

# Craniovertebral junction injuries in children. A Review

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**Abstract:** The craniovertebral junction (CVJ) is the most complex and dynamic region of the cervical spine. The wide range of movements possible at this region makes it vulnerable to injury and instability. The special anatomical features make children more prone to injuries of CVJ than adults where lower cervical spine is involved more frequently. The classical clinical manifestation in CVJ injury patients are pyramidal signs including weakness and spasticity, stigmata of CVJ anomalies (short neck, low hair line, facial or hand asymmetry, high arched palate, ), torticollis and neck movement restriction. The history of transient loss of consciousness or sudden neurological deterioration following minor trauma may be elicited. Most authors advocate conservative management (in form of immobilization) of CVJ injuries in children as is true in adults. Halo vest provides superior immobilization in upper cervical and CVJ injuries and can be used in a child as young as 1 year of age with minimal difficulty. Early surgical intervention, i.e. within 2 weeks of injury include is indicated in injuries that cannot be reduced and stabilized by external means, partial spinal cord injury with progressive neurological deficit and in children with extradural hematoma.

**Keywords:** Craniovertebral junction, fracture, children

## INTRODUCTION

C1-C2 unit is a complex segment of the cervical spine. It provides an average of 23° of flexion extension and an average of 47° of rotation<sup>1</sup>. Combination fractures of C<sub>1</sub> and C<sub>2</sub> are associated with increased morbidity and mortality than isolated C1-C2 fractures. Altantoaxial region in children is more prone to injuries than in adults where lower cervical spine is involved more frequently. Further birth-related spinal trauma is unique to children and occurs at a frequency of 1per 60,000 births<sup>2</sup>. The most common level of injury is upper cervical followed by cervico thoracic region<sup>3</sup>.

The high cervical region in children has several well described characteristics which predisposes it to injuries<sup>4-9</sup>:

1. Increased ligamentous laxity which allows excessive motion of the spine<sup>10</sup>
2. More horizontally oriented facets that allow excess translational rotation in an antero-posterior

direction.

3. Less mature bone maturation (ossification).
4. Higher fulcrum of cervical movement (C2-C3) (in adults at C5-C6).
5. Higher inertia and torque forces associated with a large head/body mass ratio (which shifts fulcrum hip to up).

Hence spinal cord is most frequently injured in cervical level in all pediatric age groups<sup>4,6,7</sup>. Children also have a higher incidence of complete spinal cord injury without radiographic abnormality (SCIWORA)<sup>11,12,13</sup>. Further, pediatric spinal injuries can present with special problems regarding external spinal immobilization and surgical intervention in a child with significant growth potential.

## EPIDEMIOLOGY

Injuries to the spinal cord and/or vertebral column are relatively uncommon in pediatric population. The incidence ranges from <1 to 10% of all spinal injuries<sup>14,11,15,16,17</sup>. Each year in United States, there are approximately 11,200 new cases of spinal cord injuries of which 1065 involve the children. Several factors differentiate the incidence, type and location of spinal injuries that occur in children compared with those occurring in adults. This variance is largely attributed

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to age dependent anatomical and biochemical features of the spine<sup>14</sup>. Young children suffer a greater proportion of cervical injuries, particularly injuries involving the upper cervical spine and cranio-vertebral junction (CVJ)<sup>15,17,18</sup>.

Fractures of the odontoid process of the axis accounts for 10-20% of all cervical spine fractures<sup>19,20,21</sup>. Among these, the Anderson and D'Alonzo type II fractures which occur at the junction of odontoid process and body of the axis form the most common type of odontoid process fracture<sup>22</sup>. The most common level of injury is upper cervical and then cervicothoracic<sup>3</sup>. Mackinnon et al described 22 neonates with birth related spinal cord injuries (SCI)<sup>3</sup>. The diagnosis was defined by the following criteria: Clinical evidence of acute cord injury for at least one day and evidence of acute cord injury, spinal cord or column injuries by imaging or electrophysiological studies. Fourteen neonates had upper cervical injuries, 6 had cervicothoracic injuries and 2 had thoraco-lumbar injuries. All upper cervical injuries were associated with cephalic presentation and the use of forceps for rotational maneuvers. Cervicothoracic injuries were associated with breach presentation..

Combination fractures of C1-C2, as already mentioned are relatively common. The occurrence of a concurrent C<sub>1</sub> fracture in presence of type II or III odontoid fractures has been reported in 5-53% of cases<sup>23</sup>. Green et al reported incidence of combination fractures of C1-C2 as 14%<sup>23</sup>.

### Etiology

Vehicular trauma is the most common cause of upper cervical injuries (56%), falls (17%) and are twice as frequent in young children. Athletic and sports related injuries (wrestling, football, diving, gymnastics etc.) comprise 13% (more common in old children). Penetrating injuries account for 4% of all spinal injuries<sup>24,25</sup>. Odontoid epiphysiolysis is typically seen in children <7 years. The neurocentral synchondrosis, which may not fuse completely until the age of 7 years represents a vulnerable site of injury in young children<sup>26</sup>. Birth injuries are a known cause for SCI (spinal cord injury) in neonates (6%)<sup>24</sup>. Menticoglou et al reported 15 neonates with birth related upper cervical spine injuries<sup>27</sup>. All were associated with cephalic deliveries requiring rotational maneuvers with forceps.

### Signs and symptoms

The craniovertebral junction which comprises of the basi-occiput, atlas, axis and their supporting ligaments constitute the most complex and dynamic region of the cervical spine. The wide range of movements possible at this region makes it vulnerable to injury and instability.

Osenbach et al in a study comparing younger (0-8 years) and old children [9-16years] found variation in cause, distribution, type of injury and severity of neurological injury<sup>24</sup>. They found that cervical spine is most frequently injured and accounted for in 63% the injured children, and upper cervical spine and CVJ were more frequently injured than lower cervical spine. Younger children sustained a higher percentage of cervical injuries (79%) than older children (54%). Furthermore, upper cervical and CVJ injuries were twice as frequent in young children. Lower cervical injury occurred with some frequency in both the groups. Neurological deficit was twice more frequent in upper cervical as compared to lower cervical injuries. The classical clinical manifestation in CVJ injury patients are pyramidal signs including weakness and spasticity, stigmata of CVJ anomalies (short neck, low hair line, facial or hand asymmetry, high arched palate), torticollis and neck movement restriction. The history of transient loss of consciousness or sudden neurological deterioration following minor trauma may be elicited..

Neurological disability caused by compressive myelopathy was graded by Kiran Kumar et al<sup>28</sup> while describing surgical management of remote, isolated type II odontoid fractures as:-

- I. Neurologically intact (hyper-reflexia with mild spasticity).
- II. Independent with minor disability
- III. Partially dependent for daily needs and
- IV. Totally dependent.

And the atlantoaxial dislocation (AAD) was classified as reducible or irreducible depending on the change in atlanto-dental interval on flexion and extension.

Irreducible Atlanto axial dislocation caused by:

- A. Malunited fracture of the odontoid.
- B. Fixed anterolisthesis of the anterior arch of C<sub>1</sub>-fractured odontoid complex.

C. Fixed retrolisthesis of anterior arch of a C<sub>1</sub>-fractured odontoid complex.

Reducible AAD was further classified as caused by mobile AAD and hypermobile AAD<sup>28</sup>.

Scott et al<sup>14</sup> in a study on treatment of atlanto occipital instability in pediatric patients categorized the CVJ injuries in 4 broad categories.

1. Atlantoaxial rotatory subluxation.
2. Atlantoaxial ligamentous instability.
3. Traumatic Atlantoaxial fracture.
4. Atlanto-occipital dislocation.

Atlantoaxial fixed rotatory subluxation is more common during childhood. It can present after minor trauma, in association with an upper respiratory tract infection or without an identifiable inciting event. The head is rotated to one side with the head tilted to other side causing the so called Cock Robin appearance. The child is unable to turn his head past the midline. Attempts to move the neck are often painful. The neurological status is almost always normal<sup>29,30,31,32</sup>. Fielding and Hawkins<sup>33</sup> described 17 children and adults with atlanto axial rotatory subluxation and classified their dislocations into 4 types based on radiographic features.

**Type-I:** Most common, unilateral anterior rotation of the atlas pivoting around the dens with a competent transverse ligament.

**Type-II:** Described as unilateral anterior subluxation of the atlas pivoting on contralateral C<sub>1</sub>-C<sub>2</sub> facet. The Atlanto-dens interval is not increased to >5mm.

**Type-III:** Described as anterior subluxation of both C<sub>1</sub> facets with an incompetent transverse ligament.

**Type-IV:** Posterior displacement of C<sub>1</sub> relative to C<sub>2</sub> with an absent or hypoplastic odontoid process.

Lui et al<sup>34</sup> described nine children with ligamentous injuries resulting in AAD and unlike children with traumatic dens injuries who can be managed with closed reduction and immobilization, these children required surgical stabilization and fusion.

Osenback et al<sup>24</sup> categorized upper cervical injuries in 4 groups :-

1. Vertebral body and/or posterior element fracture alone (42%)

2. Fracture combined with subluxation (28%)
3. Subluxation without evidence of bony fracture (11%)
4. SCIWORA (19%).

Nondisplaced fractures were more frequent in older age group whereas subluxation without bony fracture and SCIWORA occurred with higher frequency in young children. All patients with subluxation alone (ligamentous injury mainly) had only cervical spine involved and 84% sustained concomitant systemic injuries that included closed head injuries (20%), abdominal injuries (8%), long bone fractures (8%) and blunt thoracic injuries (4%). There was 60% incidence of head injury among multiple trauma victims.

Dickman et al suggested a 12% incidence of neurological deficit for C1-C2 combination fractures compared with 0% and 2% incidence for isolated atlas and axis fractures respectively.

Injuries involving some degree of subluxation with or without fracture are more likely to result in neurological injury than non displaced fractures. The inherent hyper mobility and elasticity of pediatric spine allows transient subluxation to occur after which elastic recoil returns the spine to a relatively normal anatomical alignment. This is one theory that explains the phenomenon of SCIWORA<sup>35,36</sup>.

SCIWORA is associated with a high incidence of complete neurological injury. Children with SCIWORA develop delayed neurological injury (upto 2 days) after what is considered a trivial trauma<sup>12,13,37,38</sup>. Once initiated, there is often a rapid evolution to severe and irreversible neurological injury. Delayed presentation has been reported in as high as 67% children with SCIWORA<sup>13</sup>.

In fracture odontoid inadequate immobilization with fibrous non-union occurs. As time passes, anterior subluxation occurs with increasing pain and muscle spasm in the neck. With progression there is asymmetry in the subluxation of the condylar complexes causing a rotatory dislocation that results in the head being rigidly held in a slightly flexed position and rotated to one side<sup>39</sup>. The significant step that develops between the cranial base and the spine of C2 becomes clinically obvious and palpable. As the compression progresses, the posterior arch of the atlas impinges up on the posterior columns of the spinal cord resulting in numbness and tingling in the fingertips and then in all the extremities. The spinal

motor tracts are eventually affected because of either compression or repeated episodes of microtrauma from recurrent subluxation during flexion and extension<sup>25</sup>. Any motor signs and symptom combination can result, ranging from unilateral monoparesis, progressing to cruciate paralysis, tetraplegia and respiratory failure (30%). The pathogenesis of the neurological deficit though not clear, seems to be due to repeated microtrauma to the cord (which occurs due to repetitive early dislocation), which leads to cord degeneration<sup>40-42</sup>.

## MANAGEMENT

After receiving a child or adult with trauma, physical examination is important in aiding diagnosis and in a patient with suspected spinal trauma, primary goal should be to achieve immobilization of the spine along with ensuring adequate airway, ventilation and perfusion. Spinal immobilization prevents vertebral column and spinal cord from further injury during imaging or transportation to a specialized centre. In children <8 years, head is relatively larger as compared to torso which forces the neck into a position of flexion when the head and torso are supine on a flat surface<sup>43</sup>. Nypamer and Treloar<sup>43</sup> in their series of 40 children found that all children below 8 years required elevation of the torso (mean elevation of 25mm) to eliminate positional neck flexion to achieve neutral position. In separate report, they found that semi rigid cervical collars placed on children younger than 8 years did not prevent this positional forced flexion when placed supine on standard, rigid spinal boards<sup>44</sup>.

Huerta et al concluded that no collar provided acceptable immobilization when used alone<sup>44</sup>. They found that the combination of a modified half spine board, rigid cervical collar and tape was the most effective means of immobilizing cervical spine for transport in children. Shafermeyer et al cautioned that taping across the Torso to secure the child to the supine board may have deleterious effect of child's respiration<sup>45</sup>. Pang and Hanky provided the only description of external immobilization device for neonates. They described a thermoplastic molded device that is contoured to the occiput, neck and thorax<sup>46</sup>.

## Imaging

As children can suffer multiple level injury, evaluation of entire spinal axis is essential<sup>24</sup>. The initial evaluation of C<sub>1</sub>-C<sub>2</sub> injury begins with obtaining plain X-ray films<sup>14</sup>.

Fracture or non diagnostic findings on plain radiographs are further delineated by either thin section CT<sup>24</sup>. All patients with neurological deficit undergo MRI scans to exclude an acute surgical lesion like EDH or herniated disc although the later is extremely uncommon in children<sup>47</sup>. CT-myelography is an alternative to MRI, if it not available<sup>48</sup>. Laham et al<sup>49</sup> while evaluating the role of cervical spine X-ray evaluation in children with apparent isolated head injuries categorized the children into *High risk* and *Low risk* groups. High risk were incapable of verbal communication either because of age (below 2 years) or head injury, and those who had neck pain. He used three view approach of antero-posterior, lateral and open mouth X-rays and discovered no cervical spine injury in low risk group while 70.5% in the high risk group. He concluded that cervical spine X-rays are not necessary in children with isolated head injuries who can communicate and have no neck pain or neurological deficit. Bohn et al emphasized that unexplained hypotension or absent vital signs in pediatric trauma patients are likely to result from a severe cervical cord injury and should be evaluated properly<sup>50</sup>.

Viccellio et al<sup>51</sup> evaluated the cervical spine in children younger than 18 years using NEXUS criterion (National Emergency X-radiography Utilization Studies). They used five low risk criteria viz. (1) The absence of midline cervical tenderness, (2) Evidence of intoxication, (3) Altered level of alertness, (4) Focal neurological deficit and (5) A painful distracting injury. X-rays were obtained at the direction of the physician (treating). A minimum of 3 views were obtained. The child was considered to be at low risk if all the 5 criteria were absent. If anyone of the criteria was present, the child was considered to be at high risk. They found that none of the children in low risk group had a documented cervical spine injury by radiographic evaluation. 0.98% injuries were documented in children not fulfilling the low risk criteria. The author concluded that applying NEXUS criteria to children would reduce cervical spine X-ray use by 20% and would not result in missed injuries.

The use of open mouth X-ray has been questioned by many authors in children. Swischuk et al in their study calculated a missed fracture rate of 0.007 per year per radiologist on lateral X-rays and concluded that open mouth X-ray might not be needed routinely in children less than 5 years<sup>52</sup>. Similarly Buhs et al also concluded (in a separate study) that open mouth X-ray is not necessary for clearing the cervical spine in children young

than 9 years<sup>9</sup>.

Scarrow et al failed to demonstrate any usefulness of evoked responses, flexion-extension fluoroscopy or MRI of the cervical spine in the evaluation of the cervical spine in children with altered mental status after trauma<sup>53</sup>. Ralston et al<sup>54</sup> and Dwek and Chung<sup>55</sup> in two different retrospective studies on use of flexion/extension radiography (cervical) after obtaining a static cervical X-ray, concluded that the use of flexion-extension X-rays after obtaining normal static (neutral) cervical X-ray is of questionable use. Fairholm et al<sup>25</sup> in a study on management of delayed neurological symptoms in fracture odontoid mentioned that flexion extension views will analyze the immediate reducibility of the dislocation.

Kiran Kumar et al<sup>28</sup> established the diagnosis of fracture odontoid and reducible and irreducible AAD by transtable lateral X-rays and axial and sagittal reconstructed intrathecal contrast CT scans of the CVJ in flexion and extension positions of the neck. They used MRI scans of the cervical spine in nine patients to assess the extent of cervico-medullary compression and the cord intensity changes on T<sub>2</sub>-weighed image.

Young children as compared to adults have certain X-ray features which are considered normal in them, these include pseudosubluxation of C2 on C3, over riding of the anterior atlas in relation to the odontoid on extension, exaggerated atlanto-dens intervals (ADI), and the radiolucent synchondrosis between the odontoid and C2 body<sup>56</sup>. These normal findings can be mistaken for acute traumatic injury in children after trauma. To differentiate between a traumatic and physiological subluxation, a method is recommended which involves drawing a line through the posterior arches of C1 and C3. In pseudosubluxation of C2 on C3, the C1-C3 lines should pass through, touch or be upto one mm anterior to the anterior cortex of the posterior arch of C2. If it lies 2mm or more behind the line then true subluxation should be assumed (not pseudosubluxation)<sup>57</sup>.

In odontoid epiphysiolysis in children, investigation of choice is lateral cervical spine X-ray which will often reveal the odontoid process to be angulated anteriorly and rarely posteriorly<sup>58</sup>.

## Treatment

Most authors advocate conservative management of spinal cord injuries in children as is true in adults<sup>24</sup>. Halo vest provides superior immobilization in upper

cervical and CVJ injuries and can be used in a child as young as 1 year of age with minimal difficulty. Custom moulding brace<sup>24</sup> has been used especially for lower cervical injuries. Indications for early surgical intervention, i.e. within 2 weeks of injury include<sup>24</sup>:

1. Injuries that cannot be reduced and stabilized by external means
2. Partial spinal cord injury with progressive neurological deficit
3. Extradural hematoma and
4. Herniated discs.

Recent data suggest that methylprednisolone in high doses may be beneficial in spinal cord injuries if administered within 8 hours of injury<sup>59</sup>. In NASCIS II trial patients were randomly assigned to receive a 24-hour infusion of methylprednisolone, naloxone or placebo within 12 hours after acute spinal cord injury. Again, there was no benefit overall in the methylprednisolone group; however, post hoc analyses detected a small gain in the total motor and sensory score in a subgroup of patients who had received the drug within 8 hours after their injury. As a result, this 24-hour, high-dose methylprednisolone infusion, if started within 8 hours after injury, quickly became an implied standard of care despite considerable criticism of the validity of such a post hoc analysis.

Various management techniques especially for C1-type II odontoid fracture include<sup>23</sup> semi rigid immobilization (collar), traction and then immobilization in a brace, rigid immobilization (Halo, Minerva, SOMI (Sub occipital Mandibular Immobilization), posterior fusion and anterior odontoid screw fixation. Dickman et al in a series of management of acute C1-C2 combination fractures used non operative therapy as initial management strategy in 84% patients<sup>60</sup>. Out of 25 patients, 18 were placed in Halo orthosis and two in SOMI brace for a median duration of 12 weeks. Four patients were treated by early surgical stabilization and fusion based on an atlanto-dental interval of 6mm or more. Three were treated with posterior C1-C2 wiring and fusion. One among Halo treated group failed to stabilize and posterior C1-C2 fusion was performed. All surgically treated patients achieved stabilization. The authors stressed that atlas fracture associated with type II and III odontoid fracture with an ADI equal to or exceeding 5mm should be considered for early surgical

management. In one series 6 patients with C1--type II odontoid fracture (with ADI less than 6mm) were treated with rigid immobilization using halo vest. One patient failed to stabilize and needed posterior C1-C2 fusion at 12 weeks post injury. Four patients with same type of combination fracture were treated with early surgical fusion based on an ADI of 6mm or more. Three were treated with posterior C1-C2 fusion and one patient underwent occipito-cervical fusion for multiple fractures of the posterior atlantal arch. Five patients having C1-type III odontoid fracture were successfully managed by halo immobilization for an average period of 12 weeks. Three patients had C1-Hangman combination fracture and were treated successfully with either a halo or SOMI device. Seven cases having C1-miscellaneous C<sub>2</sub> body combination fracture, were successfully treated with either halo or SOMI brace.

Lee et al performed C1-C2 posterior fusion in two patients with C1-type II combination fracture successfully<sup>61</sup>. Henry et al described 90% success rate by anterior odontoid screw fixation in 10 of his patients with same type of combination fracture<sup>62</sup>. Occipitocervical fusion has been reserved for patients with disruption of C1 arch and gross C1-C2 instability with type II odontoid fracture<sup>23</sup>.

Most reported combination fracture of C<sub>1</sub> and Hangman types have been managed conservatively with immobilization. Fielding et al<sup>63</sup> recommended that fracture of this type with angulation between C2-C3 of 11° or more be treated surgically as this type of combination fractures with equal to- or exceeding 11° angulation between C2-C3 were associated with a nonunion rate of 85% or more.

Crockard et al described the use of transoral decompression (in fracture odontoid) for myelopathy caused by odontoid fracture and recommended that this be used if reduction fails to relieve the condition<sup>64</sup>. Fairholm et al while describing the management by odontoid fracture found in most patients that even though there is marked dislocation that does not reduce in extension, a period of traction is beneficial<sup>25</sup>. Most patients will slowly reduce over a period of 10-14 days of traction applied in slight extension. The axis of the extension should be around the axis of the external auditory canal and not through long cervical vertebra. This is facilitated by placing a large soft roll under the neck and allowing gentle traction with 2-6 kg weight. In those patients in whom reduction is achieved either in

extension or after traction, a simpler transarticular C1-C2 posterior fusion is performed. In those who remain unreduced after 10 days of traction, a transoral resection of the odontoid and upper 2/3<sup>rd</sup> of the body of C<sub>2</sub> is required. External support is ensured to facilitate bony healing for which halo vest immobilization is considered a standard now (after fusion). With transarticular fusion, only a simple Philadelphia collar is necessary.

Mandabach et al found 8 out of 10 children having odontoid injuries were successfully treated with halo-immobilization alone<sup>65</sup>. They concluded that as the injury occurs through the epiphysis, it has a high likelihood of healing if closed reduction and immobilization are used. The average time to fusion was 13 weeks. Reduction was obtained by application of halo device under ketamine anesthesia and then realignment of the dens utilizing C-arm fluoroscopy. Reinges et al noted that only three young children have been reported in literature that had odontoid injuries primarily treated with surgical stabilization<sup>66</sup>. Wang et al described using anterior odontoid screw fixation as the primary treatment option in a 3 year old child with C2 epiphysiolysis<sup>67</sup>. A hard cervical collar was used postoperatively.

Atlantoaxial rotatory subluxation, more commonly seen in children has been successfully treated conservatively by manual reduction and immobilization in hard collar or halovest for period ranging from 4-10 months<sup>68</sup>. Surgical orthodesis can be considered for those with irreducible subluxations, recurrent subluxation or subluxations present for more than three weeks<sup>56</sup>.

Unlike children with traumatic injury to dens who can be managed with closed reduction and immobilization, children with AAD due to ligamentous injury require surgical stabilization and fusion. Lui et al attempted to treat two such children with halo immobilization for three-month-duration; both attempts failed to achieve stability (all nine children in their series needed surgical stabilization)<sup>34</sup>.

Further concerns of cervical traction in children are because of their relatively thinner cranium with a higher likelihood of inner table penetration, lighter body weight that provides less counterforce to traction. More elastic ligaments and less developed musculature, increasing the potential for over distraction. The placement of bilateral pairs of burr holes (parietal) and passing 22-gauge wire through them to provide a point of fixation for traction

has been described for infants with cervical spinal injuries<sup>56</sup>.

In patients with malunited fracture of odontoid, a simultaneous transoral decompression (TOD) and posterior fusion were performed by Kiran Kumar et al while treating remote isolated type II odontoid fracture. They used modified Brooke's method in 17 of their patients which consists of uniting the decorticated posterior arch of the atlas and lamina of the C2 by use of a central sublaminar braided wire without any intervening bone graft and the placement of onlay bone graft on the C<sub>1</sub>-C<sub>2</sub> arch that were held in apposition by the lateral sublaminar stainless steel wires, and the creation of notches on the posterior arch of C1 and lamina of C2 to hold the wires in place<sup>28</sup>. In one patient who had extremely thin posterior arch of atlas (C1), the modified Jain technique of occipito-cervical posterior fusion was applied after excision of the posterior arch of the atlas. This consisted of creating an artificial arch on the occipital bone 1cm superior to the posterior margin of the foramen magnum and fusing it to the lamina of the axis by use of sublaminar wirings and wedge and onlay bone grafts. In one of their patients with hypermobile AAD, the Ransford technique of posterior fusion was used. This involved fixing of a pre-moulded stainless steel loupe to the occiput and the laminae of the upper cervical spine by use of stainless steel wire. Onlay autologous rib grafts were placed between the occiput, C1 and C2 to secure a bony fusion. Early walking was encouraged in all patients with neck movement stabilized by one of the hard cervical collar for at least 3 months.

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