Revision surgery of irreducible atlantoaxial dislocation: a retrospective study of 16 cases

Mingsheng Tan · Xin Jiang · Ping Yi ·
Feng Yang · Xiangsheng Tang · Qingying Hao ·
Guangbo Zhang

Received: 13 September 2010 / Revised: 8 May 2011 / Accepted: 22 May 2011 / Published online: 8 June 2011
© Springer-Verlag 2011

Abstract There is lack data concerning anterior cervical spine revision surgeries; even more data are missing concerning posterior cervical revision surgeries, to determine the feasibility, safety, and clinical efficacy of revision surgery for irreducible atlantoaxial dislocation (RS-IAAD). Patients with IAAD-FS underwent one-stage transoral release and posterior reduction. Their medical history was documented in detail. The JOA score system was used to evaluate each patient's neurological status pre and post-operatively, and serial MRI and radiographs were used to determine the status of the reduction and the autografts. 16 patients (average age, 36 years old) underwent successful surgery. There was no intraoperative or postoperative neurological deficit except in two cases that suffered transient neurological deficit that alleviated after conservative treatment. Solid bony union was seen at the end of 3 months after surgery in all patients. The mean follow-up period was 28.8 months (range 18–66 months). No pseudarthrosis was noted. Anterior transoral release and posterior instrumented fusion remain significant surgeries with the potential for serious complications, but in the current series there were no major complications.

Keywords Revision surgery · Atlantoaxial dislocation · Transoral approach · Posterior reduction

Introduction

The fact that C1/2 is adjacent to medulla oblongata and that there is no clear anatomic margin due to prior surgical scar tissue make revision surgery more difficult and risky. Anterior transoral odontoid resection, combined with posterior decompression, instrumentation and craniocervical fusion in situ predispose cerebrospinal fluid leakage and neurological deficit at high risk [1–3]. Theoretically, IAAD can be reduced by anterior transoral soft tissue release alone plus posterior instrumentation and fusion, and some surgeons have applied this philosophy to the treatment of IAAD before [4]. However, few literatures presently dated the revision surgery with respect to IAAD-FS. In the past 8 years, we have done a series of studies in anatomy and clinic surgery. Here, we report a series of 16 craniocervical revision surgery.

Methods

Patients and procedures

Between Oct 2001 and Oct 2009, a consecutive 16 patients (9 males and 7 females) with the mean age at previous surgery of 32.1 years (range 7–56 years) were reviewed retrospectively. There were os odontoidum in three patients, occipitalization in eight cases, malunion of odontoid fracture in four cases, and malfunction of atlas transverse ligament in one case (AADL > 5 mm). The mean age at revision surgery was 36.3 years (range 10–57 years), with an interval of 42.2 months between two surgeries (range 11–158 months). All surgeries were performed by the same senior spine surgeon. 16 patients presented with neck pain, 5 patients had history of severe

M. Tan · X. Jiang (✉) · P. Yi · F. Yang · X. Tang · Q. Hao ·
G. Zhang
Department of Orthopedic Surgery, China-Japan Friendship
Hospital, Ministry of Health, Yinghua East Street,
Chaoyang District, Beijing 100029,
People's Republic of China
e-mail: michaeljiangx@yahoo.com.cn

Fig. 1 Surgical illustration. **a** Irreducible atlantoaxial dislocation: contracted scar tissue, bony graft and instruments in front and rear of C1/2 vertebral, respectively. **b** Anterior surgical procedure: resection of contracted scar, a 10 mm width bone elevator was placed into C1/2 facet joint to distract the facet joint space of over 3 mm;

c Posterior removal of wires, bony graft and C1 posterior arch as well as posterior edge of foramen magnum; C1/2 pedicle screws instrumentation is performed. **d** Step by step screw tightening cautiously under fluoroscopy and SEP monitor results in C1/2 reduction; Auto-bone graft was performed afterwards

trauma to the head or neck, and 2 patients had congenital short neck and swan neck deformity. 14 patients presented with mild to moderate spastic quadricparesis and Hoffmann's sign (+). Their sensory deficit was relatively milder than motor deficit (Fig. 1).

A retrospective review of the demographic data, clinical outcome and radiographic findings was performed. Our indications for the revision surgery were neurologic deficit and/or severe spinal cord compression. All patients' neurofunction were assessed using the Japanese Orthopedic Association (JOA) score (100 score) [5].

Preoperative plain radiograph, magnetic resonance imaging, and three-dimensional CT scan were used to determine the deformity and nature of neurological compression. Flexion–extension lateral X-ray before operation and at 3, 6, 12, 24 months and at annual follow-up thereafter were performed. The cervicomedullary angle (CMA) on each MRI was measured.

Operative technique

Intraoperative Somatosensory-Evoked Potentials (SEP) were used in all cases

Transoral anterior atlantoaxial joint release: the patient's tongue and endotracheal tube are retracted caudally by a retractor blade. A malleable retractor blade is used to displace the soft palate and uvula superiorly to provide access to the upper posterior oropharynx. Teeth guards are attached to the retractor frame protect the patient's teeth. The oropharynx and the retractors are sterilized with povidone–iodine solution after the retractors have been positioned.

An intraoperative radiograph is obtained to judge the spinal alignment after positioning and confirm the extent of

the cephalad and caudal exposure provided by the retractor system. The C1 tubercle is palpated to verify the position of the midline. A linear midline incision is created in the median of the posterior pharyngeal wall. Periosteal elevators are used to separate the anterior longitudinal ligament subperiosteally and tissue flap from the anterior elements of the C1–C2. The inferior rim of the anterior C1 arch is dissected to expose the base of odontoid process using high-speed blur. Excision of the scar tissue around the anterior lateral mass was performed. If bony formation impeding reduction was evidenced in anterior atlanto-odontoid space on 3-dimensional reconstruction, good reduction could be achieved by the resection of the anterior arch of C1 and a soft tissue release between the odontoid process and the anterior arch of C1. In most cases, little motion was obtained because the anteromedially shifted lateral mass of the atlas was overlapped and arthrodesed partially with the anterolateral surface of the vertebral body of the axis. Then an elevator of 10-mm width was inserted into the left and right facet joint space, respectively, the atlas joint space between the lateral mass of the atlas and axis elevated to approximately 3 mm, which we think is a good criterion for a successful soft tissue release and a goal for good posterior reduction and decompression of spinal cord. Repeated elevation with resection of the scar tissue in front of the C1 lateral mass and around C1 anterior arch was performed; a high-speed blur was used to handle the anterior cortical bone of C1–2, after that cancellous auto-bone graft particle from iliac crest was implanted and the incision was sutured in layers [10].

After changing to prone position, posterior removal of failed instrument and nonunion bone graft prior to decompression and instrumentation was performed. Two different techniques were used based on state of dislocation and bone abnormality. For patients who had osteosynthesis

in C0-1 joints due to congenital deformities and trauma and inability to distract joint space over 3 mm, occipitocervical fusion was recommended. Otherwise, atlantoaxial fusion was performed routinely. The aforementioned two procedures were described in literature [4, 19]. The system was constrained and the polyaxial cervical screws (Depuy spine, Raynham, MA, USA) were used. We recommend obtaining a CT scan reconstruction preoperatively in every case in which C1 lateral mass screw may be inserted. During C1-pedicle screw placement, the vertebral artery and the vein plexus between C1 and C2 superiorly and inferiorly to C1 posterior arch were exposed. If the posterior arch is thick enough (in the rostral-caudal dimension) for a screw to be safely placed inferior to the vertebral artery groove, we prefer to insert the screw through the posterior arch of C1 because it allows a longer screw to be placed; if the posterior arch was too thin at the vertebral artery groove, then the screw was placed inferior to the C1 posterior arch to avoid injury to vertebral artery.

Postoperative management: the endotracheal tube and nasogastric feeding tube are maintained until the patient's tongue swelling subsides. Typically, moderate tongue swelling can be expected after surgery, but it usually subsides within the first 24–36 h. Enteric feedings continued for 1 week followed by advancing to routine diet gradually.

After surgery, all patients were maintained in a rigid cervical collar for 6 weeks or more based on incorporation of bone graft. Patients were followed up longitudinally, and their fusion statuses, neurological statuses, and clinical and radiographic outcomes were assessed. We defined bony fusion as no absorb or translucent line around graft, no instrument failure and no movement under dynamic radiograph. We defined complete reduction as AADL ≤ 3 mm. All patients were required to complete outcome questionnaires including ADL [6]; nine were by self-administration and seven by mail.

Results

The pathology in our series was some extent of dislocation of C1/2 after first surgery; of these cases, posterior instrument failure and pseudoarthrosis after first surgery was documented in 11 cases; of these, one screw fixating an occipitocervical graft loosened, but the patient was asymptomatic; anterior instrument failure and bone non-union was noted in one case, three cases experienced only decompression to the occipital magnum with no internal fixation, and one case had only anterior decompression and bony graft with no instrumentation.

Seven patients underwent fusion level at C1–C2, 4 at C0–C2, 2 at C0–C3, 1 at C0–C4, 1 at C1–C3, and 1 at

C1–C4. Auto iliac crest bone graft was performed in all 16 patients. The operative time averaged 290 min (range 210–340 min) with the average intraoperative blood loss of 700 ml (range 500–1100 ml) (Table 1).

All 16 patients were followed up for an average of 28.8 months (range 18–66 months). At final follow-up, based on lateral radiograph, CT-scan and MRI findings, 12 of 16 cases had complete reduction, and 4 had incomplete reduction. The mean cervicomedullary angle (CMA) improved from preoperative 101.8° to postoperative 143.0° with an increase of 41.2° (CMA averaged 163.9°, 135.5°–168.9° in Chinese [7]). After revision surgery, 12 cases had CMA improved to normal; however, 4 cases had CMA lower than normal due to incomplete reduction.

No patients had neurological deterioration after revision operation; all patients had JOA (17) score improved from 8.3 of before revision surgery to 12.8 at final follow-up. Nine patients were scaled excellent, five were good, and two remained with no change due to long-term severe C1/2 dislocation despite of enlargement of canal space.

No patient developed pharyngeal wall infection; however, superficial infection with respect to posterior approach was noted in one patient 2 months later which healed after intermittent debridement and 3 weeks of antibiotics. Five cases had neck pain which remained unchanged at final follow-up. One patient (C0-2 fusion) had adjacent C2/3 disc narrowing and degeneration (Case 10). No instrument failure was noted. Three patients (cases 10, 13, 14) had some extent of bone graft absorption which was evidenced radiographically within 1 year after revision surgery; however, bone union was evidenced at C1/2 lateral facet joint and around odontoid process. All patients had solid bony union both radiologically and clinically with no loss of reduction at follow-up (Figs. 2, 3).

All patients completing questionnaires reported being satisfied with the results of their revision surgery and had ADL score improved from 50 of preoperation to 73 of last follow-up. Result from ADL revealed that 11 patients (68.7%) reported mild or no pain and 5 patients (31.3%) described their pain as “discomforting” (Table 2).

Discussion

The most important aspect of surgical treatment for patients with atlantoaxial dislocation complicated with neurological deficit is to secure a sufficient sagittal canal diameter as well as stability reconstruction [8]. The presence of instrument and bony graft as well as no clear anatomic margin contribute to the high risks of injury to medulla [9], and all this lead to the irreducibility of C1/2.

We tried halo-traction prior to revision surgery in our early series of research; however, reduction of C1-2 was

Table 1 Patient demographic and clinical data

Case	First surgery		Revision surgery		Surgical protocol	Cause	Surgical protocol
	Sex	Age (y)	Diagnosis	Age (y)	Duration after first surgery (m)		
1	M	42	Malunion of odontoid fracture + AAD	45	38	BGN + AAD	ATR + PRIF (C1-2)
2	F	9	Os odontoieum + AAD	10	14	IF + BGN + AAD	ATR + PRIF (C1-2)
3	F	37	Occipitalization + AAD, Chiari deformity	40	36	BA + AAD	ATR + PRIF (C0-2)
4	F	44	Occipitalization + AAD	51	88	IF + BGN + AAD	ATR + PRIF (C0-3)
5	M	42	Occipitalization + AAD	43	13	IF + BGN + AAD	ATR + PRIF (C1-2)
6	M	18	Malunion of odontoid fracture + AAD	19	11	IF + BGN + AAD	ATR + PRIF (C1-2)
7	M	37	Os odontoieum + AAD	52	15	IF + BGN + AAD	ATR + PRIF (C0-4)
8	F	39	Occipitalization + AAD	40	37	IF + BGN + AAD	ATR + PRIF (C1-3)
9	M	27	Occipitalization + AAD, Chiari deformity	38	136	IF + BGN + AAD	ATR + PRIF (C0-3)
10	M	15	Occipitalization + AAD	17	25	AAD + BA	ATR + PRIF (C0-2)
11	M	56	Malunion of odontoid fracture + AAD	57	14	IF + BGN + AAD	ATR + PRIF (C1-2)
12	M	47	Occipitalization + AAD, Chiari deformity	50	35	AAD + BA	ATR + PRIF (C0-2)
13	F	24	Os odontoieum + AAD	37	158	IF + BGN + AAD	ATR + PRIF (C1-2)
14	F	7	Relaxation of transverse ligament of C1	9	24	IF + BGN + AAD	ATR + PRIF (C1-2)
15	F	33	Malunion of odontoid fracture + AAD	34	12	IF + BGN + AAD	ATR + PRIF (C1-4)
16	F	47	Occipitalization + AAD	48	15	IF + BGN + AAD	ATR + PRIF (C0-2)
Average		32.1		36.3	42.2		

AAD atlanto-axial dislocation, AOM atlanto occipital malformation, IF instrument failure, PD posterior decompression, BGN bony graft nonunion, ATR + PRIF anterior transoral release + posterior reduction, instrument and fusion, BA basilar invagination

Fig. 2 Case 10, male, 17-year-old, diagnosed of congenital atlanto-occipital malformation combined with basilar invagination and C1/2 dislocation. Two years after C1/2 laminectomy and enlargement of foramen magnum, severe C1/2 dislocation was noted with C2 vertebral body and odontoid process into cranial of 25 mm. A revision surgery was performed. **a** Flexion–extension lateral radiograph prior to revision surgery showed congenital atlanto-occipital malformation, basilar invagination, C1/2 dislocation and swan-neck deformity, C1/2 reduction was not available dynamically. **b** CT sagittal scan prior to revision surgery showed evidenced of C1/2 laminectomy and enlargement of foramen magnum, no bony graft and

instrumentation were evidenced. **c** MRI sagittal T2-weight showed odontoid process into cranial and compression to the ventral medullary with CMA decreasing to 76°. **d** One month after revision surgery, lateral X-ray showed reduction of C1, odontoid process out of foramen magnum and correction of deformity. **e, f** CT sagittal scan and MRI sagittal T2-weight at 1 month after revision surgery. **g** Twelve months' follow-up, lateral X-ray showed no instrument failure, no loss of reduction, evidence of bony fusion at craniocervical region and secondary C2/3 disc degeneration. **h–i** 50 months' follow-up, lateral X-ray and CT sagittal scan showed bony union and no loss of reduction

not achieved due to the presence of fibrous tissue ossification and/or failed instrument which impeded the final correction. Therefore, we obviated halo-traction and tried surgery directly.

In literature there are several approaches documenting the surgical treatment of IAAD including transoral decompression plus odontoid resection; transoral anterior atlantoaxial release, reduction and posterior internal fixation; one-stage transoral decompression and reduction by self-designed instruments. All these contribute satisfactory

outcomes for primary surgery; however only few literatures documented their efficacy for revision surgery [2, 10–13].

Despite the contracted scar tissue around the C1 lateral mass as well as bony graft and failed instrument, anatomic reduction was achieved in 12 cases (75%) following surgery. All but two patients had neurologic function improved, and it is our opinion that restoration of cervical spine alignment and improvement of vertebral artery blood supply contribute to the neurologic improvement. All

Fig. 3 Case 5, 43-year-old male, diagnosed of congenital atlanto-occipital malformation complicated with C1/2 dislocation, 1 year after atlanto-occipital fusion and pedicle screw instrumentation, instrument failure and C1 dislocation developed. A revision surgery was performed. **a** Internal instrument failure, bony graft absorbance and irreducible C1/2 dislocation is evidenced. **b** One month after

revision surgery, lateral X-ray showed reduction of C1/2 and no signs of instrument failure. **c** CT sagittal scan at one month showed ADI of 2 mm, canal diameter at C1/2 of 22 mm. **d** Lateral X-ray of 24 months showed bony fusion, no loose of C1/2 reduction with ADI of 2 mm and no signs of instrument failure

patients completing questionnaires reported being satisfied with their results.

Based on our clinical experience, anterior release is of key importance for the RSAAD. Under the condition of no osteosynthesis around C1/2 facet joints, complete C1/2 reduction can be reached by our method. We defined criteria of successful transoral release as elevation of C1/2 bilateral lateral mass joint space by 3 mm width which is sufficient for reduction via posterior approach. That means if C1/2 bilateral lateral mass joint space can be elevated by 3 mm width, most factors from vertebral anterior aspect which impede C1/2 reduction are removed. During this process, intraoperative spinal cord monitor must be used to guide the maneuver.

This sequential reduction provides advantages for the treatment of the RS-IAAD compared with traditional decompression: (1) The anterior release is a less invasive procedure which obviates the need to dissect the odontoid process. Of the 12 cases achieving anatomic reduction, none was complicated with dura matter tearing or spinal cord injury. (2) This method obviates the procedure of removing posterior failed instrument first, while direct anterior release followed by posterior reduction and instrumentation is indicated. (3) No need for over removal of tissue, while only to the extent of elevation of C1/2

bilateral lateral mass joint space by 3 mm width. (4) The reduction is reached sequentially by instruments' pull strength. (5) Realignment of the atlantoaxial joint can prevent the subaxial spine degeneration due to C1/2 long time dislocation.

Several instrumentations have been described in the literature [14]. Anterior instruments placed on the pharyngeal wall predispose the risk of local infection; as a result, anterior screw loosening has been reported [11]. While for posterior instrument, after removal of scar tissue and decompression, there is not enough space for sublaminar wire instrumentation. Despite the good biomechanical characteristics provided by C1-2 transarticular screws described by Magerl and Seeman, intraoperative reduction is incapable of achieving them.

Posterior plate-screw system has the advantages of intraoperative reduction and not relying on the integrity of C1/2 posterior structure. They can provide adequate immediate stabilization. In our early series of studies, we proposed a modification to the technique of Goel et al. [15–17]: we preferred to insert the screw into lateral mass via the posterior arch rather than below it. Currier et al. [18], in his article, mentioned this as technique of Tan et al. and suggest that if the posterior arch is thick enough for a screw to be safely placed inferior to the vertebral artery

Table 2 Radiological and clinical outcome data

Case	Before revision surgery			Follow-up (m)	After revision surgery					
	JOA (17)	ADL	CMA (°)		JOA (17)	ADL	CMA (°)	Neurological improvement	Reduction on radiography	Complication
1	4	35	100	30	4	40	155	Poor	Complete	Nasal hponation
2	8	45	106	28	12	60	128	Good	Complete	No
3	10	55	100	24	15	75	138	Excellent	Complete	No
4	12	60	115	66	16	85	135	Excellent	Complete	Occipitocervical pain
5	12	60	95	24	15	80	149	Excellent	Complete	Occipitocervical pain
6	10	55	110	35	16	85	150	Excellent	Complete	No
7	6	40	98	18	7	45	150	Poor	Partial	Screw loosen
8	11	50	110	28	16	80	140	Excellent	Partial	No
9	8	50	100	25	10	55	135	Good	Partial	No
10	8	50	76	50	15	85	148	Excellent	Complete	Adjacent degeneration
11	11	55	106	26	15	80	146	Excellent	Partial	No
12	13	60	105	21	16	80	150	Excellent	Complete	Occipitocervical pain
13	7	45	99	33	9	80	142	Good	Complete	Deep infection
14	12	55	110	39	16	80	145	Excellent	Complete	No
15	9	45	105	24	14	85	137	Good	Partial	No
16	9	45	100	28	13	80	140	Good	Complete	Occipitocervical pain
Average	9.4	50	101.9	28.8	13.0	73	143			

RS revision surgery, CMA cervical medular angle, ADL activity daily living

※ Neurologic Improvement: according to Hirabayashik's Grade Definition (JOA)

groove, then our technique is ideal because it allows a longer screw to be placed and the bleeding is markedly reduced. In the same year, Resnick et al. [19] reported the similar technique. Another advantage is short-segment fixation. We extended the range of fusion to occiput only when osteosynthesis or destruction of atlanto-occipital joint is evidenced; otherwise, we can preserve the ROM of cervical spine as much as possible.

One patient (case 10) developed C2/3 disc degeneration probably due to the change of viscoelasticity of C2/3 disc after reduction, and two patients (cases 1, 7) showed poor clinical outcome owing to their long-term medulla compression.

Limitations of the study

There are several limitations to the present study. Chief among these is still the relative paucity of patients (of revision surgery) collected and relatively short-term follow-up is documented. This is mainly due to the paucity of patients necessitating revision surgery at craniocervical region.

References

1. Crockard HA, Johnston F (1993) Development of transoral approaches to lesions of the skull base and craniocervical junction. *Neurosurg Q* 3:61–82
2. Hadley MN, Spetzler RF, Sonntag VKH (1989) The transoral approach to the superior cervical spine: a review of 53 cases of extradural cervicomedullary compression. *J Neurosurg* 71:16–23
3. Behari S, Banerji D, Bhargava V et al (1999) Transoral decompression for craniovertebral osseous anomalies: perioperative management dilemmas. *Neurol Soc India* 47:188–195
4. Zileli M, Cagli S (2002) Combined anterior and posterior approach for managing basilar invagination associated with type I Chiari malformation. *J Spinal Disord Tech* 15:284–289
5. Ono K, Ebara S, Fuji T et al. (1987) Myelopathy hand. New clinical signs of cervical cord damage. *J Bone Joint Surg Br* 69(2):215–219
6. Granger CV, Albrecht GL, Hamilton BB (1979) Outcome of comprehensive medical rehabilitation: measurement by PULSES profile and the Barthel Index. *Arch Phys Med Rehabil* 60(4):145–154
7. Desheng W, Liming Z, Shuron D (1993) Measurement of normal value of brain stem-spinal cord angle and clinical significance. *J Harbin Med Univ* 1(1):13–17
8. Grob D (1998) Surgery in the degenerative cervical spine. *Spine* 23:2674–2683

9. Kerschbaumer F, Kandziora F, Ewald W et al (1998) Staged therapy for atlanto-axial instability in rheumatoid arthritis. *J Bone Joint Surg Br* 80:S244–S245
10. Wang C, Yan M, Zhou HT (2006) Open reduction of irreducible atlantoaxial dislocation by transoral anterior atlantoaxial release and posterior internal fixation. *Spine* 31(11):306–313
11. Yin Q, Ai F, Zhang K (2005) Irreducible anterior atlantoaxial dislocation: one-stage treatment with a transoral atlantoaxial reduction plate fixation and fusion. Report of 5 cases and review of literature. *Spine* 30(13):375–381
12. Govender S, Kumar KPS (2002) Staged reduction and stabilization in chronic atlantoaxial rotatory fixation. *J Bone Joint Surg Br* 84:727–731
13. Kerschbaumer F, Kandziora F, Klein C et al (2000) Transoral decompression, anterior plate fixation, and posterior wire fusion for irreducible atlantoaxial kyphosis in rheumatoid arthritis. *Spine* 25:2708–2715
14. Henriques T, Cunningham BW, Olerud C et al (2000) Biomechanical comparison of five different atlantoaxial posterior fixation techniques. *Spine* 25:2877–2883
15. Goel A, Desai KI, Muzumdar DP (2002) Atlantoaxial fixation using plate and screw method: a report of 160 treated patients. *Neurosurgery* 51:1351–1357
16. Harms J, Melcher RP (2001) Posterior C1–C2 fusion with polyaxial screw and rod fixation. *Spine* 26:2467–2471
17. Melcher RP, Puttlitz CM, Kleinstueck FS et al (2002) Biomechanical testing of posterior atlantoaxial fixation techniques. *Spine* 27:2435–2440
18. Currier BL, Yaszemski M (2004) The use of C1 lateral mass fixation in the cervical spine. *Curr Opin Orthoped* 15:184–191
19. Resnick DK, Benzel EC (2002) C1–C2 pedicle screw fixation with rigid cantilever beam construct: case report and technical note. *Neurosurgery* 50:426–428