Pros and Cons of C2 Nerve Sectioning/Preservation in Posterior Fusion for Congenital Atlantoaxial Dislocation

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OBJECTIVE: Deliberate C2 nerve root sectioning and its preservation have been described during posterior fusion for atlantoaxial dislocation (AAD). However, the associated outcomes have been less studied, especially in congenital AAD. Our objective was to study the clinical outcomes after C2 nerve root sectioning and the feasibility of C2 preservation in these patients.

METHODS: The data from 190 patients were retrospectively studied. The decision to cut or preserve the C2 nerve root was determined by the preoperative radiologic findings and intraoperative suitability of its preservation. During follow-up, the patients were questioned about C2 nerve-related dysfunction. The pros and cons of sectioning or preservation of the C2 nerve root and the related feasibility factors are also described.

RESULTS: Complex C1-C2 anatomy that required extensive dissection and drilling was seen in 139 patients. After C2 nerve root sectioning in 178 patients, none reported bothersome hypoesthesia, paresthesia, or dysesthesia that required medication. However, 9 patients (5.1%) developed nonhealing occipital ulcers and most required flap coverage or a skin graft. C2 nerve root preservation is feasible with an adequate inferior C1 lateral mass and normal-size ganglion. In patients with C1—occipital condyle hypoplasia, extremely oblique joints, spondyloptosis, incurved occiput, pseudo-facets, and anomalous vertebral artery, preservation of the C2 nerve root is difficult.

CONCLUSIONS: The advantages of sectioning the C2 nerve root are many. However, a subset of patients is prone to morbid occipital neuropathic ulcers. After C2 nerve sectioning, one should be cautious of such complications. C2 nerve root preservation should be strongly considered for patients with favorable anatomy.

INTRODUCTION

Posterior short segment fusion using the C1 lateral mass and C2 pedicle screws has steadily evolved to become the preferred treatment of atlantoaxial dislocation (AAD), regardless of its etiology.¹,² Recently, techniques have been described in which comprehensive drilling of the C1-C2 joints is performed to correct the dislocations in various planes.¹,² These are technically demanding because of the array of complex congenital variations in the C1-C2 bony morphology and the adjacent vertebral arteries (VAs) that can show an anomalous course in approximately 12%—20% of patients.³ Hence, a clear visualization of the entry sites for the C1-C2 screws and the joints is an absolute necessity before proceeding with any sort of instrumentation for instability of craniovertebral junction.

When accessing the C1 lateral mass, the C2 nerve root ganglion is a major limiting factor that restricts adequate visualization of the facets. It can also be bulky with an extensive perineural venous plexus. Some surgeons prefer to intentionally sacrifice the C2 ganglion to adequately visualize the C1 facet and insert screws.
under direct vision. This step, although it simplifies the procedure, is not without disadvantages such as postoperative paresthesia and dysesthesia along the nerve distribution. The functional outcomes after C2 nerve root sectioning have been studied by few investigators. Most patients in these series had C1-C2 instability resulting from a noncongenital etiology such as trauma and acquired causes. In such cases, the C1-C2 anatomy can be expected to be relatively simple compared with the congenital cases. The extent of surgical exposure and the degree of facet joint remodelling needed will be less than required for those with congenitally deformed joints. Consequently, the threshold to preserve or sacrifice the nerve root to achieve panoramic surgical visualization is likely to differ between the congenital and acquired cases.

We studied the feasibility of preservation of the C2 nerve root and the long-term outcomes resulting from C2 nerve root sectioning in patients undergoing posterior C1-C2 fusion for congenital AAD.

METHODS

From 2012 to 2017, 190 patients (aged 12—73 years) with congenital AAD who had undergone posterior C1-C2 fusion in our institute were included. Children younger than 12 years were excluded. In the initial 3 years, the C2 nerve root was deliberately sacrificed for ease of exposure. However, we observed that a few patients developed neuropathic ulcers in the occipital area that were difficult to manage. With a gradual learning curve, for patients undergoing surgery during the latter 2 years, a conscious effort was made to preserve the C2 ganglion whenever feasible. The decision to preserve or section the C2 nerve root was at the discretion of the surgeon according to the preoperative imaging and intraoperative findings. The computed tomography (CT) scans and CT angiograms (CTAs) were studied to determine the presence of complex bony and vascular anomalies that might preclude successful preservation of C2 nerve root owing to the lack of adequate joint visualization and C1 screw placement. The factors evaluated included assimilated C1, hypoplasia of the C1—occipital condyle complex, incurring of the occipt, pseudofacets, and the severity of deformed joints on the CT bone window. The CTA was studied for the presence of an anomalous VA. Intraoperatively, the posterior vertical height of the C1 lateral mass was assessed after drilling a notch underneath the C1 pedicle (the junction of the C1 posterior arch to its lateral mass) to determine the suitability of screw and plate placement. The size of the C2 ganglion was also evaluated. A large ganglion necessitated sectioning to widen the surgical corridor. The details are presented in Discussion section. In most of the patients (n = 178), the C2 nerve root had to be sacrificed, with its preservation successful only in 12.

Surgical Technique

After placing the patient supine in skeletal traction, the C1 and C2 were exposed through a midline skin incision and subperiosteal dissection. The C1 lateral mass and C2 isthmus were defined. The C2 ganglion or nerve root was identified bilaterally, and the periureal venous plexus was coagulated and cut. Care was taken to not sever the nerve root close to the spinal cord to avoid a cerebrospinal fluid leak. Before coagulating the venous plexus, we confirmed from the preoperative CTA that the patient did not have an anomalous VA lying close to the ganglion. Dividing the C2 nerve roots helped with the panoramic visualization of the C1-C2 joints and easy placement of the facet screws. In the patients in whom the C2 nerve root was intended to be preserved, the nerve root was dissected and mobilized. Next, the root was gently retracted to dissect the joint space. The C1-C2 joints were then opened, cartilage along the surfaces was excised, and comprehensive drilling was performed. The screws (C1 lateral mass and C2 pedicle) were then inserted, and spacers were placed in the joint cavity and fastened with Goel’s plate/polyaxial screws and rods. Reduction was achieved in multiple planes by rod manipulation, the details of which have been previously described. Postoperatively, the neck was immobilized in Philadelphia collar for 3 months.

Follow-Up and Assessment

At regular (1.5-, 3-, and 6-month) intervals, the patients underwent routine clinical and radiologic assessments. Thin-slice sagittal and coronal reconstructed CT scans were examined at 6 months and 1 year to determine the presence of bony fusion and maintenance of reduction. The presence of bony trabeculae between the C1 and C2 facets or between the lamina and arch on the CT scan without any gap between them (and with no mobility on flexion—extension radiographs) indicated fusion. In the presence of metallic spacers, particular attention was given to the areas that were lateral, anterior, and posterior to the spacers. Cystic lucencies around the implants or along the endplates and linear defects within the bridging trabeculae suggested the presence of nonfusion.

Additionally, at their latest follow-up examination, the patients were questioned regarding possible C2 nerve-related symptoms. This evaluation was based on variables such as hypoesthesia, paresthesia, dysesthesia, and neuralgia over the area of the C2 nerve distribution. To assess the degree of involvement, the findings were then graded using a 10-point scale (0—10). Any requirement for medication for C2 nerve-related symptoms was also recorded. The patients were further questioned whether the symptoms, if any, had disturbed their daily activities.

RESULTS

All the patients had congenital AAD with subluxation in the anteroposterior, vertical, rotatory, or lateral plane or a combination of these. Of the 190 patients, 124 were male and 66 were female. In 139 patients (73.2%), the C1-C2 anatomy was complex with highly deformed oblique joints. The remaining had relatively less oblique joints and a robust C1 lateral mass. Of these, 20 underwent surgery during the latter 2 years of the study period, and we were able to preserve the C2 nerve root in 12 of them.

The AAD was irreducible in 138 patients and reducible in 52 patients. Bony abnormalities were more often seen (89.9%) in the irreducible AAD group, and os odontoideum (46.2%) was commonly associated with the reducible AAD group. Most patients (82.3%) with irreducibility had an assimilated C1 arch. Most also had an associated decreased vertical height of the C1 lateral mass due to hypoplasia of the C1—occipital condyle complex. The joints were oblique in patients with irreducible AAD compared with those
C2 Nerve Dysfunction

None of the patients who underwent C1-C2 fixation after sacrifice of the C2 nerve root presented with spontaneous complaints of nerve dysfunction. On questioning, 54 patients (30.3%) reported hypesthesia over the C2 dermatome (mean score, 0.84). Paresthesia and dysesthesia were noted in 39 (21.9%) and 34 (19.1%) patients, respectively. The corresponding mean scores were 0.72 and 0.56. None of the patients reported that their daily activity had been affected by these symptoms, and none required any specific medications. At subsequent follow-up visits, the sensory disturbances remained the same and did not require any intervention. The data are summarized in Table 1.

Of the 190 patients, 9 (5.1%) developed an occipital neuropathic ulcer (Figures 1 and 2). Of these, 3 were challenging to treat and required wound coverage in the form of flap rotation or advancement by a dedicated plastic surgery team (Figures 1 and 2). In 2 patients, the defect was covered with a split-thickness skin graft. In 4 patients, the ulcers were superficial and healed with expectant management.

Two patients in whom C2 nerve root preservation was attempted but required sacrifice developed monoparesis of upper limb, possibly a result of root manipulation during dissection and screw insertion. However, they improved during the follow-up period.

C2 Nerve Preservation

Because of the occipital ulcer formation in some of our patients, we tried to preserve the C2 nerve root whenever feasible. In 12 patients, C2 nerve root preservation was possible (Figure 3). Of these 12 patients, 10 had reducible AAD with an intact C1 arch and relatively flat joints. Also, the robust inferior lateral mass of the C1 and normal course of the VA facilitated C1 screw insertion without sectioning the nerve root. In 2 patients with irreducible AAD, the C1 lateral mass was occipitalized; however it was well formed with no associated anomalous VA. In these patients, the C2 nerve root preservation was feasible. None of the 12 patients with C2 nerve root preservation experienced C2 nerve-related pain.

Apart from these 12 patients, we had 3 other patients in whom the C2 nerve root could be mobilized and the joints drilled and manipulated. However, kinking of the nerve root occurred after tightening the Goel plate, and it had to be sacrificed. In 2 others, excessive traction on the C2 nerve root occurred during drilling of the joint and screw insertion that necessitated sacrifice. These 2 patients developed transient monoparesis of upper limb as described.

DISCUSSION

Generally, the workspace around the C1-C2 joint is constrained. The wide range of associated bony abnormalities, deformed joints, and VA anomalies further add to the surgical complexity. One such important structure is the C2 ganglion, which is situated in the posterior surface of the C1-C2 joint. It lies in the intervertebral space bounded superiorly by the posterior arch of C1 and inferiorly by the C2 lamina. Anatomic studies have shown that the C2 ganglion occupies almost three fourths of the foraminal height of C2.14

Using a posterior surgical approach to access to this area, the C2 ganglion can hinder adequate visualization of the C1 facets. To overcome this difficulty, the C2 ganglion is often deliberately sacrificed.4,11-13 This aids in precise identification of the anatomy of the C1 lateral mass for screw placement. Also, the C1-C2 joint cavity can be better visualized for packing the bone grafts and spacer insertion, thereby increasing the likelihood of solid bony fusion.8,16 In addition, the operative time and blood loss are reduced secondary to avoidance of dissection of the periganglionic venous plexus.5,10

However, the purported benefits of C2 nerve sectioning are not without any drawbacks.5 Sacrifice of the C2 nerve can cause postoperative numbness over the occiput and retroauricular area. Some patients can develop dysthesia along the C2 distribution.

Table 1: Postoperative Symptoms Related to C2 Nerve Dysfunction

<table>
<thead>
<tr>
<th>Parameter</th>
<th>C2 Sectioned* (n = 178)</th>
<th>C2 Preserved† (n = 12)</th>
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<tbody>
<tr>
<td>Paresthesia</td>
<td>39 (21.9)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Point scale [mean]</td>
<td>0.72</td>
<td>NA</td>
</tr>
<tr>
<td>Hypoesthesia</td>
<td>54 (30.3)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Point scale [mean]</td>
<td>0.84</td>
<td>NA</td>
</tr>
<tr>
<td>Occipital neuralgia</td>
<td>NA</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Point scale [mean]</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Dysesthesia</td>
<td>34 (19.1)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Point scale [mean]</td>
<td>0.56</td>
<td>NA</td>
</tr>
<tr>
<td>Occipital ulcer formation</td>
<td>9 (5.1)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Medication required for symptom relief</td>
<td>No</td>
<td>NA</td>
</tr>
<tr>
<td>Disturbs daily activities</td>
<td>No</td>
<td>NA</td>
</tr>
<tr>
<td>Surgical intervention needed</td>
<td>5†</td>
<td>NA</td>
</tr>
</tbody>
</table>

Data presented as n (%).
NA, not applicable.
*Transient traction-related neurologic deficit occurred in 2 patients.
†Unilateral in 3 patients and bilateral in 9.
‡Split-thickness skin graft/flare for occipital ulcer.

in the patients with reducible AAD. Pseudofacets were seen in 19.5% of patients. An anomalous VA was more prevalent in those with irreducible AAD (24.5%). These complex bony and vascular anomalies prevented reaching the C1-C2 joints without C2 nerve root sacrifice.

All 190 patients underwent short segment posterior C1-C2 fusion. The average blood loss in the patients with C2 nerve sacrifice was 454.2 mL and in the C2 nerve preservation group, it was 576.4 mL. The operative duration was 117.9 and 142.5 minutes in the C2 nerve sacrifice and preservation groups, respectively. The follow-up duration ranged from 3 to 48 months (mean, 23.4). The overall fusion rate was 91.8% at the 6-month follow-up examination.
However, whether these clinical symptoms affect patients’ functional outcomes and their quality of life is uncertain. Although a few studies have reported on this subject, we describe our own cohort of patients undergoing posterior C1-C2 fusion for congenital AAD and the various clinical consequences of sectioning the C2 nerve root.4,5,8-11

In contrast, an attempt to preserve the C2 ganglion requires its mobilization to visualize the C1 lateral mass. Its preservation can be associated with occipital neuralgia, which has been reported in approximately 35% of patients.4 Impingement on the C2 ganglion by the plate/screw construct is the likely cause of this neuralgia. Also, the limited exposure and inadequate drilling at the C1-C2 joint could impair the desired manipulation and bony fusion. Few investigators have studied the C2 nerve-related effects after C1-C2 fusion.4-11 The patient cohort in most of these series had AAD secondary to trauma, a degenerative process, or relatively simple congenital forms such as os odontoideum.4,8,10,11 In a review by Elliott et al., occupational numbness after C2 nerve root sectioning was reported in approximately 12% of patients. However, most were not very concerned about their symptoms. The functional outcomes and pain scores were not significantly different from those for the patients with C2 nerve root sacrifice.5,5,8,10,11 Also, the neuropathic pain that occurred after C2 nerve root preservation was more troublesome and affected patients’ quality of life.4,5 Our study also supports the findings that C2 nerve root sectioning might not be that distressing in the postoperative period. The magnitude of improvement in neurologic status that follows surgery possibly overshadows these relatively minor symptoms.

An important observation from our study is that a few patients developed serious and troublesome neuropathic occipital ulcers. These were challenging to treat and required a multidisciplinary team approach. This subset of patients, although small, should not be ignored. One might argue that immobilization with a cervical collar per se could have predisposed the patients to the development of occipital ulcers. This was unlikely because we have
not encountered such deep ulcers in our clinical practice in patients immobilized after spinal fixation for subaxial degenerative diseases. It is possible that the existing hypoesthesia in the C2 dermatome could have accentuated the formation of the ulcers. In the absence of intact sensation over C2 area, patients will not perceive the pain sensation as they would normally and tend to present at a late stage with frank skin changes and ulceration. Such a complication after C2 nerve sectioning has not been previously described. Hence, we recommend preservation of the C2 nerve root whenever it is feasible. The relevant factors are presented in the next section.

Determinants of C2 Nerve Root Preservation or Sectioning

Various factors direct the feasibility of C2 nerve preservation in patients with congenital AAD (Figures 4 and 5). The feasibility is dependent on the osseo-vascular anatomy around the C1-C2 joints and the morphology of the C2 ganglion.

1. Preservation of C2 nerve root (Figure 4)
   A. Osseous anatomy
      (I) Height of the C1 lateral mass beneath the C1 pedicle
      For C1 screw insertion, the available mean vertical height between the inferior margin of the posterior arch of the atlas to that of the inferior facet is approximately 4 mm. Therefore, patients with a robust (sufficient vertical height) C1 lateral mass beneath the C1 pedicle/arch and no associated anomalous VA are potential candidates for C2 nerve root preservation. This is irrespective of whether the C1 arch is normal or an assimilated one. In fact, the well-formed lateral mass chunk in an assimilated C1 provides a stronger bony mass for C1 screw purchase. Furthermore, the entry point for the screw can be brought slightly lower (close to the inferior margin), with the screw directed more cranially and thereby providing adequate space between the undersurface of the C1 posterior arch/pedicel and the tulip of the screw. This possibly lessens the chance of nerve root irritation by the screw head. In patients with an intact C1, such an inferior entry point, along with cranial angulation of the screw trajectory, could violate the normal occipito-atlantal joint.

2. Sectioning of the C2 nerve root (Figure 5)

B. Morphology and size of the C2 ganglion

C2 nerve root preservation can be attempted in those in whom the thickness of C2 ganglion is normal (approximately 5-7 mm). A large-size ganglion will limit access to the C1 lateral mass and the available space for C1 screw insertion. Recently, certain measurements such as a height for screw index have been developed to predict C2 nerve dysfunction in patients undergoing lateral mass fixation and is based on the difference in height between the C2 ganglion and its foramen.14

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Figure 3. (A, B) Intraoperative photographs (C1-C2 posterior fusion) showing preserved left C2 nerve root (asterisk in A) (Goel’s plate and screw technique) and bilateral C2 nerve roots (arrow in B) (polyaxial screw and rod fixation).
A. Osseous anatomy

(I) Height of the lateral mass beneath the C1 pedicle

At times, patients with an occipitalized C1 arch will have associated hypoplasia of the C1–condyle complex. This reduces the available vertical height of the lateral mass for C1 screw placement. In such cases, we drill a small portion of the inferior C1 lamina to create space to accommodate the Goel plate/C1 screw tulip. This step would be difficult without sectioning the C2 ganglion.

(II) Hypoplasia of the C1–occipital condyle complex

(III) Highly oblique C1–C2 joints

(IV) Extreme dislocation/spondyloptosis

(V) Incurved occiput

(VI) Pseudofacets

In these anomalies (numbers II–VI), the C2 isthmus lies close to the C1 arch or base of the occiput with restricted space to reach the C1-C2 joint. Some patients will have pseudofacets that will require extensive drilling to gain access to the actual C1-C2 joint. In such cases, we suggest cutting of the nerve root to access the C1-C2 joint and improve joint visualization.

B. Morphology and size of the C2 ganglion

Occasionally the C2 nerve root will be larger and hinder access to the C1 lateral mass and C1-C2 joint. In such cases, the C2 nerve root will need to be cut.

C. Vascular anomaly

An anomalous VA that lies in proximity to the C2 ganglion and the C1–C2 joint is not uncommon in patients with congenital AAD. It can be safeguarded only if it is delineated and mobilized off the C2 nerve root. This step requires C2 nerve sectioning because the VA is located ventral to the nerve root. In our study, approximately three fourths of the patients had complex C1-C2 joint anatomy. The anomalous VA added to the surgical difficulty. In these patients, C2 nerve root preservation was almost impossible. However, based on our unpleasant experience of occipital neuropathic ulcers in some patients, we would strongly suggest C2 nerve root preservation in favorable circumstances. To summarize, its preservation in congenital AAD is possible in patients with a robust C1 lateral mass and normal-size ganglion in the absence of other bony and vascular anomalies. Its preservation is difficult in the presence of C1 inferior facet–condylar hypoplasia, pseudofacets, highly deformed joints, an anomalous VA, and a bulky C2 ganglion.

3. Overcoming Surgical Difficulties and Alternative Solutions

As described, whenever the height of C1 lateral mass is reduced, drilling off a small portion of the inferior C1 lamina would accommodate the Goel plate/C1 screw tulip. Alternatively, the C1 pedicle screw can be inserted such that the shaft of the screw does not irritate the nerve root. However, the construct should be combined with C2 pars or translaminar screws. The disadvantage with this procedure is that it takes the construct away from the force of action and can cause C1-C2 redislocation. Similarly, one might fuse the occipital squama to the cervical spine, which can result in delayed complications unless multiple segments are included. Such multisegmental fusion adversely affects the quality of life because of severe restriction of neck movement.

When the C2 nerve root is bulky and is preserved, the Goel plate, rather than the rod construct, will be a good choice. The chance of nerve root irritation by the screw shaft is less likely with the former because the screw head lies flush with the plate. In a screw–rod construct, the screw head tends to slightly protrude above the posterior aspect of the C1 lateral mass and is more likely to cause nerve root irritation. A partially threaded screw is likely to lessen the nerve irritation. At times, the preserved nerve root can become slightly stretched over the surface of the Goel plate, which is 2 mm thick. In 3 of our patients, the nerve root had to be cut for similar reasons after an initial attempt to preserve it.
With no pain perceived over the C2 dermatomal distribution, the Philadelphia collar can accentuate the development of occipital ulcers. Therefore, one must be vigilant about the possibility of such a complication in AAD patients undergoing C2 nerve sectioning. Frequent clinical examinations are recommended during the follow-up of these patients. An alternative orthosis would be to use a sternal occipital mandibular immobilizer brace or halo whenever skin changes or superficial ulcers are detected at the initial stages to halt their progression to a full blown ulcer.

Although the present study had only a few subjects in whom the C2 nerve root preservation was attempted, we noted that 2 such patients developed transient upper limb monoparesis in the postoperative period. This could be explained by the traction on the cervical cord during retraction of the C2 ganglion. None of C2 preservation group complained of occipital neuralgia. However, a definite conclusion cannot be drawn from the small number of patients.

Study Limitations
The present study was a retrospective series and did not have a sufficiently large comparison group in whom C2 nerve root preservation was performed.

CONCLUSIONS
The pros and cons of sectioning the C2 nerve root should be remembered while performing posterior fusion for congenital AAD. In these patients, the C1-C2 anatomy might not be always favorable for preservation of the C2 nerve root. Few patients develop neuropathic occipital ulcers which adds to their morbidity. Hence, we would suggest an effort to preserve the C2 nerve root whenever feasible. This can be attempted in patients who have relatively less oblique joints with sufficient vertical height of the C1 lateral mass, normal-size C2 ganglion, and no anomalous VA.
REFERENCES


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