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ORIGINAL ARTICLE

Feasibility and outcome of stand-alone trans-articular screw fixation in atlantoaxial instability in children less than 8 years of age

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Abstract

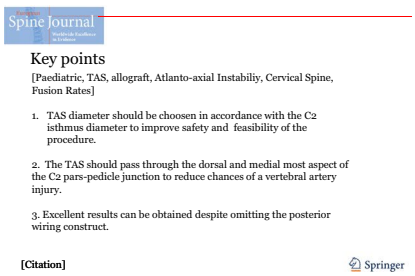
Purpose To study the feasibility and outcome of stand-alone trans-articular screw (TAS) fixation for atlantoaxial instability (AAI) in children less than 8 years of age.

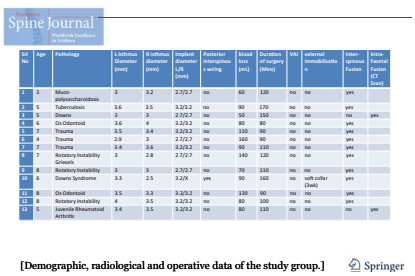
Methods This prospective study was conducted between 2009 and 2014. Thirteen children suffering from AAI were operated for a TAS fixation. Feasibility of TAS fixation was assessed on CT scan and a screw diameter was chosen based on C2 isthmus diameter. Demographic data collected included the etiology for AAI, age, and sex. Intra-operative data recorded was the duration of surgery, blood loss, vertebral artery injury or any adverse event. Radiological evaluation included pre- and post-operative atlantodens interval (ADI) and space available for cord (SAC) and fusion was evaluated at 3, 6, 12 and 24 months. Statistical analysis was done using SPSS software and statistical significance was set at $p < 0.05$.

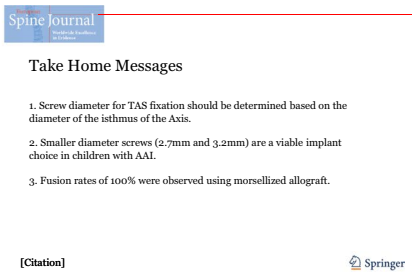
Results The mean age of the final study group was 6.1 ± 1.5 years, with nine males and four females. Mean isthmus diameter on the left and right side was 3.3 ± 0.3 and 3.2 ± 0.2 mm, respectively. Five patients had an isthmus diameter of < 3.2 mm and a 2.7 mm Herbert screw was used in them and in nine patients, a CCS of 3.2 mm was used. Mean pre- and post-op ADI and SAC improved from 5.5 ± 0.8 to 3.1 ± 0.1 mm, respectively, and 9.8 ± 2.8 to 14 ± 0.6 mm, respectively. Fusion was seen in all patients.

Conclusions Stand-alone TAS with morselized allograft is safe, feasible and successful in managing AAI in children below 8 years of age.

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







Keywords Pediatric · TAS · Allograft · Atlantoaxial instability · Cervical spine · Fusion rates

Introduction

The management of atlantoaxial instability (AAI) in children is fraught with many challenges. Various modalities of fixation for AAI (trans-articular screw fixation, C1–C2 segmental fixation) are reported to have excellent outcomes in adults [1–3]. However, many of these techniques are not

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considered suitable for children [4]. Instead, these children are offered alternative fixation options, such as Gallies' fixation and other posterior wiring techniques. Poor fusion rates and dependence on external immobilization remain a significant drawback of posterior wiring techniques [5, 6]. In this study, we report the feasibility of stand-alone TAS fixation without supplemental posterior wiring, in children less than 8 years of age using screws of smaller diameter [2.7 mm Herbert screw and 3.2 mm cannulated cancellous screws (CCS)]. We also replaced the inter-spinous structural autograft with morselized allograft and report the outcomes of using allograft alone.

Materials and methods

This was a prospective study, conducted at a single institute over a period of 5 years (2009–2014). Patients, 8 years of age or lesser, with a mobile, reducible AAI and a minimum of 24-month follow-up were included in the study. We excluded patients with an irreducible AAI, isthmus diameter of less than 2.7 mm, basilar invagination and/or C1 lateral mass/C2 articular mass destruction.

Children less than 8 years of age, having symptomatic AAI, were evaluated radiologically to determine the feasibility of TAS fixation. The study protocol included assessing plain radiographs, dynamic radiographs, CT scan in flexion and extension and MRI scans. All patients underwent CT scanning using a multidetector CT. All images were reconstructed with a slice thickness of 1 mm and reconstruction interval of 1 mm. Consideration for the feasibility of TAS fixation was based on (1) reducibility of C1/C2 joint complex, (2) isthmus thickness and (3) the presence of anomalous anatomy. AAI was deemed mobile and reducible when dynamic X-ray/CT scan or pre-operative traction revealed an ADI of less than 4 mm. High riding vertebral artery (HRVA) was defined as an isthmus diameter of less than 2.7 mm on the parasagittal CT cut, 3 mm lateral to the lateral spinal canal. This was chosen as the cut-off (instead of 4 mm in adults) for a HRVA as the outer diameter of the cannulated cancellous screws (CCS) used in this series for TAS fixation is 2.7 mm (and not 3.5 mm). An isthmus diameter of < 2.7 mm precludes the possibility of placing TAS fixation safely.

All the patients were operated by a single surgeon (Dr. V.K).

Demographic data collected included the etiology for AAI, age, and sex. Intra-operative data recorded was the duration of surgery, blood loss, vertebral artery injury (VAI), neurological injury or any adverse event. Radiological evaluation included pre- and post-operative atlantodens interval (ADI) and space available for the cord (SAC). Post-op radiographs were evaluated at 3, 6, 12 and 24 months for fusion, both interarticular and inter-spinous. If inter-spinous fusion was not seen at 9 months, a CT scan was done to assess for intra-articular fusion. "Complete" fusion was defined as radiographically continuous bone formation across the treated levels.

Surgical technique

The patient was positioned prone on the operative table on padded rolls. Pre-operative traction was applied with 5 kg weight. A 'military tuck' position was given and fixed in place rigidly with the Mayfield head holder system after confirmation of satisfactory C1–C2 reduction on the lateral radiograph. The skin incision was centered over C2. Only the inferior arch of C1 and complete C2 was subperiosteally exposed. The C1–C2 joint was opened with a McDonald or pen-field and the joint surface was curetted and packed with chips of morselized allograft. The entry point at C2 was determined based on a retrograde projection drawn out on the intra-operative lateral image and anatomical landmarks on visualization. From the tip of the anterior tubercle of the atlas, a line was produced passing through the dorsal most point of the C2 pars-pedicle junction and continued distally to the C2/3 joint line (Fig. 1a). This marks the superoinferior entry plane. The mediolateral angulation was determined by starting at a point on the superoinferior plane just 5 mm medial to the dorsal most and medial aspect of the C2 pars-pedicle junction (Fig. 1b). This ensures a slightly laterally directed screw to purchase the thickest mass of the C1 lateral mass. The drill guide was passed percutaneously, via a stab incision at the T1 T2 region, into the primary exposure and docked on the entry point. Once the drill guide was in place, a high-speed burr was used to create a starter hole in C2 and a 1.2-mm guide wire was passed via the C2 into the anterior cortex of C1 under lateral C-arm guidance. It is essential to drill as dorsally and medially as possible within the isthmus to minimize potential injury to the vertebral artery. Implant choice was dependant on the thickness of the isthmus on CT scan. For those children with a C2 isthmus thickness between 2.7 and 3.2 mm, a 2.7 mm Herbert screw was used. Moreover, for children with an isthmus diameter between 3.2 and 4 mm, a 3.2 mm cannulated cancellous screw was used. The same procedure was repeated on the contra lateral side and screws are passed.

The lamina of the C2 vertebra and inferior C1 arch were decorticated with a high-speed burr before application of the bone graft. Freeze dried cancellous allograft from the bone bank was morselized and placed between the posterior arch of C1 and the spinous process of the C2 vertebra and along bilateral facet joints.

All statistical analyses were performed using SPSS version 22.0 (SPSS Inc., Chicago, IL, USA). A p value was set at < 0.05 for significance.

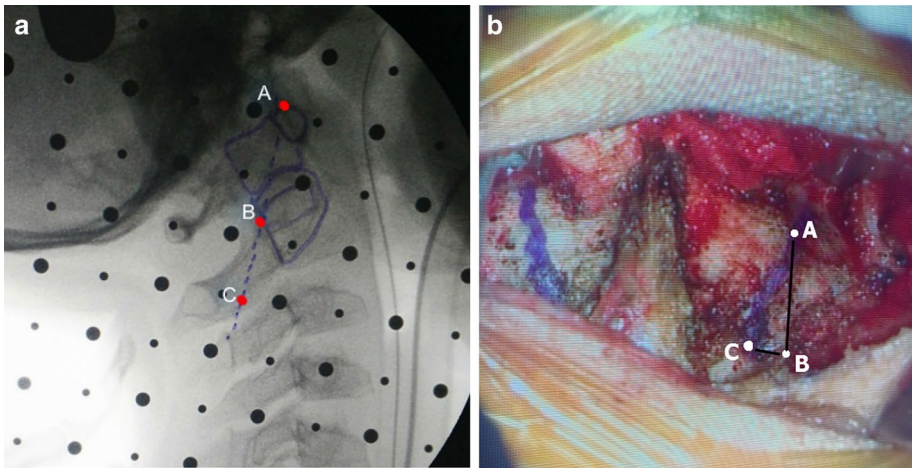

Fig. 1 **a** Trajectory planning and entry point for each screw requires carefully determining important landmarks from radiologically and in the operative field anatomically. On the lateral image point A is the final target, just superior to the anterior tubercle of C1 in a well reduced atlantoaxial joint. Point B is the dorsal and medial most aspect of the C2 pars-pedicle junction which is exposed intra-operatively. Connecting these two points, a retrograde projection is made to get a plane of entry above C2/3 facet joint, marked by point C here. **b** Point A marks the dorsal and medial most part of the pars-pedicle junction of C2. Point B is a vertical projection of point A on the superoinferior plane. Point C is a point on the pre-determined superoinferior plane that is 5 mm medial to point B

Results

Between 2009 and 2014, a total of 15 children below 8 years of age suffering from AAI were operated for TAS fixation. Of them, two patients had not completed 2 years of follow-up and were excluded from the study. The remaining 13 patients fulfilled all our inclusion criteria and formed part of the final study group. The final study group comprised nine boys and four girls, with a mean age of 6.1 years (3–8 years). In this series, there were seven different causes of AAI, the commonest being post-traumatic AAI seen in three children (Table 1).

Pre-op radiographs and CT scan revealed that TAS fixation was feasible in 12 patients bilaterally and unilaterally (left side) in one patient with AAI due to Down syndrome (High riding vertebral artery on the right side, 2.5 mm isthmus diameter). In this patient, the unilateral TAS was combined with an inter-spinous wiring. A patient suffering from rotatory AAI had an assimilated C1 and another patient had an aplasia of the posterior arch of atlas. Both were deemed safe for stand-alone TAS fixation.

Mean isthmus thickness on the left and right side were 3.3 ± 0.3 and 3.2 ± 0.2 mm, respectively. Five patients had an isthmus thickness of less than 3.2 and a 2.7 mm Herbert screw was used in all these patients. In the remaining eight patients, a CCS of 3.2 mm was used. Mean screw length was 30.4 ± 1.8 mm. Mean pre- and post-op ADI and SAC improved from 5.5 ± 0.8 to 3.1 ± 0.1 mm, respectively, and 9.8 ± 2.8 to 14 ± 0.6 mm, respectively, both of which were statistically significant.

Intra-operative parameters revealed a mean surgical time of 115 ± 27 min and a mean blood loss of 95 ± 35 ml. No evidence of vertebral artery perforation was noted. No other complications were recorded.

Radiographically evident fusion was recorded with a mean interval of 6 months, ranging from 3 to 12 months in these children. Two patients (one child suffering from JRA and one child suffering from Down syndrome) showed no inter-spinous fusion and CT scan done at 9 months revealed intra-articular fusion between C1 and C2 facet joints (Fig. 2a–d). Fusion was found to be present in all patients at final follow-up 100%.

Discussion

Trans-articular screws have been successfully used in C1–2 fixation in adult patients with fusion rates of 96–100% [7]. A handful of studies exploring the feasibility of TAS fixation in children have also reported good results [4, 8, 9]. The mean age of the children in these studies range from 9 to 12 years. The age at which a child's craniovertebral region starts resembling adult size and characteristics is reported to be between 8 and 10 years of age [10, 11]. The aim of this study was to assess the feasibility of TAS fixation before a child's craniovertebral junction attains adult size and characteristics, i.e., before 8 years of age. In this series of 13 patients, the mean age was 6.1 years and ranged from 3 to 8 years. To the author's knowledge, this is the only study to have reported the feasibility and outcomes of TAS fixation in this age group.

Table 1 Demographic, radiological and operative data of the study group

S. no.	Age	Pathology	*L* isthmus diameter (mm)	*R* isthmus diameter (mm)	Implant diameter *L*/*R* (mm)	Posterior inter-spinous wiring	blood loss (ml)	Duration of surgery (min)	VAI	External immobilization	Inter-spinous fusion	Intra-facetal fusion (CT Scan)
1	3	Mucopolysaccharidosis	3	3.2	2.7/2.7	No	60	120	No	No	Yes	
2	5	Tuberculosis	3.6	3.5	3.2/3.2	No	90	170	No	No	Yes	
3	5	Downs	3	3	2.7/2.7	No	50	150	No	No	No	Yes
4	6	Os odontoid	3.6	4	3.2/3.2	No	80	80	No	No	Yes	
5	7	Trauma	3.5	3.4	3.2/3.2	No	110	90	No	No	Yes	
6	4	Trauma	2.9	3	2.7/2.7	No	160	90	No	No	Yes	
7	7	Trauma	3.4	3.6	3.2/3.2	No	90	110	No	No	Yes	
8	7	Rotatory instability Griesels	3	2.8	2.7/2.7	No	140	120	No	No	Yes	
9	8	Rotatory instability	3	3	2.7/2.7	No	70	110	No	No	Yes	
10	6	Downs syndrome	3.3	2.5	3.2/X	Yes	90	160	No	Soft collar (3 weeks)	Yes	
11	8	Os odontoid	3.5	3.3	3.2/3.2	No	130	90	No	No	Yes	
12	8	Rotatory instability	4	3.5	3.2/3.2	No	80	100	No	No	Yes	
13	5	Juvenile rheumatoid arthritis	3.4	3.5	3.2/3.2	No	80	110	No	No	No	Yes

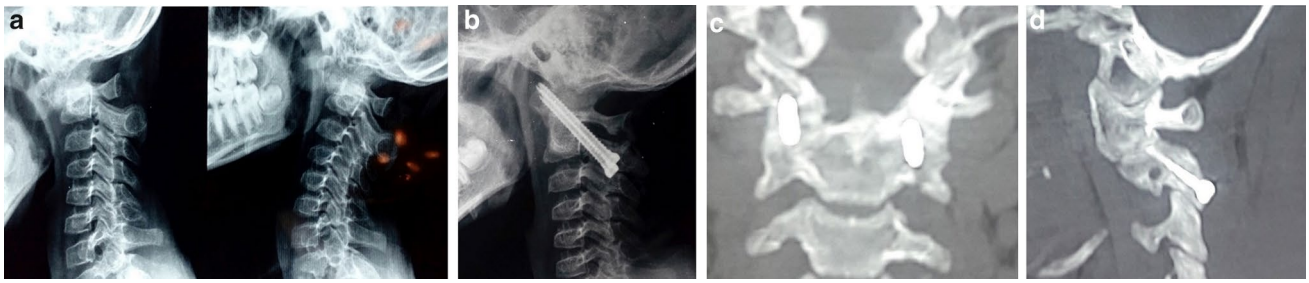

Fig. 2 **a** Pre-operative lateral dynamic radiograph of the cervical spine of 5-year-old child suffering from juvenile rheumatoid arthritis showing AAI. **b** Nine-month post-operative image showing no inter-spinous fusion. **c**, **d** Coronal and sagittal images showing solid facet fusion

In children suffering from AAI, a TAS fixation was planned only when the child's craniovertebral anatomy was deemed safe and viable for the procedure. This was determined by studying the C2 isthmus diameter on the pre-operative CT scans. We assessed the narrowest C2 isthmus diameter on 3 mm parasagittal CT scan images starting from the lateral spinal canal border and opted for a screw diameter in accordance. Children with an isthmus diameter between 2.7 and 3.2 mm were considered safe for TAS fixation with a 2.7 mm cannulated cancellous screw and children with an isthmus diameter of more than 3.2 mm, underwent fixation with a 3.2 mm screw. Five children had an isthmus diameter of less than 3.2 mm and underwent fixation using a Herbert screw (2.7 mm) and the other eight underwent fixation with a 3.2 mm CCS. Only one child with AAI secondary to Down syndrome had a C2 isthmus thickness of 2.5 mm on the right side and that side was deemed unsuitable for TAS fixation. Instead, he underwent a unilateral TAS with wiring and morselized allograft. Considering the plasticity of the child's bone, 2.7 mm was chosen as the cut-off for the isthmus diameter to place a 2.7 mm screw [12].

Trans-articular screw fixation is often contra-indicated in patients with a HRVA. Traditionally, a high riding vertebral artery is defined as a C2 isthmus diameter of 4 mm or less [13]. Four millimeters is chosen as the cut-off as there needs to be an adequate bone stock to accommodate the 3.5 mm screw used in TAS fixation [13]. As we used screws with a smaller diameter, the cut-off for a high riding vertebral artery was set at 2.7 mm. Using these smaller diameter screws, we could place screws in 25 out of a total of 26 atlantoaxial joints safely. The one screw that had to be omitted was due to a high riding vertebral artery (2.5 mm pars thickness). In this series, all 13 patients were found to be anatomically suitable for at least one C1–C2 screw and bilateral suitability was seen in 96% children. In comparison, Brockmeyer et al. studying the suitability of 3.5 mm screws for TAS fixation in the pediatric age group found at least one C1–2 joint was anatomically suitable in 91% cases, whereas bilateral suitability was seen in 86% cases [8]. The increased suitability of the procedure reported in this series is due to the use of smaller diameter screws. In this series, none of the 13 children had any neuro vascular compromise and no vertebral artery perforation was noted. We attribute this to (1) use of smaller diameter screws, (2) careful assessment of pre-operative CT scans, (3) only going ahead with the plan when the C1C2 joint is fully reducible and (4) the use of a retrograde trajectory as described earlier. Using this technique, the screw passes through the most dorsal and medial aspect of the C2 isthmus, reducing the likelihood of violating the vertebral artery groove. There was no pre-determined fixed entry point as described by Magerl et al. but an entry point based on each case [14]. Using a similar study population, Gluf and Brockmeyer had reported two vertebral artery perforations in their study of 36 patients [4].

Trans-articular screw fixation as described by Magerl et al. is accompanied with inter-spinous wiring and structural autograft between C1 and C2 spinous processes [14]. Multiple reports of sustained excellent outcomes despite posterior graft/wire failure are reported. In Matsumoto et al. series, they reported 18 cases (34.6%) of loosening of posterior wiring construct in 52 cases [15]. Despite this there was a 95% fusion rate. It reported 100% fusion rates even though all his cases had some degree of wire loosening, to which he concluded that adding a wire construct is not required [16]. From these results, it is apparent that the posterior wiring construct may not significantly contribute to the fusion process. However, the addition of the posterior wire is associated with a 7–17% risk of neurologic deterioration, and unless deemed essential, should be avoided [16]. Posterior wiring itself may not be possible if AAI occurs in conjunction with the absence/hypoplasia of the posterior arch of C1 [17]. In few cases, the posterior arch of C1 itself may be removed surgically for decompression. Besides posterior inter-spinous wiring, autograft harvest also increases the operative time, blood loss, predisposes the patient to intra-operative and post-operative complications associated with wiring and graft harvesting. Wang et al. obtained 100% fusion rates despite omitting the supplementary posterior wiring and structural graft. Instead, they used morselized autograft between C1

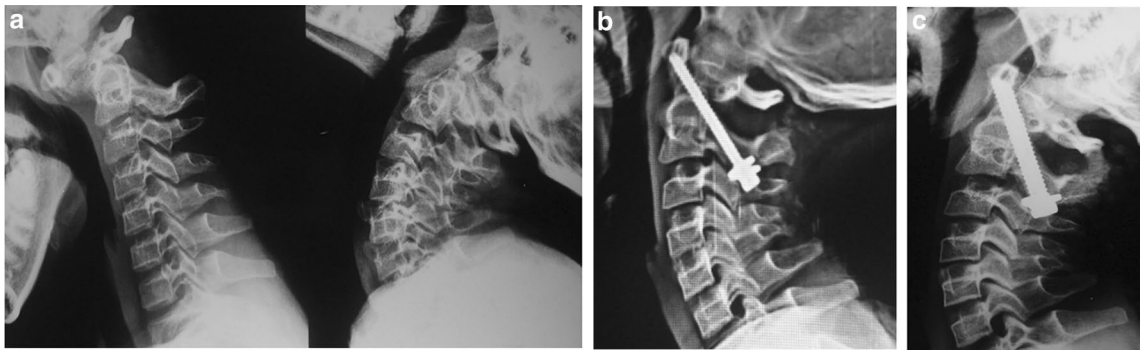

Fig. 3 **a** Six-year-old boy suffering from AAI due to an Os odontoid. Pre-operative lateral radiograph showing instability. **b** Immediate post-operative lateral radiograph showing good reduction and fixation using two 3.2-mm cannulated cancellous screws. No inter-spinous wiring done and morselized allograft used. **c** One-year post-operative lateral image showing solid inter-spinous fusion

arch and C2 laminae [18]. While autograft is the ideal graft material with osteo-conductive, osteo-inductive and osteo-genic properties, harvesting it in children is associated with a 10–40% risk [19]. To reduce the risk associated with autograft harvest, we used freeze-dried allograft from the bone bank. The source of this graft was the head of femur and proximal tibia bone removed from individuals undergoing replacement surgery for osteo-arthritis of the knee/hip. In our series, using morselized allograft, fusion was seen in all our patients (100%) at final follow-up (Fig. 3a–c). Two patients did not show inter-spinous fusion on lateral radiographs at final follow-up, however, CT scans revealed fusion at the facet level. We attributed our low mean surgical time of 115 ± 27 min and a mean blood loss of 95 ± 35 ml to (1) Not using autograft and (2) not using sublaminar wires to supplement the TAS fixation, and (3) careful pre-operative planning.

Potential concerns regarding the effects of atlantoaxial fusion on the normal craniovertebral growth and development exists. As the craniovertebral junction attains adult size and characteristics by 8–10 years of age, the impact of fusion at and around this age should have a minimal influence on growth. However, the mean age in this study was 6.1 years, with the procedure being performed in children as young as 3 years old. Long-term follow-up is needed to study the implications of fusion on the normal growth and development of the upper cervical spine in these children. The smaller diameters of screws used in this study also raise concerns of implant breakage and the subsequent need of revision surgery. We did not face this issue in our study group as all the children achieved fusion.

Limitations of our study are that it was a small cohort, managed at a single center by a single surgeon. Similar studies need to be conducted on a larger scale to reinstate the outcomes and emphasize the feasibility of smaller diameter screws.

Conclusion

Trans-articular screw fixation in the pediatric population is feasible and is associated with high fusion rates. Added advantages of this technique are the avoidance of a post-operative halo vest, elimination of the need for instrumentation in the canal, applicability despite anomalies involving the C1 post arch and non-dependence on autograft harvest. Flexibility in implant diameter allows implantation in cases which were previously considered to be unsafe due to a high riding vertebral artery. This makes this technique a valuable procedure in the armamentarium of a spine surgeon for the management of pediatric atlantoaxial instability.

Compliance with ethical standards

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

Ethical statement The manuscript submitted does not contain information about medical device(s)/drug(s). Bombay Hospital research ethics committee BRHC 1013/12. Permission to reproduce copyrighted material granted.

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