ORIGINAL ARTICLE ASIAN JOURNAL OF MEDICAL SCIENCES

Clinicoradiological evaluation and outcome assessment of single-stage posterior correction of atlantoaxial dislocation

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Submission: 28-03-2023 Revision: 27-06-2023 Publication: 01-08-2023

ABSTRACT

Background: Atlantoaxial dislocation (AAD) refers to loss of stability between atlas and axis vertebrae, resulting in loss of normal articulation. It may be due to congenital, traumatic, or inflammatory reasons. Various techniques of atlantoaxial fixation have been described and used in patients over the years, but each has its own merits and demerits. **Aims and Objectives:** The aim of the study was to study clinicoradiological evaluation and outcome assessment of single-stage posterior correction of AAD. **Materials and Methods:** This is a prospective study done in the Department of Neurosurgery, JA group of hospitals, Gajra Raja Medical College, Gwalior, from July 2021 to July 2022. The study was conducted on 20 randomly selected patients whose presentation and imaging showed indication for surgery. **Results:** C1 lateral mass C2 pars screw fixation had one Frankel grade improvement in 6 months in 96% patient with a fusion rate of 100% and the patient also has modified Japanese orthopedic association score improvement of 15.25 ± 0.98. **Conclusion:** This study reviewed single-stage posterior correction of AAD using C1 lateral mass and C2 pedicle screw and rod fixation; it enables the stabilization of atlantoaxial motion and preserves motion at occipito-atlantal level with high fusion rates, good neurological recovery, and minimal complications.

Key words: Atlantoaxial dislocation; Atlantoaxial subluxation; Comprehensive review; Classification; Diagnosis; Treatment

Access this article online

Website:

http://nepjol.info/index.php/AJMS

DOI: 10.3126/ajms.v14i8.53643

E-ISSN: 2091-0576

P-ISSN: 2467-9100

INTRODUCTION

Atlantoaxial dislocation (AAD) is an uncommon condition. AAD results due to loss of stability between the atlas and axis (C1–C2), resulting in loss of normal articulation. The Atlantoaxial joints can lose stability due to traumatic, inflammatory, idiopathic, or congenital abnormalities.[1] Although it occurs in all age Groups, AAD is most often seen in adolescents and young adults.

AADs have been extensively studied and reported in the literature with subsequent treatment recommendations. Neurologic symptoms occur when the spinal cord or adjacent nerve roots are involved. AAD may occur as a result of abnormalities or trauma associated with the C1-C2 articulation, causing excessive movement of this joint. This includes the articulation between the anterior arch of C1 and the odontoid process of C2, as well as the facet joints of posterior elements. The following three patterns are noted:

- Flexion-extension
- Distraction
- Rotation.

The most common abnormalities involve the transverse ligament or odontoid process.[1] Transverse ligament and the facet capsules maintain the integrity of the atlantoaxial articulation. The transverse ligament is the

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primary restraint against anterior translation of the C1 on C2, whereas the odontoid is the primary restraint against posterior translation. Symptomatic AAD occurs when subluxation or dislocation causes the odontoid process, or posterior arch of the atlas, to impinge on the spinal cord causing neurologic manifestations. In addition, motion of the C1-C2 segment can also cause the compression of adjacent or exiting nerve roots. There is no evidence that individuals with asymptomatic AAD are at higher risk for the development of symptomatic AAD.

In addition, children appear to be more susceptible to AAD secondary to their steeper dens-facet angle and rich vascular folds in the atlanto-axial joint.

AAD was initially classified by Greenberg[2] into two subcategories – reducible and irreducible. Greenberg further devised a treatment strategy based on this system. Wang has recently proposed a novel classification system that aims to standardize AAD classification and its treatment strategy. Referred to as the "Wang classification system,"[3] it draws from Greenberg's system and is primarily based on classifying dislocations as reducible or irreducible AAD. According to this system, which includes preoperative evaluation using dynamic radiographs, reconstructive CT Scan, and skeletal traction test, the Wang classification categorizes AAD into four types: instability (type I), reducible dislocation (type II), irreducible dislocation (type III), and bony dislocations (type IV). This novel classification system offers a better diagnosis and treatment protocol for clinicians suspecting AAD in a patient.

Various techniques of atlantoaxial fixation have been described and used for the treatment of patients with atlantoaxial instability, but each has its advantage and disadvantages.

There are several surgical techniques to manage C1-C2 instability. Since the first description of the sublaminar wiring technique by Gallie[4] in 1939, various modifications of the techniques had been suggested by Goel and Laheri.[5] Unfortunately, studies have shown that the non-union rates for these techniques are as high as 80% (range 3–80%). Although these techniques had been developed and modified to achieve better result of the stabilization of atlantoaxial complex, the operations are still challenging due to various complications and technical difficulties.

These unsatisfactory outcomes led to the development of newer techniques for C1-C2 fusion instrumentation including the use of C1-C2 transarticular screws.[6] This technique requires pre-instrumentation reduction of any subluxation. In 1994, Goel and Laheri reported the use of plates and C1 lateral mass and C2 pars interarticularis screws.[7] In 2001, Harms and Melcher further popularized the technique of posterior C1-C2 fusion with C1 lateral mass screw and C2 pedicle screw.[8] Although this technique is technically demanding, it has been shown to give superior biomechanical and clinical results. The advantages of this new technique are safer trajectory of screws, potential for post-instrumentation reduction, and avoidance of damage to the C1-C2 facet joint. Posterior screw fixation utilizing C1-C2 transarticular screws and C1 lateral mass screws with C2 pars screws are the final two alternative methods of posterior C1-C2 fixation. The C1 lateral mass screw with C2 pedicle screw construct was initially created with plates and screws by Goel et al.,[5] in the 1980s. The method has recently gained popularity, and a variety of instrumentation is now available for application with this new technique.

Aims and objectives

- To study various clinical and radiological aspect of congenital AAD
- To study role of single-stage posterior correction in congenital AAD.

MATERIALS AND METHODS

Duration of study

- This study was conducted for 1 year

Design of study

- The study design was a prospective study.

Place of study

- Department of Neurosurgery, Jayarogya and Associated hospital, GRMC, Gwalior. A tertiary care center.

Congenital AAD

- Defined as atlantodental interval >5 mm in children and >3 mm in adults.

A consecutive series of patients with AAD admitted in the Department of neurosurgery, Jayarogya and Associated Hospital, GRMC, Gwalior from July 2021 to July 2022 were surgically treated by either C1 lateral mass C2 pars-interarticularis screw-rod fixation or Occipito-C2 wiring technique. All aspects of our research were assessed and approved by Research Ethics Committee. Dislocation or reduction was assessed before surgery, immediately after surgery, and at the final follow-up with both X-ray and CT scan. Etiology, instrumentation, levels fused, and complications were documented. We assessed all patients clinically for neurological recovery and followed over a period of 6 months. Neurological outcome using Frankel classification (Grade A: no function, Grade B: sensory only, Grade C: some sensory and motor preservation, Grade D: useful motor function, and Grade E: normal function) and Japanese orthopedic association score (JOA) [Annexure 1] were assessed for all patients in pre-operative period, at 6 months and 1 year. The JOA score was developed by the JOA in 1975. Since then, it has become one of the most frequently used outcome measures to evaluate functional status in patients with cervical myelopathy.

Inclusion criteria

- Patients admitted to the Neurosurgery Department with symptoms of craniovertebral compression and radiologically proven AAD, treated by single-stage posterior fixation.

Exclusion criteria

- Patient not giving consent for posterior fixation
- Patients of AAD with other comorbidities
- Irreducible AAD.

Statistical analysis

The results are presented in frequencies, percentages, and mean±SD. The Paired t-test was used to compare the mean change in continuous variables from pre-operative to subsequent time periods. The $P<0.05$ was considered statistically significant. All the analysis was carried out on SPSS16.0 version (Chicago, Inc., USA).

RESULTS

The present study was conducted in the department of neurosurgery, Jayarogya and associated hospital, GRMC, Gwalior, with the objective to study the various clinical and radiological aspects of congenital AAD and surgical outcomes with single-stage posterior correction.

A total of 31 patients were included in the study and analyzed. In our study, in demographic distribution, more than half of the patients were >30 years (54.8%) followed by <20 (35.4%). Mean to be 30.83±16.12. In terms of gender distribution, more than half were males (64.5%) and females 35.5%.

Table 1 shows the distribution of patients according to symptoms. Weakness of all four limbs and difficulty in walking was present in all the patients. Stiffness and numbness in all four limbs were present in 58% of patients. Table 2 shows the comparison of mean change in JOA motor dysfunction score of upper limb from pre-operative to 3 months and 6 months in C1-C2 screw and rod fixation. Change was found to be statistically significant. Table 3 shows the comparison of mean change in motor dysfunction score of lower limb from pre-operative to 3 months and 6 months in C1-C2 screw and rod fixation.

Table 1: Distribution according to age

Age in years	No. (n=31)	Percent
<20	11	35.4
20–30	3	9.6
>30	17	54.8

Table 2: Distribution according to gender

Gender	Number	Percent
Male	20	64.5
Female	11	35.5

Table 3: Distribution of patients according to symptoms

Symptoms	Number	Percentage
Neck pain	16	51.6
Weakness of all four limbs	31	100
Bladder-bowel dysfunction	4	12.9
Difficulty in walking	31	100
Stiffness	18	58
Suboccipital headache	3	9.6
Trauma	4	12.9

Change was found to be statistically significant. Table 4 shows the comparison of mean change in JOA total average score from preoprative to 3 months and 6 months in C1-C2 screw and rod fixation. There was a significant mean change.

DISCUSSION

The present study was designed and conducted at the Department of Neurosurgery, Gajra Raja Medical College, Gwalior to study clinic-radiological aspect of AAD along with role of single stage posterior correction in AAD.

Our study indicates that AAD is an important issue affecting middle aged and adolescent population.

Yang et al.,[13] reported most of their cases in adolescent age group. Similarly, Martinez-Del-Campo et al.,[11] reported mean age in their series to be 39.9 years with range 7 months to 88 years. One-third of their patients were younger than 40 years. Liang et al.,[12] reported mean age 44 years. Mean age in our series was 30.28±16.36 range (6-56).

More than half of our patients were males 64.5% and 35.5% were female. Martinez-Del-Campo et al.,[11] reported 64 (53%) male and 56 (47%) female patients in their study. Male predominance is probably due to increased number of male patients being investigated, which is an important social feature in the Indian society.

The presentation of AAD may range from minor axial neck pain to death (Table 5). All of our patients had weakness

Table 4: Comparison of mean change in JOA motor dysfunction score of upper limb from preoperative to 6 months in c1-c2 fixation

Time period	Mean change in JOA motor dysfunction score	P-value
Preoperative to 3 months	1.58±0.71	0.0001
Preoperative to 6 months	1.79±0.65	0.0001

JOA: Japanese orthopedic association score

Table 5: Comparison of mean change in JOA motor dysfunction score of ll from preoperative to 6 months in c1-c2 fixation

Time period	Mean change in motor dysfunction score in ll	P-value
Preoperative to 3 months	2.08±0.77	0.0001
Preoperative to 6 months	2.75±0.75	0.0001

JOA: Japanese orthopedic association score

of all four limbs with difficulty in walking (100%). Yang et al.,[13] reported 90% of their patients with pyramidal signs. Numbness and tingling in all four limbs were present in 18 (58.0%) followed by neck pain in 16 (51.6%) of patients in our study. Yang et al.,[13] reported 70% of their patients with numbness and 50% with neck pain, similar to our results. 4 (12.9%) of our patients had bladder dysfunction, 3 (9.6%) had suboccipital headache, and 1 (3.2%) patient had difficulty in breathing. While Martinez-Del-Campo et al.,[11] in their study reported neck pain to be most common presenting complaint in 83 (69%), followed by numbness and weakness in 52 (43%), bladder bowel incontinence in 20 (17%), and gait ataxia in 13 (11%). They also reported that 37 (31%) of their patients had no neurological deficit preoperatively.

Weakness of all four limbs and walking difficulty was predominant symptom in our series. The probable reason is late presentation at a tertiary care center for definitive management in our Indian society.

In our study (Table 6), the pre-operative neurological status of all patients who underwent C1 lateral mass C2 pedicle screw fixation fell in Frankel grade C. All of our patients had one Frankel grade improvement at 6 months.

Similar results were seen in a study by Kwan et al.,[14] who reported one Frankel grade improvement in their 85.7% of patients at final follow-up.

In this study, we assessed patients on the basis of Modified JOA score along with Frankel grading.

The mean improvement in the motor dysfunction score of upper limb from pre-operative period to 3 months and 6 months was statistically significant.

Table 6: Comparison of mean change in JOA total average score from preoperative to 3 months to 6 months in screw rod fixation

Time period	Mean change in JOA in total average score	P-value
Preoperative to 3 months	4.08±1.58	0.0001
Preoperative to 6 months	5.37±1.61	0.0001

JOA: Japanese orthopedic association score

Comparison of mean change in JOA motor dysfunction score LL from pre-operative to 3 months and 6 months was statistically significant. JOA sensory dysfunction score in pre-operative period was 2.25±0.53 which increased to 2.58±0.50 at 6 months and 2.75±0.44 at 1 year. Comparison of mean change in JOA sensory dysfunction score from pre-operative to 6 months ($P=0.003$) and 1 year ($P=0.0001$) was statistically significant.

(Table 7) JOA total average score was 9.88±1.87 at pre-operative which increased 15.25±0.98 at 6 months. There was a significant ($P=0.0001$) mean change in JOA total average score from pre-operative to 3 months and 6 months period. Similarly, Martinez-Del-Campo et al.,[11] reported a preoperative mean JOAS score of 12.0±3.5 which increased to 14.8±3.4 postoperatively ($P<0.001$) and 14.9±3.5 at last follow-up ($P<0.001$). Similarly, Liang et al.,[12] reported a preoperative mean JOAS score 7.3 ± 1.8 which increased to 13.1 ± 1.4 at last follow-up ($P<0.001$). These results were similar to our study when compared at last follow-up.

JOA score can be used to assess the improvement in different domains of neurological impairment and can be followed over time with easy reproducibility and without any observer bias.

In our study (Table 8), the mean of ADI decrease after operation was statistical significance ($P<0.01$). The mean and SD of ADI before operation and after operation was 7.88±1.36 and 4.4±1.25. Sutipornpalangkul and Thanapipatsiri[15] in 2014 did atlantoaxial transarticular screw fixation and posterior fusion using polyester cable and published mean and SD of ADI before operation and after operation which was 4.86±3.07 and 2.76±1.38 mm, respectively. They also reported it to be statistically significant. Both studies show similar results with reduction in mean ADI after reduction and fixation of C1-C2 joint.

In our study, we did C1 and C2 pedicle screw and rod fixation with atlantoaxial instability (Table 9). We observed excellent outcomes in terms of fusion rates which was 100%. All of cases achieved union with average union time of 5.5 months. Rezaee et al.,[10] reported similar fusion rates in 3.5 months and Kwan et al.,[14] reported 100%

Table 7: Comparison of mean age Group in various studies

S. No.	Study	Mean age (years)
1	Rezaee et al.[10]	34.67
2	Martinez-Del-Campo et al.[11]	39.9
3	Liang et al.[12]	44.5
4	Present study	30.28

Table 8: Comparison of male to female ratio in various studies

S. No.	Study	Mean age (years)
1	Martinez-Del-Campo et al.[11]	1.14:1
2	Liang et al.[12]	1.89:1
3	Present study	1.85:1

Table 9: Comparison of various signs and symptoms of AAD in various studies

S. No.	Study	Symptoms
1	Yang et al.[13]	Pyramidal signs (90%) Numbness and Tingling (58%) Neck pain (51%)
2	Martinez et al.[11]	Neck pain (69%) Numbness (52%) Weakness of limbs (52%)
3	Present study	Neck pain (51.6%) Numbness (58%) Weakness of all limbs (100%) Difficulty in walking (100%)

fusion rates in 5.3 months. Yoon et al.,[19] have achieved excellent bone fusion using C1 lateral mass screw and C2 pedicle screw compared to the results of other methods without any procedure-related complication. Dickman and Sonntag[16] reported similar 86% union rate in their C1-C2 fixations with wires and autograft (n=74). In a similar study conducted at the University of Toronto by Coyne et al.,[17] reported (19%) of their wiring fusions failed. We had similar results in terms of fusion rates when compared to other studies. P. Bourdillon et al[18] indicates that Goel and Harms fusion is to be considered the first line of choice for C1-C2arthrodesis.

Early post-operative complications included neck pain as the main compliant of all patients after surgery which significantly reduced in due course of time. Graft site pain was also a limiting factor in early mobilization of patients, but we did not encounter its persistence beyond 10 days. Sawin et al.,[20] reported a bone graft donor site morbidity, 4% during rib and 25% during iliac crest harvests. In our study, surgical site infection occurred in 2 cases (6.4%) limited to subcutaneous plane only and subsided with antibiotics. Martinez-Del-Campo et al.,[11] reported 5.8% wound healing complications similar to our series. Martinez-Del-Campo et al.,[11] also reported vertebral artery injury in three patients (2.5%). In our study, we encountered vertebral artery injury in one patient. Furthermore, we encountered death of one patient. We did encountered cerebellar infarct in one patient (with dominant vertebral artery injury intraoperatively) in post-operative period which lead to gradual deterioration and finally death of patient on fourth postop day. In a study by Martinez-Del-Campo et al.,[11] no mortality was reported as they had vertebral artery injury in three patients but of non-dominant side. Similarly Rezaee et al.,[12] Kwan et al.,[14] and Harms and Melcher[21] also did not reported any mortality in their studies.

Limitations of the study

Patients requiring two or multi staged corrective procedures were not included in this study.

CONCLUSION

This study reviewed single-stage posterior correction of AAD using C1 lateral mass and C2 pedicle screw and rod fixation, it enables the stabilization of atlantoaxial motion and preserves motion at occipito-atlantal level with high fusion rates, good neurological recovery, and minimal complications.

ACKNOWLEDGMENT

The authors are acknowledged to present part of this paper in the form of paper Presentation at MP chapter NSICON 2022.

ETHICAL COMMITTEE APPROVAL

Letter No. 1004, dated 16/07/2021.

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AK- Definition of intellectual content, literature survey, prepared first draft of manuscript, implementation of study protocol, data collection, data analysis, manuscript preparation and submission of article; **VKK-** Concept, design, clinical protocol, manuscript preparation, editing and revision, statistical analysis; **AS-** Review manuscript, preparation of tables; **AvdS-** Coordination and manuscript revision; **MJ-**Table editing and review; **AviS-** Coordination and manuscript revision.

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Source of Support: Nil, **Conflicts of Interest:** None declared.