screws (PS) were used in 1 patient, PS + facet hooks in 3, and multiple thoracic hooks in 31. In the surgical method using multiple thoracic hooks, down-going transverse hooks and up-going facet hooks were combined to block up the thoracic spine; we had used this method since 1991 (Fig. 1) [7]. We selected PS alone for short occipito-thoracic fusion (“O–T1”), and mainly used multiple thoracic hooks for fusion from the occipital bone to the 4th thoracic vertebra or longer (“O–T4” or longer distance). Instrumentations and fusion areas are shown in Table 3. Postoperatively, a SOMI brace was applied for 4–6 months.

Clinical evaluation

We firstly investigated the incidence of adjacent-level failures (e.g., vertebral fractures, subluxations, and the dislodgement of distal anchors). Then, correlation of disease parameters and joint involvements to the adjacent-level failures (i.e., age, gender, duration of RA, type of RA, medication, number of artificial joints, and neurological status before surgery) were investigated. Furthermore, the adjacent-level failures were divided into two types (Types A and B) according to their levels (Fig. 2) and the

Fig. 1 Schema of multiple thoracic hooks (Reprinted from [5]). multiple thoracic hooks are combination of transverse hooks and facet hooks. In the case who received fixation from the occipital bone to T5, we used down-going transverse hooks at T2 and T3, and up-going facet hooks at T4 and T5

characteristic of each type was evaluated. Type A included the fracture at the lowest level of the fusion area, and Type B at the level inferior to the fusion area. Hirano et al. reported a similar classification in 2010 [8]. All parameters were analyzed using Chi-square test and Mann–Whitney test. A value of $P = 0.05$ was considered to be significant.

Results

Incidence of adjacent-level failures (Fig. 3)

Nine of 35 patients (26 %) suffered ten vertebral fractures as adjacent-level failures after surgery. There were no posterior element fractures with the anterior vertebral column fractures. No patient developed new thoracic subluxation at the adjacent level, or had implant failure or dislodgment of distal end anchors. The adjacent-level failures occurred in patients who received fixation of “O–T4” or longer distance (T4 in 1 of 2 cases, T5 in 7 of 25, and T7 in 1 of 1). The patients were divided into two groups according to fusion area. Six patients

Table 3 Fusion area and anchors of the distal end vertebra

Anchors	Fusion area (from occiput to)							Total
	T1	T2	T3	T4	T5	T6	T7	
PS alone	2							2
PS + facet hook		3	1				1	5
MTH				2	24	2		28
Total	2	3	1	2	24	2	1	35

PS pedicle screw, MTH multiple thoracic hooks

Fig. 2 Classification of adjacent-level failures; type A: fracture at the lowest level within the fusion area, type B: fracture at the level inferior to the fusion area

Fig. 3 The fixation level of the adjacent-level failure cases. The failures occurred in patients who were fixed O–T4 or longer

received a fixation that was shorter than O–T4 fusion, and 29 patients received a fixation that was longer than or equal to O–T4 fusion. The number of cases that showed adjacent-level failures is shown in Table 4. The percentage of patients who showed adjacent-level failures was greater in the longer fixation group (9 of 24, 31 %) than in the shorter group (0 of 6, 0 %), but this was not statistically significant ($P = 0.14$). (Table 4).

Correlation of disease parameters to the adjacent-level failures

There were no differences in the characteristics (i.e., gender, age, duration of RA, type of RA, medication, the number of artificial joints, and neurological status before surgery) between patients with and without adjacent-level failures. The results are summarized in Table 5.

Table 4 Correlation of fusion area to the adjacent-level failures

Fusion area (from occiput to)							Total
T1	T2	T3	T4	T5	T6	T7	
Adjacent-level failures							
0/1	0/3	0/1	1/2	7/24	0/2	1/1	9/35
Shorter fixation (T1–T3)			Longer fixation (T4–T7)				$P = 0.14^*$
0/6 (0 %)			9/29 (31 %)				

* Chi-square test

Table 5 Correlation of disease parameters to the adjacent-level failures

	Adjacent-level failures (+) ($n = 9$)	Adjacent-level failures (–) ($n = 26$)	P value
Gender	M:2, F:7	M:2, F:22	0.62*
Age at surgery	64.6	64.5	0.97**
Duration of RA	20.6	23.9	0.52*
Type of RA	MES; 3 MUD; 6	MES; 5 MUD; 21	0.68*
Medication	PSL = 5.9 mg	PSL = 4.6 mg	0.14**
	MTX = 2.5 mg	MTX = 1.6 mg	0.66**
	BAs = 1 case	BAs = 1 case	0.93*
No. of artificial Joints	0.89	1.46	0.37**
Neurological status	II = 1, IIIA = 3, IIIB = 5	IIIA = 7, IIIB = 19	0.33*

MES more erosive subject, MUD mutilating-type disease, PSL prednisolone, MTX methotrexate, BAs biological agents

* Chi-Square test

** Mann–Whitney test

Classification of adjacent-level failures

In ten vertebral fractures, seven occurred at the lowest level of the fusion area (Type A) and three at a level inferior to the fusion area (Type B). Type A included six compression fractures and one burst fracture. These fractures occurred within 1 month after surgery, and all cases were cured with no symptoms by conservative treatment. Type B included one compression fracture and two burst fractures. Two-thirds of them were unstable fractures, and one patient finally developed sudden paraplegia resulting from collapse of the adjacent vertebra (T6).

Case series

Case 1 (Fig. 4)

A 69-year-old woman had rheumatoid arthritis with typical mutilating-type joint involvements. She had suffered RA for 30 years and cervical myelopathy for 3 months. Her

preoperative neurological status was Ranawat class IIIA. The lateral radiograph in the neutral position showed AAS + VS and severe anterior subluxation at C3/4 and C5/6 levels, and MRI revealed cervical cord compression at C3/4 and C5/6. “O–T5” fusion using RRS Loop Spinal System® (Robert Reid Inc. Tokyo, Japan) with multiple thoracic hooks was performed. One month after surgery, she had a vertebral fracture of T5 at the lowest level of the fusion area (Type A). This fracture had not worsened and was cured with no complaint by conservative treatment. Two years after surgery, she developed a vertebral fracture of T6 at a level inferior to the fusion area (Type B) which had collapsed. She had kyphotic deformity, but she did not have new neurological deficit. We continued with routine follow-up carefully.

Case 2 (Fig. 5)

A 66-year-old woman had MES type RA. She had suffered RA for 19 years. Her preoperative neurological status was Ranawat class IIIA. She previously underwent “O–C2” fusion for AAS + mild VS and had achieved solid fusion. Seven years after surgery, SAS occurred and her myelopathy had gradually worsened. A lateral view of the cervical spine X-ray showed severe anterior subluxation and instability at the C3/4 level, and MRI revealed cervical cord compression at C3/4. She underwent “O–T5” fusion using RRS Loop Spinal System® (Robert Reid Inc. Tokyo, Japan) without perioperative complications. However, 3 months after surgery, she developed sudden paraplegia resulting from the collapse of T6 at a level inferior to the fusion area (Type B) and instrumented fusion was extended to L2. This patient remained bedridden even after revision surgery.

Discussion

The cervical spine is frequently affected in rheumatoid arthritis (RA), and various surgical methods have been reported to manage these cervical lesions, including atlanto-axial fusion and occipito-cervical fusion. Several studies have reported that, in operated patients, additional long fusion surgery have been caused by newly developed instability and decompensation of the adjacent segments after the atlanto-axial or occipito-cervical fusions [9–14]. Newly developed subluxation below a fused segment may occur in 20–36 % of RA patients after cervical fusion [12–14]. However, there are few reports about adjacent-level failures after occipito-thoracic fusion for severe destructive cervical lesions in RA patients. Hirano et al. [8] reported that 11 of 56 patients (19.6 %) suffered thoracic spine lesions (10 fractures and 1 subluxation) after occipito-thoracic fusion during the follow-up period of 36 months.

Fig. 4 a Typical mutilating-type RA patients with cervical myelopathy (IIIA). Destructive cervical spine involvements (a X-ray, b MRI). She underwent O–T5 fusion with multiple thoracic hooks. Postoperative X-ray (c, d). Postoperative 1 month, T5 fracture at the

lowest level within the fusion area (f). Postoperative 2 years, T6 fracture at the level inferior to the fusion (g). T6 had collapsed and she had kyphotic deformity (h)

Fig. 5 MES type RA patient with cervical myelopathy (IIIA). She had cranio-vertebral involvement (AAS and mild VS) and underwent O–C2 fusion (a, b). 7 years later, anterior subluxation and instability at the C3/4 level had occurred (c, d). O–T5 fusion with multiple

thoracic hooks for salvage surgery (e, f). Postoperative 3 months, T6 burst fracture at the level inferior fusion caused sudden paraplegia (g, h). She underwent extended surgery to L2 (i, j)

In our study, 9 of 35 cases (26 %) developed adjacent-level failures. These results indicated that the incidence of adjacent-level failures after OTF was high.

To avoid adjacent-level failures after occipito-thoracic fusion, we evaluated their risk factors. Consequently, there were no significant differences in preoperative characteristics between patients with and without adjacent-level failures. However, there might be a couple of interesting different tendencies as follows. Firstly, regarding the fixation area, adjacent-level failures occurred in patients who received fixation of “O–T4” or longer distance, which suggested that the long fusion might cause adjacent-level failures due to greater mechanical stress. In a similar way, Song et al. [15, 16] reported that fusion may increase mechanical stress at adjacent disc levels and lead to adjacent segment disease, and that multi-level fusion accelerated the severity of adjacent-level degeneration as compared to single-level fusion in degenerative cervical disease.

Secondly, fractures at the lowest level of fusion area (Type A) were stable and were cured without symptom by conservative treatment. On the other hand, fractures at the level inferior to the fusion area (Type B) were few, but may cause sudden paraplegia. We, therefore, should carefully observe patients with type B fractures.

Thirdly, concerning distal anchors, Hirano et al. [8] report that spinal compression fracture at the lowest level within the fixation area is a serious problem that can lead to pullout of pedicle screws. We used multiple thoracic hooks for cases receiving fusion of “O–T4” or longer distance, and there were no cases that developed back-out of distal anchors. Adjacent-level failures frequently occur after occipito-thoracic fusion in rheumatoid patients, because they have multiple spinal involvement and severe osteoporotic bone. Thus, we should use safety anchors at the distal end of the stabilization area so as not to induce renewed neurological deficits due to adjacent-level failures. We believed that multiple thoracic hooks were stronger and safer than pedicle screws for the fragile spine.

Conclusion

Based on our results, it was difficult to completely prevent the development of adjacent-level failures, but surgeons should avoid long fusion such as fusion of “O–T4” or longer distance as much as possible, and make careful observation especially for adjacent vertebral fractures at a level inferior to the fusion area. In addition, multiple thoracic hooks construct is recognized as a reliable anchoring method to resist fragile bone.

Conflict of interest None.

References

1. Ochi T, Iwase R, Yonemasu K et al (1988) Natural course of joint destruction and fluctuation of serum C1q levels in patients with rheumatoid arthritis. *Arthritis Rheum* 31(1):37–43
2. Omura K, Hukuda S, Katsuura A et al (2002) Evaluation of posterior long fusion versus conservative treatment for the progressive rheumatoid cervical spine. *Spine* 27(12):1336–1345
3. Matsuyama Y, Kawakami N, Yoshihara H et al (2005) Long-term results of occipitocervical fusion surgery in RA patients with destruction of the cervical spine. *J Spinal Disord Tech* 18(Suppl):S101–S106
4. Shimizu T, Tanouchi T, Ino M et al (2009) Revision surgeries following cervical fusion in rheumatoid arthritis: What should we learn from these cases?. In: 38th annual meeting of Japanese Society for Spine Surgery and Related Research Abstract book, p 380
5. Tanouchi T, Shimizu T, Fueki K et al (2012) Neurological improvement and prognosis after occipito-thoracic fusion in patients with mutilating-type rheumatoid arthritis. *Eur Spine J* 21(21):2506–2511
6. Tanouchi T, Shimizu T, Fueki K et al (2008) Complication and prognosis after occipito-thoracic fusion in mutilating type of rheumatoid arthritis. In: 37th annual meeting of Japanese Society for Spine Surgery and Related Research Abstract book, pp 79–80
7. Shimizu T, Edakuni H, Shimada H et al (1996) Occipito-cervical, thoracic fusion. In: Koichiro Hayashi (ed) *OS now No21. Medical view*, Tokyo, pp 35–47 In Japanese
8. Hirano K, Matsuyama Y, Sakai Y et al (2010) Surgical complications and management of occipitocervical fusion for cervical destructive lesions in RA patients. *J Spinal Disord Tech* 23(2):121–126
9. Ishi K, Matsumoto M, Takahashi Y et al (2010) Risk factors for development of subaxial subluxations following atlantoaxial arthrodesis for atlantoaxial subluxations in rheumatoid arthritis. *Spine* 35:1551–1555
10. Iizuka H, Iizuka Y, Kobayashi R et al (2013) Effect of a reduction of the atlanto-axial angle on the cranio-cervical and subaxial angles following atlanto-axial arthrodesis in rheumatoid arthritis. *Eur Spine J* 22(5):1137–1141
11. Mori K, Imai S, Omura K et al (2010) Clinical output of the rheumatoid cervical spine in patients with mutilating-type joint involvement: for better activities of daily living and longer survival. *Spine* 35(13):1279–1284
12. Kraus DR, Peppelman WC, Agarwal AK et al (1991) Incidence of subaxial subluxation in patients with generalized rheumatoid arthritis who have had previous occipital cervical fusions. *Spine* 16:S486–S489
13. Matsunaga S, Ijiri K, Koga H (2000) Results of a longer than 10-year follow-up of patients with rheumatoid arthritis treated by occipitocervical fusion. *Spine* 25:1749–1753
14. Mori T, Matsunaga S, Sunahara N et al (1998) 3- to 11-year followup of occipitocervical fusion for rheumatoid arthritis. *Clin Orthop Relat Res* 351:169–179
15. Song KJ, Choi BW, Jeon TS, Lee KB, Chang H (2011) Adjacent segment degenerative disease: is it due to disease progression or a fusion-associated phenomenon? Comparison between segments adjacent to the fused and non-fused segments. *Eur Spine J* 20(11):1940–1945
16. Song KJ, Lee SK, Song JH, Choi BY (2009) The comparison of multi-level fusion versus one-level fusion to the development of adjacent level degeneration in anterior cervical arthrodesis with PEEK cage and plate augmentation for the degenerative cervical spinal disorders. *J Korean Orthop Assoc* 44(6):613–618