

Treatment of pediatric atlantoaxial instability with traditional and modified Goel–Harms fusion constructs

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Abstract There are several treatment options for rigid fixation at C1–C2 including Brooks and Gallie type wired fusions and C1–2 transarticular screws. The use of a Goel–Harms type fusion, a construct with C1 lateral mass screws and C2 pedicle screws, has not been extensively described in pediatric patients. Here, we describe its relatively safe and effective use for treating pediatric patients by retrospective chart review of patients treated by the senior author for atlantoaxial instability with a Goel–Harms-type constructs during a 3-year period (2005–2007). Six patients were treated using Goel–Harms-type constructs. Five patients were treated utilizing a construct containing C1 lateral mass screws and C2 pedicle screws; one patient was treated using construct containing C1 lateral mass screws and C2 trans-laminar screws. The patients ranged in age from 7 to 17 years old (mean 12.7). All patients had findings of an os odontoideum on CT scans and three of the six patients had T2 hyperintensity on MRI. Three of the six patients presented with transient neurologic deficits: quadraplegia in two patients and paresthesias in two patients. In each patient C1 lateral mass and C2 screws were placed and the subluxation was reduced to attain an anatomical alignment. No bone grafts were harvested from the iliac crest or rib. Local morsalized bone and sub-occipital skull graft was used. All patients tolerated the procedure well

and were discharged home on post-operative day 3–4. The patients wore a hard cervical collar and no halo-vests were needed. All patients had solid fusion constructs and normal alignment on post-operative imaging studies performed on average 14 months post-operatively (range: 7–29). The results demonstrated that Goel–Harms fusions are a relatively safe and effective method of treating pediatric patients with atlantoaxial instability and are not dependent on vertebral anatomy or an intact ring of C1. Follow-up visits and studies in this limited series of patients demonstrated solid fusion constructs and anatomical alignment in all patients treated.

Keywords Atlantoaxial instability · Os odontoideum · Goel–Harms fusion · Pediatrics · Cervical spine

Introduction

Management of upper cervical instability, particular atlantoaxial instability, can be challenging in the pediatric population. The goal of surgical intervention is to decompress the neural elements and generate a biomechanical construct that is stable and promotes a solid arthrodesis. Various treatment options have been described. Traditional posterior wiring techniques, including Brooks and Gallie fusions, have a long history of success in all age groups [7, 8, 16, 29]. More recently, the introduction of transarticular C1–2 fusions have been described and when technically possible have been shown to be effective in adults as well as pediatric patients [1, 2, 6, 13, 18, 19, 39].

Harms and Melcher described a method of posterior C1–C2 fixation [23]. A similar technique was previously described by Goel and Laheri [20]. The Goel–Harms construct consists of placement of screws into the lateral

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Table 1 Patient clinical Characteristics

	Age (years)	Sex	Co-morbid	Pre-op deficits	T2WI ^a	Flex-ex (mm) ^b	Follow-up ^c (months)
Patient 1	14.3	F	None	Transient paresthesias, and quadraplegia	Y	5	29
Patient 2	11.5	M	Chiari I	Transient paresthesias	N	8	12
Patient 3	7.5	F	Trisomy 21	None	N	NA	8 ^d
Patient 4	17.1	F	Trisomy 21	None	Y	NA	20
Patient 5	14.3	M	None	Transient quadraplegia	Y	10	12
Patient 6	11.4	F	Seizure disorder	Neck pain	Y	7	7

NA not available

^a T2WI presence of hyperintensity on T2 weighted MRI images

^b Flex-ex amount of movement in millimeters (mm) at C1–2 on flexion–extension images

^c Follow-up date patient had last follow-up visit with imaging

^d Patient died from an unrelated condition prior to her 12 month follow-up

mass of C1 coupled to C2 pedicle screws. Additionally, a modification of the traditional Goel–Harms construct was described that uses C2 trans-laminar screws [2, 28]. These techniques have a high rate of fusion and can limit some of the potential morbidity associated with other techniques. Placement of C1 lateral mass screws has been described in pediatric patients and, although technically challenging, has been successful even in children less than 8 years of age [25, 28]. The use of a Goel–Harms construct for treatment of atlantoaxial instability has not been extensively described in pediatric patients. We describe our experience with two types of Goel–Harms constructs in an exclusively pediatric patient population. Three of the six patients presented with transient neurologic deficits: quadraplegia in one patient; paresthesias in one patient; and one patient had both quadraplegia and paresthesias.

Clinical materials and methods

We retrospectively reviewed patients that underwent a C1–2 fusion at the Children’s Hospital of Philadelphia (CHOP). All patients had pre-operative imaging that demonstrated an os odontoideum and atlantoaxial instability. All fusions were performed with intra-operative fluoroscopy. The patients were positioned in a Mayfield head holder and under fluoroscopic guidance were reduced and anatomically aligned. C1 screws were placed into the lateral masses and C2 pedicle screws were placed with fluoroscopic guidance (Synthes Axon System, Synthes, West Chester, PA, USA). Prior to placement of the instrumentation, the C1–2 joint was prepared and decorticated with a curette and a high-speed drill with an M8 bit. A C1 laminectomy was performed if it was felt that additional decompression was needed. After the instrumentation was placed, the lateral masses and lamina of C1 and the pars and lamina of C2 were decorticated. The

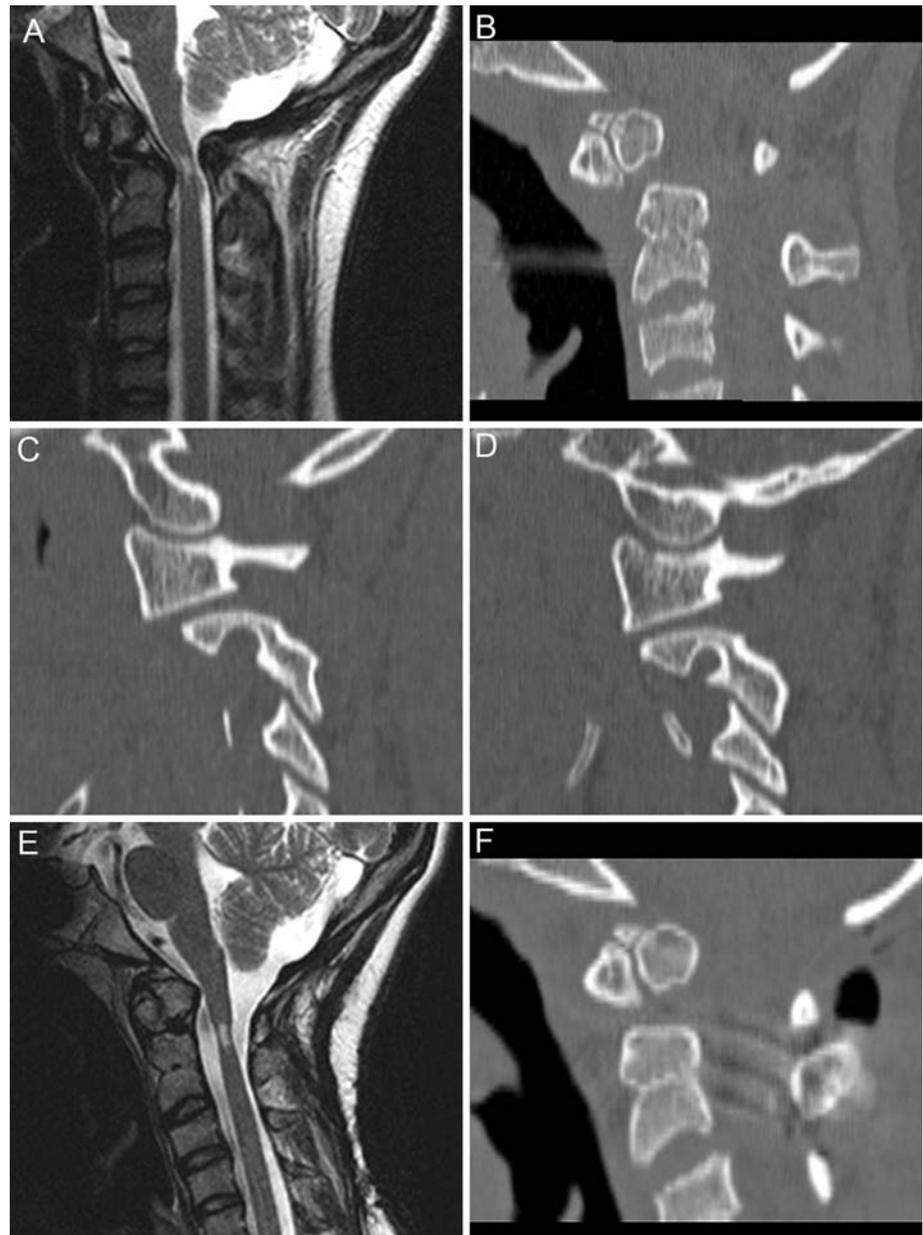
fusions were supplemented with DBX putty (Synthes, West Chester, PA, USA), local morselized bone, and allograft bone chips. No autograft was harvested from the iliac crest or the rib. In some cases where additional bone was needed, bone was harvested from the lateral sub-occipital bone with a burr with special attention paid to maintaining the midline keel so that it was available if future extended fusion operations were needed (Fig. 5a). All patients had routine neuro-monitoring throughout their operative course. The patients were placed in a Miami J cervical collar post-operatively and monitored in the pediatric intensive care unit. All patients had routine follow-up in the neurosurgical office and post-operative imaging confirming an arthrodesis. All patients underwent follow-up cervical spine imaging within 12 months of surgery. There were two exceptions, one patient died from an unrelated condition prior to 12 months and one patient had imaging at her latest follow-up (7 months). Confirmation of a stable arthrodesis was made with CT imaging on all patients requiring a laminectomy and, at minimum, dynamic flexion–extension testing in patients not undergoing laminectomy.

Results

We retrospectively reviewed patients who underwent a Goel–Harms type C1–2 fusion at the Children’s Hospital of Philadelphia (CHOP) between 2005 and 2007. All patients had pre-operative imaging that demonstrated an os odontoideum and atlantoaxial instability. The age range of the patients was 7–17 years old (mean 12.7 years old); four were female and two male (Table 1). Two patients had Downs syndrome and one patient had a Chiari I malformation.

The patients were brought into the operating room where, if possible, a closed reduction was performed under

Fig. 1 Representative radiographic images of Patient 1. Pre-operative MRI (a) and CT (b) demonstrating an os odontoideum with T2WI hyperintensity at C1–2 and focal stenosis. Sagittal images on the right (c) and left (d) demonstrate that the patient's anatomy is not ideal for transarticular screws. Post-operative MRI (e) and CT (f) demonstrate anatomic alignment and decreased stenosis

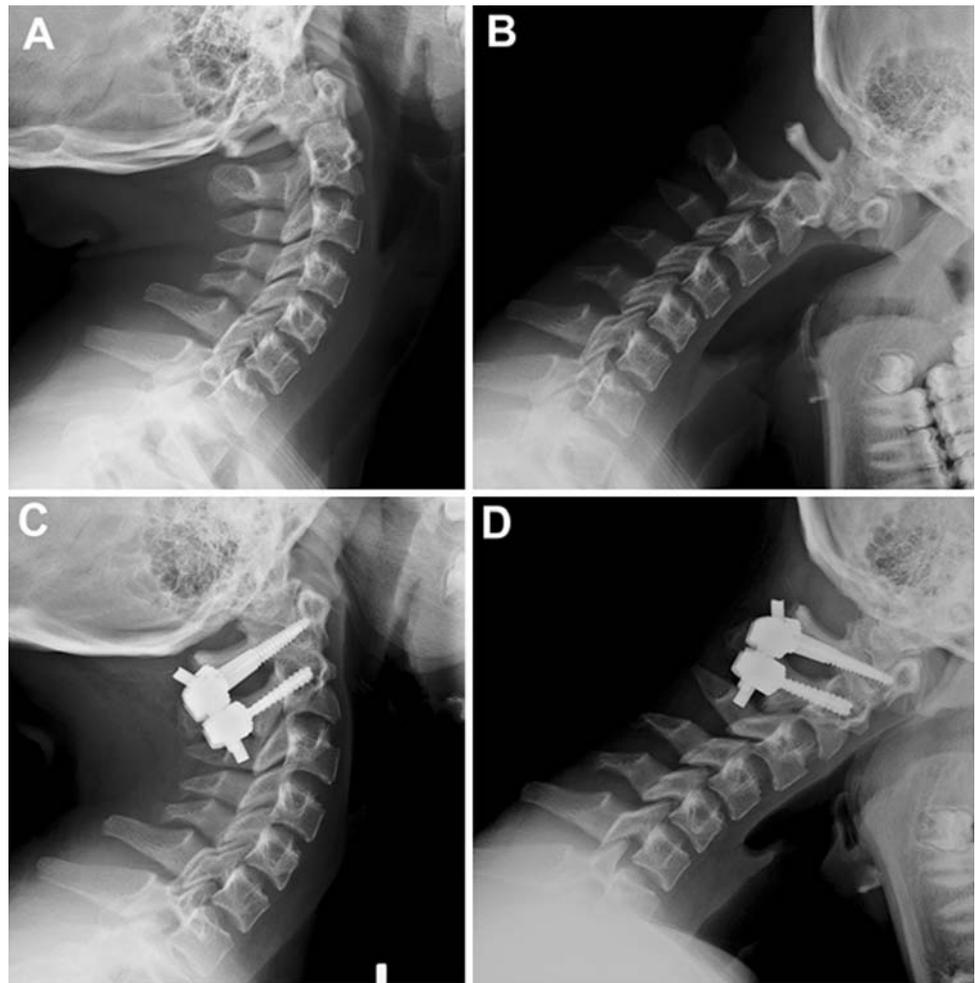


fluoroscopic guidance. All patients had C1 lateral mass screws and 5 of 6 had C2 pedicle screws. In one patient (patient 6) the pedicles were too small and C2 translaminar screws were placed. The placement of the instrumentation, the degree of reduction, and alignment was confirmed intraoperatively with fluoroscopy. Two patients had a C1 laminectomy performed secondary to compression (patients 3 and 4). All patients had post-operative CT scans that demonstrated proper placement of the instrumentation and anatomic alignment.

The patients were all discharged home on post-operative day 3 or 4. They remained in a cervical collar for one month. Post-op CT scans obtained on average 14 months post-operatively (range 7–29 months) confirmed solid

fusion constructs in all patients. All patients underwent follow-up cervical spine imaging within 12 months of surgery. There were two exceptions, one patient died from an unrelated condition prior to 12 months (patient 3) and one patient (patient 6) had imaging at her latest follow-up (7 months). Confirmation of a stable arthodesis was made with CT imaging on all patients requiring a laminectomy and, at minimum, dynamic flexion–extension testing in patients not undergoing laminectomy. There were no instances of iatrogenic vertebral artery injury. Two patients (patients 1 and 2) experienced transient paresthesias in a C2 dermatome that completely resolved within 2 weeks. There were no wound infections or wound dehiscences.

Fig. 2 Flexion–extension films of Patient 1. Pre-operative extension (a) and flexion (b) views demonstrating abnormal motion at C1–2. Follow-up post-operative imaging demonstrating anatomic alignment without motion on extension (c) or flexion (d) views



Representative cases

Patient 1: The patient was a 14-year-old female with no significant past medical history. The patient experienced transient loss of sensation bilaterally to both upper and lower extremities and inability to move after being hit in the back by a wave while in the ocean. The patient described paresthesias throughout her extremities and an abnormal gait, which continued for 48 h.

A computed tomography (CT) scan and magnetic resonance imaging (MRI) of the cervical spine demonstrated separation of the superior aspect of odontoid process from the rest of C2 vertebral body, consistent with an os odontoides (Fig. 1a, b). There was a focal area of increased T2WI signal seen at the C1–2 level with focal narrowing. On flexion–extension there was abnormal motion at C1–2: the distance between the back of the anterior arch of C1 and the body of C2 was 10 mm in the flexed position, reducing to 5 mm in extension (Fig. 2a, b).

The CT scan demonstrated unfavorable anatomy for a transarticular screw bilaterally (Fig. 1c, d). Similar unfavorable anatomy for transarticular screws was seen in three

of the six patients. The patient underwent a Goel–Harms fusion with placement of C1 lateral mass screws and C2 pedicle screws. The patient was reduced in the operating room. The patient had neuromonitoring throughout the OR course without any changes.

Post-operatively the patient was placed in a cervical collar for 1 month. Immediate post-operative imaging demonstrated proper placement of the instrumentation with good reduction and anatomic alignment (Fig. 1e, f). She was discharged home on post-operative day four.

On follow-up, the patient had a solid fusion construct on CT scans and maintained proper alignment on flexion–extension films (Fig. 2c, d). On follow at 29 months the patient had imaging that demonstrated a solid fusion construct. She was without symptoms and had returned to her pre-injury activities including playing basketball on her high school team.

Patient 4: The patient was a 17-year-old female with a history of Trisomy 21. The patient was found to have an os odontoides and had T2 hyperintensity on MR imaging. The canal was narrowed, and therefore a decision was made to perform a laminectomy in the patient in addition to



Fig. 3 Representative radiographic images of Patient 4. **a** Immediate post-operative coronal CT image at the level of the C1–2 joints. Follow-up images performed at over 20 months demonstrating fusion across the C1–2 joints on coronal (**b** and **c**) and sagittal (**d** and **e**) images

a C1–2 fusion. The patient demonstrated a solid bony fusion across the C1–2 joints at follow-up (Fig. 3a compared to Fig. 3b–e). This construct was stable at 20 months, her latest follow-up with imaging.

Patient 6: The patient was a 11-year-old female with no significant past medical history. The patient presented with neck pain and was found to have an os odontoideum (Fig. 4a, f). The CT scan demonstrated unfavorable anatomy for a transarticular screw bilaterally (Fig. 4b, c) and C2 pedicle screws (Fig. 4d). The patient was taken to the operative room where C1 lateral mass screws (Fig. 4h) and C2 trans-laminar screws (Fig. 4i) were placed. Bone was harvested through from the lateral sub-occipital bone with a burr (Fig. 5a) that did not significantly extend the incision (Fig. 5b).

Post-operative imaging demonstrated good reduction (Fig. 4e, l). A 3-D reconstruction demonstrated that compared to a pre-operative scan (Fig. 4j) there was an increase in the canal diameter post-operatively (Fig. 4k). On follow-up at 7 months the patient had imaging that demonstrated a solid fusion construct with bone growth between the lamina and reconstitution of the occipital defect (Fig. 5c, d). This fusion mass represented (Fig. 5d) was typical for all the treated patients.

Discussion

Treatment of atlantoaxial instability can be challenging in the pediatric patient. There are a number of treatment options including: posterior wiring; C1–2 transarticular screws; and Goel–Harms type fusion constructs. Each type of fusion has a unique set of risks and benefits and the choice of technique is related to the anatomy of the patient and their disease process and to the familiarity of the surgeon with any particular technique.

Wiring and cable techniques have a long history of effectively treating problems in the upper cervical spine. However, they are believed to be biomechanically less stable compared to transarticular screw and Goel–Harms constructs [11, 21, 24, 27, 35, 42]. Also, unlike wiring techniques, C1–2 transarticular screws and Goel–Harms constructs do not require that the patient be placed in a halo vest post-operatively. Additionally, C1–2 transarticular screws and Goel–Harms constructs provide stabilization and promotes fusion at just one intervertebral level, the diseased level, and minimize the degree to which it limits the range of motion of a patient.

C1–2 transarticular screws have been shown to be very effective at providing a solid fusion construct in pediatric

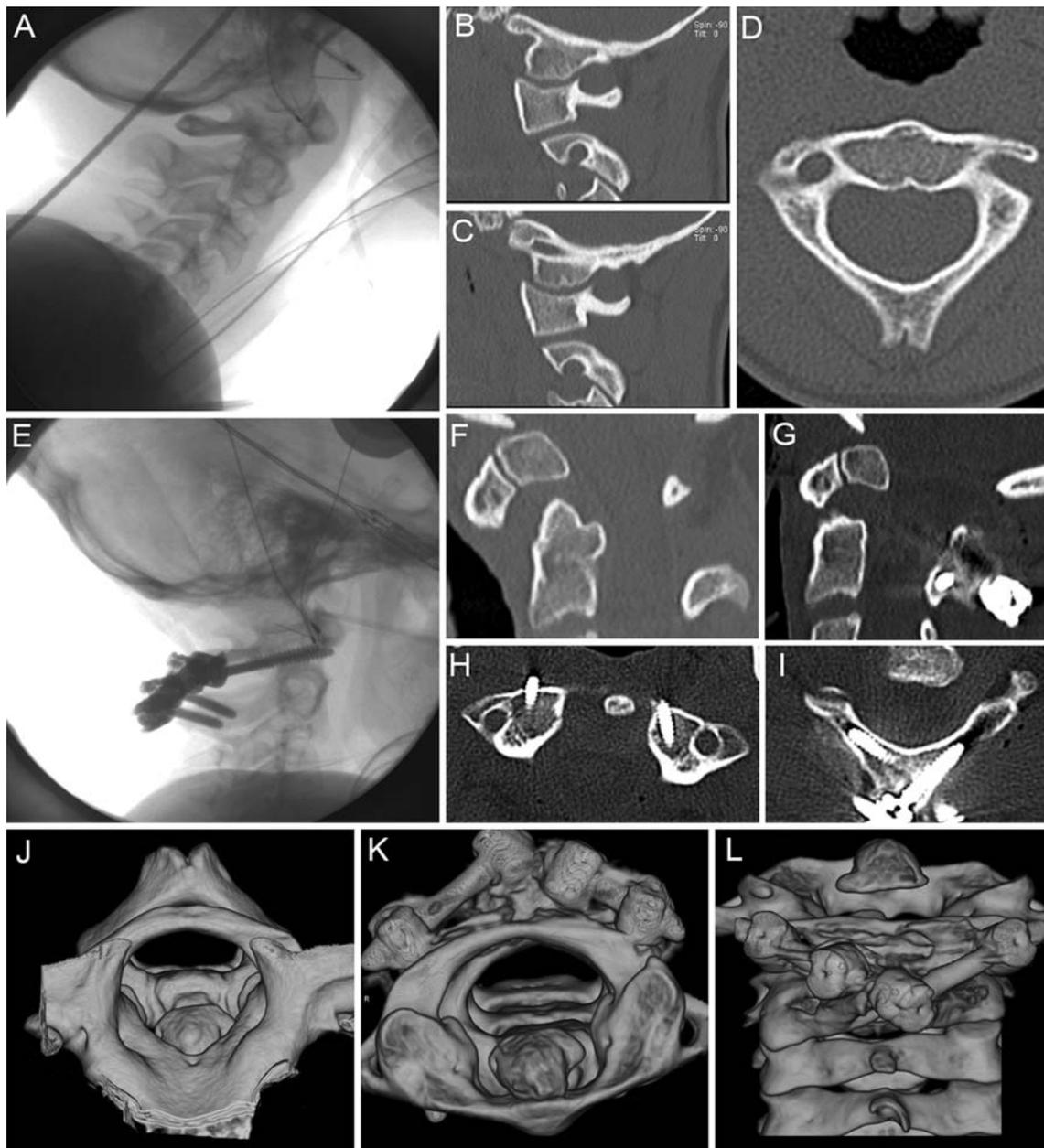


Fig. 4 Representative radiographic images of Patient 6. Pre-operative radiograph (a) demonstrating an os odontoideum that is evident on a pre-operative sagittal CT scan (f). Sagittal images on the right (b) and left (c) an axial scan at C2 (d) demonstrated that the patient's anatomy was not ideal for transarticular screws and or C2 pedicle

screws. A post-operative radiograph (e) and CT scans demonstrate proper reduction (g) and placement of C1 lateral mass screws (h) and C2 transarticular screws (i). 3-D reconstructions demonstrated an increase in the canal diameter from the pre-operative (j) to the post-operative (k) period and a solid construct (l)

patients with atlantoaxial instability [1, 2, 6, 18]. While transarticular screws can be safe and effective choice, their placement is technically demanding and in some patients, there are some potential advantages to the use of a Goel–Harms construct. First, the vertebral arteries have to have a particular anatomy to allow safe placement of C1–2 transarticular screws. If the artery has an unusual course, as has been shown to occur in 10–20% of adult patients, then a

screw can not safely be placed on that side [26, 31, 32, 37]. In one study on pediatric patients, the authors found that the anatomy of the vertebral artery prevented safe placement of a transarticular screw in 11% of joint spaces [6]. The Goel–Harms construct is thought to be less-dependant on the anatomy of the vertebral artery due to greater flexibility in screw trajectory. However, even with Goel–Harms constructs there remains a risk to the vertebral artery.

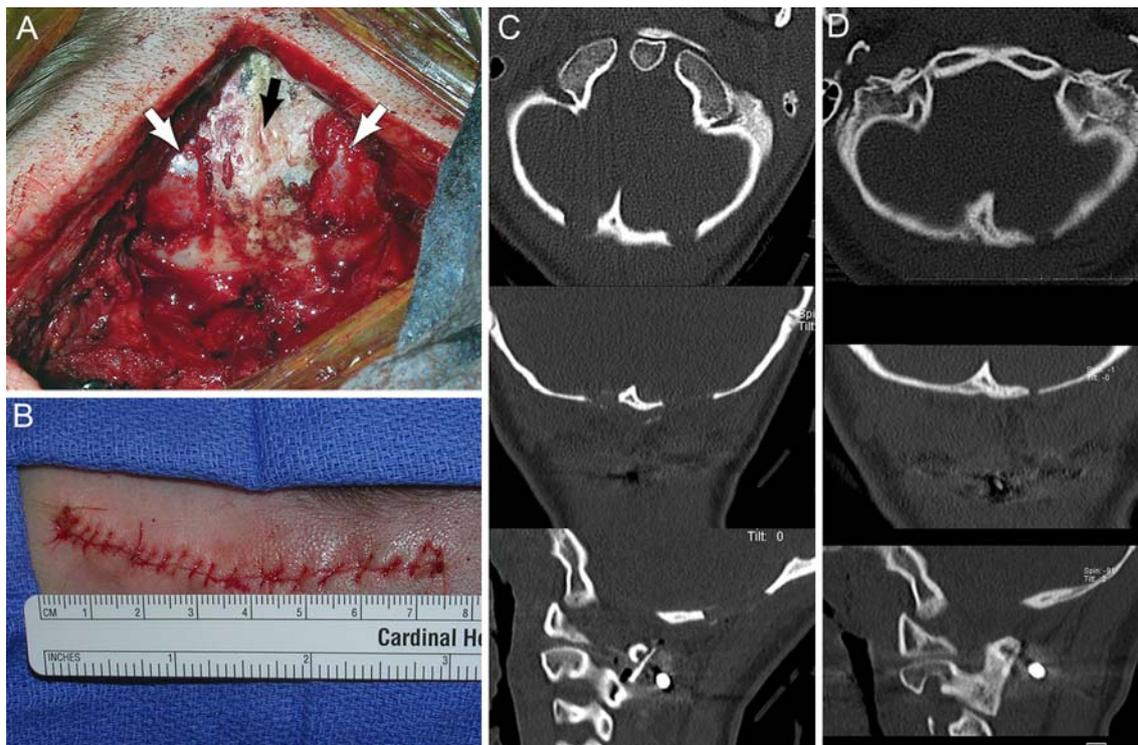


Fig. 5 Intra-operative image demonstrating harvesting bone from the sub-occipital bone (patient 6). **a** Bone was harvested from the lateral sub-occipital bone (*white arrows*) with maintenance of the midline keel (*black arrow*). **b** Typical appearance of a closed wound.

c Immediate post-operative CT images demonstrating the defects from the bone harvest. **d** Follow-up CT images demonstrating reconstitution of the bone harvest site and a solid fusion construct

Brooks or Gallie type fusions require the passage of sub-laminar wires in the cervical spine. Likewise, after placement of transarticular screws, most surgeons augment the construct with a posterior fusion construct that involves passing sub-laminar wires and placement of an autograft taken from the iliac crest. The passage of sub-laminar wires in the cervical spine carries a 7–17% risk of neurologic deterioration, although these risks are felt to be lower at the atlantoaxial level [10, 14, 17, 30, 40, 41]. Also, if the posterior arch of C1 needs to be removed surgically for decompression or if it is incomplete as occurs in 1.5–5% of the population [15, 34], it would be difficult or impossible to place sub-laminar wires. Hypoplasia of the posterior arch of C1 is seen more often in some of the patients commonly treated for atlantoaxial instability [12, 36]. For example, it was seen in 28% of patients with Down syndrome [33]. Because of the need for a C1 arch that will support posterior wiring, patients with congenital absence or a surgically removed C1 arch would not be candidates for transarticular screws or posterior wiring techniques but could be effectively treated with a Goel–Harms construct.

In most studies on C1–2 fusion autograft was taken from the hip or rib [1, 2, 5, 7, 8, 13, 18, 20, 23, 25]. Goel–Harms constructs do not require a structural graft so

hip graft may not be needed. In the six patients presented here no hip autograft or rib was harvested. Instead we describe the use of local bone supplemented with sub-occipital bone if need. Not using hip graft may reduce the overall morbidity of the procedure by avoiding the 10–40% morbidity associated with iliac crest bone harvest [3, 4, 9, 22, 38].

The proper placement of C1–2 transarticular screws requires that any subluxation at C1–2 be reduced prior to passage of the screw and the sub-laminar wires. Some patients cannot be reduced, and therefore could not undergo safe placement of C1–2 transarticular screws or passage of sub-laminar wires. A Goel–Harms construct allows intraoperative reduction of C1 on C2 after the placement of the screws [23].

There are disadvantages to the use of C1 lateral mass screws and C2 pedicle screws or trans-laminar screws. Because more instrumentation is used compared to transarticular screws, these constructs do have a greater cost. Also, compared to transarticular screws, Goel–Harms constructs do have an increased profile. However, in the patients presented here and in the large number of fusion constructs containing C1 lateral mass screws and C2 screws placed by the senior author, we have not had a case

of wound dehiscence or breakdown, even in patients that were treated with radiation.

Conclusions

We describe the relatively safe and effective use of Goel–Harms constructs, both traditional and modified, for treatment of atlantoaxial instability in pediatric patients. Treatment resulted in long-term solid fusion constructs with anatomic alignment in all patients. No autograft was harvested from the hip or rib. Future studies will help to determine which treatment option is ideal for each patient. Also, larger studies with longer-term follow-up will help to determine the overall durability of Goel–Harms constructs in the pediatric patient population.

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