Markus Weißkopf
Detlef Naeve
Michael Ruf
Jürgen Harms
Dezsö Jeszzenszky

Therapeutic options and results following fixed atlantoaxial rotatory dislocations

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D. Naeve · M. Ruf · J. Harms
Department of Orthopedic
and Spine Surgery,
SRH-Klinikum Karlsbad-Langensteinbach,
Karlsbad, Germany

M. Weißkopf (✉)
Department of Orthopedic Surgery,
University Hospital of the RWTH Aachen,
Pauwelsstr. 30, 52074 Aachen, Germany
Tel.: +49-241-8089410,
Fax: +49-241-8082530,
e-mail:
MWeisskopf@orthopaedie-aachen.de

D. Jeszzenszky
Department of Orthopedic Surgery,
Kantonsspital St. Gallen,
St. Gallen, Switzerland

Abstract Atlantoaxial rotatory dislocation (AARD) represents a rare pathological condition of the upper cervical spine that is frequently misdiagnosed, leading to a delay in therapy. In a long-term assessment of clinical and radiological results, three different therapeutic options with regard to the length of the dislocation-therapy interval (DTI) were evaluated. Twenty-six patients were treated for AARD from December 1988 until April 2000. Proper diagnosis was established after an average interval of 15 months. Three different therapeutical protocols were followed in order to reduce the dislocation: (1) closed transoral reposition under general anesthesia; (2) temporary transoral fixation utilizing the Harms T-plate; (3) definitive transoral fusion. The eight patients treated by closed reduction had the best pain relief. The average visual analogue scale (VAS) score was 96.6 points,

while the rotatory motion of the upper cervical spine, as assessed by dynamic MRI, was 25.3° to each side. The length of the dislocation-therapy-interval (DTI) averaged 1.4 months. A mean VAS Score of 92.3 points was recorded in the ten patients treated with a temporary fixation of C1/C2. In this subgroup the DTI had an average length of 5.3 months. The mean rotation to each side was 13.9°. In the eight patients who underwent definitive fusion the mean VAS score was 60.6 points, while the average length of the DTI was 40.5 months. In conclusion, the clinical outcome and the subjective well-being following AARD deteriorates with increasing length of the dislocation-therapy interval.

Keywords Atlantoaxial dislocation · Transoral approach · Temporary fixation

Introduction

Fixed atlantoaxial rotatory dislocation (AARD) represents a rare entity of upper cervical spine (C-spine) pathology [11, 16, 21, 24, 26, 27] (Table 1). Frequently, there is a considerable delay in establishing the proper diagnosis (weeks or months after the initial dislocation), since the pathological changes depicted on the conventional radiographs are only discrete. Additionally, this entity is rarely considered in the routine diagnostic work-up of torticollis [1, 7, 8, 22, 23, 31].

Conservative treatment is the first step in the treatment of fixed AARD. Frequently analgesics, halter traction or closed reduction maneuvers will successfully correct the dislocation. (Table 1). Surgical reduction is indicated in the following cases: when conservative measures fail to reduce fixed AARD; in the case of re-dislocation; in highly unstable injuries; and in patients not demonstrating appropriate compliance [2, 28]. The aim of this study was to introduce various therapeutic strategies and to assess their value over the long term. The results are presented according to both the persistence of motion and the patients' subjective well-being. The influence of the disloca-

Table 1 Review of the AARD literature

Author	Summary
Boos et al. 1997 [4]	Case report of a fighter pilot rescued by ejection seat, who sustained an atlanto-axial dislocation with disruption of the transverse ligament of the axis. Surgical treatment consisted of a transarticular arthrodesis with Gallie fusion
El Khoury et al. 1984 [6]	Report of AARD in three infants who were treated conservatively
Engelhardt et al. 1995 [7]	Report of three infants with delayed diagnosis of AARD. Conservative treatment consisted of immobilization for 4–8 weeks
Fielding and Hawkins 1977 [8]	Original article and classification of fixed AARD. Delayed diagnosis in 15 of 17 patients. Treatment included skull traction in all patients. In 13 patients additional surgical fusion was necessary
Hicazi et al. 2002 [12]	Cross-sectional clinical and radiologic study with a control group. Twenty-three out of 33 patients with acute torticollis were investigated by dynamic CT. All patients had atlantoaxial subluxation without fixation. Conservative treatment was successful in all cases
Moore and Frank 1995 [21]	Case report of a traumatic AARD featuring a bilateral facet joint fracture. Therapy included closed reduction and surgical stabilization
Phillips and Hensinger 1989 [23]	Article reviewing therapy of AARD in 23 children. Conservative treatment was performed in 20 patients. Open reduction and posterior stabilization was necessary in three cases. Success of closed reduction depended on the duration of the dislocation-therapy interval
Robertson and Swan 1992 [24]	Case report of a rugby player, who sustained a traumatic AARD with initial tetraparesis. Therapeutic regimen consisted of closed reduction that led to complete remission of the neurologic deficits
Schwarz 1995 [26]	Case studies of two infants presenting with torticollis, who had a delayed diagnosis because of insufficient radiological work-up. Both cases were treated with closed reduction
Subach et al. 1998 [27]	Retrospective review of 20 children, seen during a 7-year period, who had an atlanto-axial subluxation. Conservative management was successful in 14 cases. Posterior fusion was necessary in six patients because of unsatisfactory reduction or recurrence. Success of conservative treatment depended on the duration of the subluxation
Vinchon et al. 1995 [28]	Case report of a patient with atlanto-axial instability that lead to neurological deficits caused by vertebrobasilar insufficiency. After posterior surgical stabilization, no more symptoms were reported

tion-therapy interval (DTI) on the treatment strategy is to be evaluated.

Review of the literature

Several causes leading to fixed AARD have been reported. Traumatic dislocation of the upper C-spine [2], inflammation of the nasopharynx [9], and postoperative dislocation generated by ear, nose, and throat (ENT) procedures [27] may be preceding events. Also, congenital alterations leading to ligamentous laxity, as encountered in Down's syndrome or juvenile rheumatoid arthritis, may predispose to AARD. Many authors believe that the higher incidence of AARD in children is the consequence of the specific anatom-

ical features in this age group. The joint surface of the lateral mass is shallower and more horizontally oriented. In addition, the relative elasticity of the ligaments, the not yet fully developed neck muscles, and the relatively large head might be predisposing factors for AARD [16, 27, 29].

In a physiological rotation of the atlantoaxial joint complex, the odontoid process represents the center of rotation. With increasing rotation, a reversible subluxation in the C1/C2 joint (massae laterales) occurs. This complex movement, featuring a combination of translation and rotation movement, is restricted with regard to extreme motions by the transverse atlantal ligament and the alar ligaments [8, 31].

In fixed AARD, the physiological reduction or return to the neutral position following a rotational movement is

not achieved [6]. Causes for the failed reduction might include mechanical obstacles, such as an interposition of synovia. Another possible reason is extreme rotational motions that traumatize the ligamentous structures. In the classification of atlantoaxial malalignment, translational dislocations are differentiated from rotational dislocations [2, 8]. Fixed AARD has to be discerned from acute torticollis, in which motion can be demonstrated between C1 and C2 [12].

Because of the rare incidence and the discrete pathological changes on conventional X-rays, the final diagnosis of AARD is frequently delayed [7, 8, 19, 22, 23]. The exact anterior-posterior view on plain radiographs will always illustrate an eccentric position of the odontoid process. In addition, the rotational dislocation of the atlas leads to an elliptical appearance of the arch on the lateral view.

Besides the eccentric alignment of the odontoid process, a slanted position of the articulation is seen on the CT scans, which is impressively demonstrated on the sagittal and frontal 2D, as well as 3D reconstructions [26]. For the precise definition of a fixed rotational dislocation, functional CT studies as described by Dvorák are recommended [5]. In a dynamic investigation the head is rotated into the maximum right and left positions. An exact parallel adjustment of the gantry with respect to the articulation

of C1/C2 is obligatory. In the case of a fixed AARD, there will be no change in the rotational relationship between C1 and C2. Magnetic resonance imaging (MRI) is of special importance in the presentation of neural structures, whereas its utilization in the evaluation of ligamentous structures is not yet fully elucidated [3, 4, 30, 32].

The persistence of AARD leads to degeneration of the atlantoaxial joint complex [27]. Because of potential sequelae, such as severe headache or facial asymmetry in infants, it is mandatory to establish the proper diagnosis of this rare entity as early as possible, in order to initiate adequate therapy. It must also be considered that an additional minor trauma to a persisting dislocation might cause a severe injury of the C-spine with neurological complications [8].

Methods

From 1988 through 2000, 26 patients (18 females, 8 males) with an average age at time of reduction of 12.3 years (range: 4–38 years, SD: ± 8.66 years) were treated for AARD at our institution. The cause of the AARD was trauma in 16 cases; an inflammation of nasopharynx in five cases, previous ENT surgery in two cases, spontaneous dislocation in two instances, and one case of congenital malformation was noted (Table 2).

Table 2 Survey of patient data (URT upper respiratory tract, TO transoral, M male, F female, ENT ear, nose, and throat)

Patient	Age (years)	Sex	History	DTI (months)	Treatment	VAS pre-treatment	VAS post-treatment
1	38	M	Trauma	39	TO reduction and definitive fusion	44	42
2	36	M	Trauma	42	Posterior reduction and definitive fusion C0–C3	17	19
3	15	F	Congenital malformation	180	TO reduction and definitive fusion	82	80
4	13	F	URT Inflammation	10	TO reduction and definitive fusion	10	52
5	8	F	Trauma	9	TO reduction and definitive fusion	46	97
6	21	F	Trauma	17	TO reduction and definitive fusion	9	42
7	9	M	Trauma	9	TO reduction and definitive fusion	45	79
8	11	M	Iatrogenic post ENT surgery	18	TO reduction and definitive fusion	68	74
9	12	M	Trauma	3	TO reduction and temporary fixation	1	86
10	7	F	Trauma	6	TO reduction and temporary fixation	68	98
11	6	F	URT inflammation	6	TO reduction and temporary fixation	47	100
12	26	M	Trauma	2	Posterior reduction and temporary fixation	29	84
13	7	F	URT inflammation	3	TO reduction and temporary fixation	38	96
14	7	F	URT inflammation	16	TO reduction and temporary fixation	46	95
15	9	F	Trauma	8	TO reduction and temporary fixation	65	66
16	8	M	URT inflammation	4	TO reduction and temporary fixation	7	100
17	9	F	Trauma	4	TO reduction and temporary fixation	45	98
18	23	F	Trauma	1	TO reduction and temporary fixation	46	100
19	10	F	Trauma	3	TO closed reduction	53	85
20	4	F	Spontaneous	0.04	Spontaneous reduction	60	100
21	5	M	Trauma	0.1	Closed reduction	29	100
22	6	F	Trauma	1	TO closed reduction	86	100
23	8	F	Spontaneous	4	TO closed reduction	49	100
24	8	F	Iatrogenic post ENT surgery	1	Closed reduction	69	100
25	9	F	Trauma	1.5	TO closed reduction	10	91
26	5	F	Trauma	0.5	TO closed reduction	47	97

Fig. 1 Sketch of transoral closed according to Jeszenszky. The image illustrates that the spinous process of the axis is locked by one hand, while the index finger of the other hand presses in counter rotation to the dislocation on the lateral mass of the atlas through the posterior wall of the pharynx

According to the Fielding and Hawkins classification:

- Six patients had a type I dislocation (rotation and anterior displacement of the atlas less than 3 mm)
- Seventeen patients suffered a type II dislocation (rotation and anterior displacement of the atlas of 3–5 mm)
- Three cases showed a type III dislocation (rotation and anterior displacement of the atlas more than 5 mm)

The rotation angle of the dislocation averaged 29° (range: $9\text{--}46^\circ$, SD: ± 10.84). In 14 cases the atlas was rotated in relation to the axis to the right, in 12 cases to the left. The correct diagnosis was established on average 15 months after the onset of symptoms (range: 1 day to 15 years, SD: ± 35.35 months). Three different therapeutic strategies were followed:

- Closed reduction was attempted in all cases. If necessary the transoral method according to Jeszenszky [14] was performed (Fig. 1)
- If open reduction was necessary, temporary transoral fixation utilizing a Harms T-plate was the preferred surgical treatment
- In cases where degenerative changes or incongruence of the atlantoaxial joint were found on the preoperative CT scans, or in cases where severe lesions of the joint surface were recognized during surgery, definitive fusion was performed

All but two of the open reduction maneuvers were performed via the modified transoral approach by Schmelzle and Harms [25]. In order to reduce fixed AARD, one case was treated with a posterior temporary C1/C2 fixation, while another patient underwent a posterior fusion from C0–C3.

After closed reduction patients were immobilized in a halo body jacket for 8–12 weeks. Implants were removed after 3 months

in patients who underwent temporary fixation. As part of the routine clinical and radiological follow-up, the patients were examined after 3 months and 6 months, as well as 1 year post-reduction. The last follow-up was carried out on average after 48.1 months (range: 25–133 months, SD: ± 25.98). Clinical function of the cervical spine was assessed for the major directions of motion, i.e., flexion/extension, rotation, and side-bending. In addition, the rotation of the C-spine was assessed both in flexion and extension. The atlantoaxial rotation was determined by MRI, during which scans were obtained in a neutral position, maximum right rotation and maximum left rotation. Axial slices of 3 mm thickness of C1 and C2 were attained on a Magnetom scanner (Magnetom Expert, 1.0 T, spin-echo sequence, T1: ($T_R/T_E=700/15$) and T2: ($T_R/T_E=2000/90$)).

For the assessment of degenerative alterations of the atlantoaxial articulation, plain radiographs of the cervical spine in two planes plus a transoral view were obtained. The classification of Kellgren [17] was modified in order to differentiate between four grades of degeneration (Table 3).

The complaints of the patients were evaluated both before and after reduction, utilizing the visual analog score (VAS) for spinal trauma from the Hannover Medical School [18]. The patients were requested to assess their personal well-being themselves. A maximum total score of 100 could be achieved if the patient judged himself to be completely pain-free without loss of function.

The influence of the therapeutic option on both the self-reported VAS and the atlantoaxial motion, as assessed by MRI, was evaluated for significant difference utilizing the Bonferroni *t*-test (Version 8.2; SAS Institute, Cary, NC, USA) by a statistician not involved with data acquisition and blinded to patients' identity. Results were calculated in two-sided simultaneous confidence intervals. Since two different analyses of variance were applied, a Bonferroni adjustment of significance level had to be performed.

Results

Closed reduction was performed in eight patients. On the last follow-up no re-dislocation was seen. Five of these patients were completely pain-free. The personal assessment of patients' comfort yielded on the VAS an increase from an average 50.4 points (range: 10–86 points, SD: ± 23.4) to 96.6 points (range: 85–100 points, SD: ± 5.7). Degenerative alterations could not be detected in this group. The rotational range of motion (ROM), assessed by dynamic MRI, was 25.3° to each side (range: $7.5\text{--}41^\circ$, SD: ± 12.2). The ROM on the physical examination of the whole C-spine did not show any loss of motion (average rotational movement to each side: 72.5° ; range: $45\text{--}85^\circ$, SD: ± 12.5) (Fig. 3). The average length of the DTI was 1.4 months (range: 0.04–3 months, SD: ± 1.4).

A temporary stabilization of C1/C2 was carried out in ten patients. On follow-up examination, three patients presented without any complaints. The patients of this subgroup demonstrated an improvement of the VAS scores from an average 39.2 points (range: 1–65 points, SD: ± 21.8)

Table 3 Modified classification for C-spine degeneration after Kellgren

Grade	None (1)	Mild (2)	Moderate (3)	Severe (4)
Radio-morphologic alterations	No degenerative changes	Narrowing of disc space	Narrowing of disc space + subchondral bone sclerosis	Narrowing of disc space + subchondral bone sclerosis + osteophytes

preoperatively to 92.3 points (range: 66–100 points, SD: ± 10.9) postoperatively. There was no statistically significant difference in the postoperative VAS score compared with the “closed reduction” subgroup [-28.701 ; $+20.051$]. Radiological signs of beginning degeneration (grade 1) were seen in two cases. In one patient a diminution of the C2/C3 disc space was recognized, while another patient presented degenerative changes in the right lateral mass of the atlantoaxial joint. Rotational atlantoaxial joint movement could be demonstrated by dynamic MRI in six patients (23.3° , on average, to each side). In four patients the atlantoaxial joint fused spontaneously. One out of these four patients had a malrotation of 17° to the right. Prior to the open reduction maneuver this patient presented with a right rotational dislocation of 36° . The rotational ROM of the C1/C2 joint for the complete subgroup scored 13.9° (range: 0° – 35° , SD: ± 12.9). Again, no statistically significant difference was found in comparison with the “closed reduction” subgroup [-28.227 ; $+2.502$]. Physical examination did not reveal any loss in the rotational ROM of the C-spine (average movement to each side: 70.5° ; range: 48° – 90° , SD: ± 12.4). In this subgroup the average dislocation-therapy interval was 5.3 months (range: 1–16 months, SD: ± 4.3).

All patients ($n=8$) of the definitive fusion group demonstrated a solid fusion on their last clinical and radiological evaluation. Minor complaints were reported by one individual, while the remaining seven patients suffered from neck pain. In this group an improvement of the VAS score was also noted. The score increased from an average of 40.1 points (range: 10–82 points, SD: ± 26.8) to 60.6 points (range: 19–97 points, SD: ± 25.9). The postoperative VAS score was significantly worse statistically in comparison with the “closed reduction” and “temporary fusion” subgroups – [-61.694 ; -10.306] and [-56.051 ; -7.299], respectively. In the neighboring C2/C3 segment radiological signs of degeneration were reported in three of eight patients. Of these three cases, two showed mild (grade 1) and one moderate alterations (grade 2). In this subgroup, all patients showed a loss in ROM (average motion to each side: 40.9° ; range 15° – 60° , SD: ± 14.1) during the clinical examination (Fig. 3). A reduction in flexion/extension motion was noted in three patients. The average dislocation-therapy interval for the definitive fusion group was documented with an average delay of treatment of 40.5 months (range: 9–180 months, SD: ± 57.86).

Discussion

Unfavorable long-term sequelae of untreated fixed AARD necessitate early intervention in order to regain physiologic mobility of the upper C-spine. Advances in imaging techniques have optimized the diagnostic work-up of this rare condition [5]. When fixation between C1 and C2 is demonstrated, the success rate of conservative treatment

decreases in proportion to the length of the DTI. In the initial phase most of the pediatric patients will respond to immobilization and analgesics as conservative measures [12]. In cases of persisting AARD, i.e., 1 week after establishing the diagnosis, closed reduction utilizing halter traction can be attempted [27]. Since prolonged traction might interfere with children’s compliance, we prefer a closed reduction under general anesthesia, if necessary by means of the transoral method after Jeszenszky [14]. With this maneuver we could reduce a fixed AARD with a DTI up to 4 months. Closed reduction was followed by an immobilization period with a halo body jacket for 8–12 weeks. In all these patients persistence of motion of the atlantoaxial joint could be demonstrated on dynamic MRI studies during final follow-up. No spontaneous fusion occurred among these patients.

In the remaining 18 patients a closed reduction was not possible or could not be maintained. In these cases an open reduction was performed. Depending on the intraoperative situation of the atlantoaxial joint, either a temporary stabilization or a definitive fusion was then carried out.

While definitive fusion is a safe procedure to prevent re-dislocations, it inevitably leads to a loss of function in the main rotational segment of the C-spine. Arthrodesis of the atlantoaxial joint will theoretically result in a loss of rotation of 40° to each side [5]. Younger patients, in particular, can easily compensate for this loss of rotation in the lower C-spine segments. A review of the literature with regard to long-term results following such a fusion reveals an accelerated degeneration in the adjacent segments in approximately 30% of cases after 10 years [13]. Finite element studies of the C-spine simulating a fusion demonstrated an increased compressive force during flexion and side-bending [20].

In consideration of this background data, our primary option was temporary fixation of C1/C2 utilizing the Harms T-plate via a transoral approach in cases where open reduction was indicated [25]. Six out of the ten patients treated with temporary stabilization showed motion of the atlantoaxial articulation after removal of the plate. Four patients demonstrated no rotational motion in the MRI during their last follow-up. The reason for the spontaneous fusion could be increased scarring in the prevertebral area after two surgical approaches.

As an example the patient with the longest follow up after temporary fixation presented with a free ROM on the physical examination (Fig. 2). The respective dynamic MRI showed 28° rotation to the left and 35° to the right. The degenerative changes of the lower adjacent segment that was recognized on the final radiological follow-up could be a consequence of surgical therapy. Also, the disc-space narrowing of C2/C3 might be a late sequel of the trauma to the upper C-spine that resulted in fixed AARD and posterior arch fracture. Another patient out of this group was reduced and stabilized via a posterior ap-

Fig. 2 Lateral X-ray **a** and CT scan **b** of the cervical spine of a 22-year-old woman who sustained an upper cervical spine injury, showing a fixed AARD measuring 45° in addition to a posterior arch fracture of atlas. Diagnosis was established 1 week after trauma. Initial treatment consisted of closed reduction and stabilization in halo fixation. Failed reduction led to transfer to our spinal center. Since closed reduction was no longer feasible, an open reduction using the Harms T-plate for temporary fixation was performed 1 month after the accident. Postoperative X-rays (**c**, **d**). The plate was removed 3 months later. Follow-up examination after 12 years revealed an asymptomatic patient (VAS: 100). Beginning degenerative signs were seen in the C2/C3 segment (**e**, **f**). The dynamic MRI revealed good atlantoaxial rotation of 35° to the right and 28° to the left (**g**)

Fig. 3 Range of motion (ROM) of the cervical spine during final follow-up physical examination following different treatment forms. The letters on the left side of the bars indicate the direction of AARD

proach utilizing lateral mass screws in C1/2. This approach was recently introduced by Harms [10]. Further studies will have to show if a posterior stabilization might reduce the spontaneous fusion rate. Although four patients of this subgroup developed a spontaneous fusion, the overall VAS results showed a marked long-term improvement. Also, the averaged ROM of the six patients who showed motion on the MRI was comparable to the averaged ROM of the closed-reduction subgroup (Fig. 3). An explanation for this outcome may be the relatively young mean age of these patients (10.3 years) and, in comparison with the definitively fused subgroup, the shorter average DTI (5.3 months vs 40.5 months) of the temporarily fused patient.

As part of the radiological work-up of fixed AARD, dynamic CT scans as described by Dvorak, as well as plain radiographs and axial CT cuts were carried out in order to exactly define the true extent of the fixed dislocation. Radiation exposure during the CT examination represents a major disadvantage, especially in the evaluation of children. Here, the MRI offers radiation-free images of the vertebral bodies, although the picture quality of the osseous structures is reduced in comparison to CT scans [15]. In our patients, the dynamic MRI was carried out in 22 of 26 patients. Prior to the evaluation of patients with AARD, we validated this procedure in two healthy individuals. We believe that MRI can be a valuable diagnostic tool that can be utilized in follow-up examination of patients with AARD.

Conclusion

Our results demonstrate that closed reduction with temporary external stabilization is superior to the open reduction maneuvers. However, early establishment of the proper diagnosis is of utmost importance in order to successfully perform such a closed reduction. If open reduction becomes necessary, temporary stabilization offers similar results to those of closed reduction. Longer DTIs frequently necessitate definitive fusion, leading to a significantly worse clinical outcome.

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