

Clinical Study

Postoperative occipital neuralgia with and without C2 nerve root transection during atlantoaxial screw fixation: a post-hoc comparative outcome study of prospectively collected data

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Abstract

BACKGROUND CONTEXT: Although routine transection of the C2 nerve root during atlantoaxial segmental screw fixation has been recommended by some surgeons, it remains controversial and to our knowledge no comparative studies have been performed to determine whether transection or preservation of the C2 nerve root affects patient-derived sensory outcomes.

PURPOSE: The purpose of this study is to specifically analyze patient-derived sensory outcomes over time in patients with intentional C2 nerve root transection during atlantoaxial segmental screw fixation compared with those without transection.

STUDY DESIGN: This is a post-hoc comparative analysis of prospectively collected patient-derived outcome data.

PATIENT SAMPLE: The sample consists of 24 consecutive patients who underwent intentional bilateral transection of the C2 nerve root during posterior atlantoaxial segmental screw fixation (transection group) and subsequent 41 consecutive patients without transection (preservation group).

OUTCOME MEASURES: A visual analog scale (VAS) score was used for occipital neuralgia as the primary outcome measure and VAS score for neck pain, neck disability index score and

FDA device/drug status: Not approved (C1 lateral mass screw; C1 posterior arch screw; C2 pedicle screw; and C2 laminar screw).

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Japanese Orthopedic Association score for cervical myelopathy and recovery rate, with bone union rate as the secondary outcome measure.

METHODS: Patient-derived outcomes including change in VAS score for occipital neuralgia over time were statistically compared between the two groups. This study was not supported by any financial sources and there is no topic-specific conflict of interest related to the authors of this study.

RESULTS: Seven (29%) of the 24 patients in the transection group experienced increased neuralgic pain at 1 month after surgery either because of newly developed occipital neuralgia or aggravation of preexisting occipital neuralgia. Four of the seven patients required almost daily medication even at the final follow-up (44 and 80 months). On the other hand, only four (10%) of 41 patients in the preservation group had increased neuralgic pain at 1 month after surgery, and at ≥ 1 year, no patients had increased neuralgic pain. The difference in the prevalence of increased neuralgic pain between the two groups was statistically significant at all time points (3, 6, 12, and 24 months postoperatively) except at 1 month postoperatively. The intensity of neuralgic pain, which preoperatively had not been significantly different between the two groups, was significantly higher in the transection group at the final follow-up.

CONCLUSIONS: C2 nerve root transection is not a benign procedure and, in our experience, more than a quarter of the patients experience increased neuralgic pain following C2 nerve root transection. Because the prevalence and intensity of postoperative neuralgia was significantly higher with C2 nerve root transection than with its preservation, we recommend against routine C2 nerve root transection when performing atlantoaxial segmental screw fixation. © 2013 Elsevier Inc. All rights reserved.

Keywords: C2 nerve root transection; C2 nerve root preservation; Atlanto-axial screw fixation; Occipital neuralgia, outcomes

Introduction

In few spinal operations do surgeons intentionally cut a nerve root, let alone a normal one. However, for posterior atlantoaxial segmental screw fixation, routine transection of the C2 nerve has been advocated because it can make the operation easier. More specifically, transection of the nerve improves exposure of the screw insertion sites and the C1-C2 facet joints, increases the fusion bed area, and allows easier control of bleeding from the venous plexus at C1-C2 [1–3]. Furthermore, proponents of routine transection state that because the C2 nerve has a mainly sensory function of supplying the greater occipital nerve, transection of the nerve should only cause mild sensory deficit over the corresponding dermatome. Goel et al., the originators of this technique, reported that sharp transection of the C2 ganglion did not lead to significant or annoying symptoms in their series [1], and several surgeons have reported similar results [2–6]. On the other hand, McCormick and Kaiser stated that in their experience, patients who required sectioning of the C2 nerve root reported significant discomfort related to pain and numbness involving the posterior scalp [7]. Unfortunately, these diametrically opposed viewpoints on the results of C2 nerve root transection [1–7] are based on retrospective analyses or anecdotal experience, providing level III evidence at best.

To complicate matters further, several authors have reported that some patients in whom the C2 nerve root is preserved during atlantoaxial segmental screw fixation complain of postoperative dysesthesia or neuralgia [8–12]. Therefore, it is not clear whether preservation or transection of the C2 nerve root results in better outcomes—particularly

those related to occipital neuralgia. We were unable to find, in a MEDLINE search, a prospective comparative study focusing on patient-derived outcomes of C2 nerve root transection versus preservation. Furthermore, we could not find a study evaluating temporal changes in patient-derived outcomes following root transection. The analysis of the change in patient's symptoms over time can lead to better understanding of the genuine impact of this procedure on everyday life. Given the limitations of previous studies regarding the true impact of C2 nerve root transection, the purpose of this controlled study was to compare patient-derived sensory outcomes over time in patients with and without intentional C2 nerve root transection during atlantoaxial segmental screw fixation.

Materials and methods

Inclusion and exclusion criteria

This study was approved by our institutional review board. This was a post-hoc analysis of prospectively collected data regarding postoperative outcomes following atlantoaxial fixation and arthrodesis done by the first author. Between July 2004 and December 2007, 24 patients underwent intentional bilateral transection of the C2 nerve root for better exposure and bone grafting during posterior atlantoaxial segmental screw fixation using C1 lateral mass or posterior arch screws and C2 pedicle or laminar screws. The nerve root was transected whenever such an operation was performed, regardless of the patient age, gender, and cervical spine pathology. Our intention had been to report

EVIDENCE & METHODS

Context

Transection of the C2 nerve roots is a technique used during C1–C2 arthrodesis with screw fixation.

Contribution

In this retrospective study, the authors found that such transection was associated with a significantly greater occurrence of C2 neuralgia (29% vs 10%).

Implication

Transection may cause neuralgia after C1–C2 instrumented arthrodesis. Alternative techniques or protection of the nerve may have less morbidity.

—The Editors

on the outcomes of these transections in a prospective manner. However, in December 2007, the first author altered the technique to preserve the root. Therefore, the design of the study changed from a single-arm prospective observational outcome study to become a comparative analysis of root transected versus preserved patients. Therefore, this was a post-hoc analysis of the prospectively collected data between 2004 and 2007 on the patients who had undergone C2 nerve root transection and prospectively collected data between 2007 and 2010 on the patients in whom the C2 nerve root was preserved. Between December 2007 and November 2010, the same surgeon performed posterior atlantoaxial segmental screw fixation without C2 nerve root transection except in six patients in whom unilateral transection was performed. Among those patients, all consecutive patients who underwent either bilateral transection (transection group) or no transection on either side (preservation group) and had fixation and fusion confined only to C1–C2 levels were enrolled in this study. We excluded patients with unilateral transection of the C2 nerve root, or

fixation extended above or below C1–C2 to limit the number of confounding variables and to allow clearer determination of the effect of C2 nerve root transection.

Surgical technique of C2 nerve root transection

In the transection group, the C2 nerve root was transected on both sides during the surgical approach. After a midline approach and exposure of C1–C2 areas in a prone position, the C2 nerve root was exposed by removing the overlying atlanto-episthophic ligament, which is a fibrous sheath housing the C2 nerve root between the caudal border of the C1 posterior arch and the cranial part of the C2 pars interarticularis [2,13–15].

Three methods were used for the transection. In the initial four cases, the C2 nerve root was sharply transected using a knife or Metzenbaum scissors according to the description of Goel et al. [1]. Hemostasis was subsequently achieved using a bipolar electrocautery and/or thrombin-soaked gelatin sponge (Spongostan, Ethicon, Somerville, NJ) and cottonoid pledgets. Because we found that this technique led to profuse bleeding, we altered our technique in the subsequent 10 cases in which we transected the root using low-powered monopolar electrocautery with a sharp needle tip. In the last 10 cases, the nerve root was first cauterized using bipolar electrocautery several times and subsequently transected using a knife or Metzenbaum scissors. This resulted in the least amount of blood loss. The nerve root was not ligated in any case.

The nerve root was transected at an area just proximal to the dorsal root ganglion in the belief that transection proximal to the dorsal root ganglion might result in less postoperative pain. To reduce bleeding, however, the dorsal root ganglion was not completely dissected before transection in all cases, so in some cases the nerve root was transected through the dorsal root ganglion rather than proximal to it. In addition, if the dorsal root ganglion was located too medially [13], the nerve root was transected through the ganglion to prevent the leakage of cerebrospinal fluid.

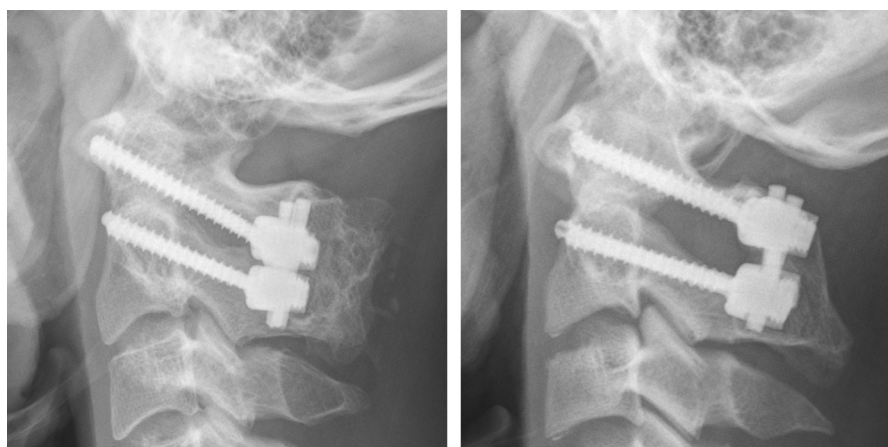


Fig. 1. Lateral radiographs showing C1 lateral mass screws (Left) and C1 posterior arch screws (Right).

Surgical technique of screw placement and bone grafting

C1 lateral mass or posterior arch screws and C2 pedicle or laminar screws were placed in the usual manner described previously [16–19]. To simplify insertion of C1 screws, in the transection group, the C1 lateral mass screws were inserted under the C1 posterior arch (Fig. 1, Left) in all but one case, in which the screws were inserted into the posterior arch. This method was typically used because the C1 lateral mass was widely exposed as the root had been transected. In the preservation group, on the other hand, only posterior arch screws, which were inserted through the posterior arch instead of under the arch (Fig. 1, Right), were used to minimize C2 root irritation by these screws. For C2 fixation, pedicle screws were preferred in both groups. Laminar screws were used on occasion when the pedicle was too small to accommodate a screw on the preoperative CT scans. Atlantoaxial arthrodesis was done by extra-articular (only in the transection group), intra-articular, and/or posterior fusion.

Data collection

Demographic data and clinical data including diagnosis, operative techniques of nerve root transection and bone grafting, and the type of screws used were prospectively recorded in a database. In addition, a questionnaire form, consisting of a 10-point visual analog scale (VAS) score for both neck pain and occipital neuralgia, with 0 indicating no pain and 10 indicating the worst pain; pain drawing for the distribution and character of the neck and neuralgic pain; and drawing for the distribution of totally anesthetic area, was completed by the patients preoperatively and at each follow-up, along with the neck disability index (NDI) score. The questionnaire forms were filled out by the patients themselves before meeting the surgeon at follow-up visits and submitted to a research coordinator who tabulated and scanned the forms. For the patients with preoperative myelopathy, the 17-point Japanese Orthopedic Association score for cervical myelopathy (JOA score) was evaluated preoperatively and at each visit, and the recovery rate of the JOA score was calculated using Hirabayashi's method [20]. Follow-up visits were typically done at 1, 3, 6, and 12 months after surgery and at every 1 or 2 years thereafter. Radiographs including flexion-extension lateral views were taken at every follow-up at 3 months and later, and three-dimensional computed tomography (CT) scans were taken between 6 and 12 months after surgery in all patients. Bone union was defined as having obvious bone union with fusion mass in sagittal reconstruction CT scan images and/or lateral radiographs and no segmental motion on flexion-extension lateral radiographs.

Data analysis

An independent experienced spine surgeon who was not involved in the care of the patients analyzed all data and

radiographs. Special attention was paid to a detailed assessment of patient-derived outcomes related to occipital neuralgia, including the change in VAS score over time and the pain distribution. In addition, change over time in the proportion of patients who had increased neuralgic pain postoperatively versus preoperatively, either because of newly developed occipital neuralgia or aggravation of pre-existing occipital neuralgia, was statistically compared between the two groups. The frequency of increased neuralgic pain was also compared among the three different methods of C2 nerve root transection.

Statistical analysis

Continuous variables were compared using Student *t* test and categorical variables were compared using Fisher exact test. SPSS software package version 19.0 (SPSS, Chicago, IL) was used to perform the statistical analysis and the level of significance was set at a two-tailed $p < .05$.

Results

Patient characteristics, surgical procedures, and functional outcomes

Demographic and clinical data of the two groups are summarized and compared in Table 1. Twenty-four consecutive patients in the transection group and 41 consecutive patients in the preservation group were enrolled in this study. There was no significant difference in age ($p = .182$), gender ($p = .197$), nature of C1–C2 pathology ($p = .680$), and prevalence of preoperative myelopathy ($p = .443$) and/or occipital neuralgia ($p = .159$) between the two groups. The mean follow-up was 59 months (range,

Table 1
Demographic and clinical data

| Variables | Group | | p Value |
|---|-----------------------|------------------------|-------------------|
| | Transection (N=24) | Preservation (N=41) | |
| Age (y)* | 48±20 | 41±20 | .182 [†] |
| Gender, n (%) | | | |
| Man | 14 (58) | 16 (39) | .197 [‡] |
| Woman | 10 (42) | 25 (61) | |
| Nature of C1–C2 pathology, n (%) | | | |
| Congenital | 10 (42) | 19 (46) | .680 [‡] |
| Traumatic | 6 (25) | 13 (32) | |
| Rheumatologic | 4 (17) | 3 (7) | |
| Other | 4 (17) | 6 (15) | |
| Preoperative myelopathy, n (%) | | | |
| Yes | 12 (50) | 16 (39) | .443 [‡] |
| No | 12 (50) | 25 (61) | |
| Preoperative occipital neuralgia, n (%) | | | |
| Yes | 4 (17) | 14 (34) | .159 [‡] |
| No | 20 (83) | 27 (66) | |
| Follow-up (mo)* | 59±16 | 23±9 | .000 |

* Mean±standard deviation.

[†] Student *t* test.

[‡] Fisher's exact test is used.

Table 2
Surgical procedures

| Procedures | Group | | p Value* |
|------------------------------|----------------------|-----------------------|----------|
| | Transection n (%) | Preservation n (%) | |
| C1 screw | | | |
| Lateral mass screw | 46 (96) | 0 (0) | <.001 |
| Posterior arch screw | 2 (4) | 82 (100) | |
| C2 screw | | | |
| Pedicle screw | 39 (81) | 59 (72) | .164 |
| Laminar screw | 9 (19) | 23 (28) | |
| Extra-articular facet fusion | | | |
| Yes | 23 (96) | 0 (0) | <.001 |
| No | 1 (4) | 41 (100) | |
| Intra-articular facet fusion | | | |
| Yes | 15 (62) | 33 (80) | .147 |
| No | 9 (38) | 8 (20) | |
| Posterior fusion | | | |
| Yes | 22 (92) | 41 (100) | .133 |
| No | 2 (8) | 0 (0) | |

* Fisher's exact test.

25 to 80 months) in the transection group and 23 months (range, 12 to 43 months) in the preservation group; the difference was statistically significant ($p<.001$).

Surgical procedures performed in the two groups are summarized in Table 2. In the transection group, lateral mass screws were placed at C1 in all patients except 1, whereas in the preservation group posterior arch screws were used in all patients ($p<.001$). At C2, either pedicle or laminar screws were used in both groups and the difference was not statistically significant ($p=.164$). Whereas all but one patient underwent extra-articular fusion of the C1-C2 facet joints in the transection group, none did in the preservation group ($p<.001$). There was no statistically significant difference in the proportion of patients who underwent intra-articular fusion of the C1-C2 facet joints

Table 3
Functional outcome scores

| Outcome scores | Group | | p Value* |
|----------------------------|------------------------|-------------------------|----------|
| | Transection Mean±SD | Preservation Mean±SD | |
| JOA score [†] | | | |
| Preoperative | 11.5±2.7 | 12.0±3.7 | .677 |
| At final follow-up | 14.3±2.3 | 14.4±3.3 | .948 |
| Recovery rate (%) | 54.7±33.3 | 58.9±33.2 | .739 |
| NDI score (%) [‡] | | | |
| Preoperative | 38.8±18.3 | 35.2±24.0 | .531 |
| At final follow-up | 23.0±16.2 | 22.0±17.4 | .818 |
| VAS score for neck pain | | | |
| Preoperative | 4.9±2.7 | 3.9±2.6 | .138 |
| At final follow-up | 2.2±1.6 | 1.8±2.4 | .463 |

JOA, Japanese Orthopedic Association; NDI, neck disability index; SD, standard deviation; VAS, visual analog scale.

* Student *t* test is used.

[†] JOA score of only those patients with preoperative myelopathy including 12 patients in the transection group and 16 patients in the preservation group.

[‡] NDI score converted to a percentage scale.

($p=.147$) or posterior fusion ($p=.133$) between the two groups. All patients had solid union on plain radiographs and CT scans between 6 and 12 months after surgery in both groups. Leakage of cerebrospinal fluid at the site of root transection was not seen either during or after surgery in any patients.

Functional outcome score assessment for the two groups is summarized in Table 3. The preoperative and postoperative NDI score, VAS score for neck pain, and JOA score and recovery rate were not significantly different between the two groups.

Occipital neuralgia in the transection group

In the transection group, four patients had unilateral occipital neuralgia preoperatively, whereas the other 20 did not. Among the 20 patients without preoperative neuralgia, five (25%) complained of new bilateral neuralgia postoperatively. Fig. 2, Left shows the change in the intensity of neuralgic pain over time in these five patients. Although one of them had complete resolution at 24 months after surgery, the other four had persistent pain until the final follow-up at 70 to 80 months. Two of the four did not require medication for the neuralgia at final follow-up (70 and 75 months, respectively) when the VAS score decreased to 3 in both patients, although both used occasional medication including gabapentin (Pfizer, New York, NY) during the follow-up periods. The other two required almost daily medication including pregabalin (Pfizer) even at final follow-up (71 and 80 months, respectively) when the VAS score was 6 and 5, respectively. In both patients, epidural and C2 ganglion blocks were tried but they were not effective.

Among the four patients who had preoperative unilateral occipital neuralgia (with VAS of 3, 4, 5, and 8, respectively), two patients (with preoperative VAS of 3 and 8, respectively) had improvement after surgery (Fig. 2, Right) and required only intermittent nonsteroidal anti-inflammatory medications during follow-up. At the last follow-up at 70 and 71 months, respectively, neither patient required medication with VAS of 1 and 3, respectively. The other two patients, who had unilateral neuralgia preoperatively (VAS of 4 and 5, respectively), developed bilateral neuralgia postoperatively. The intensity of the pain was exacerbated in both patients postoperatively (with VAS of 9 and 10, respectively) at 1 month after surgery, followed by slight improvement over time with VAS at final follow-up of 6 at 44 months and 7 at 49 months, respectively. Both patients required almost daily medication including pregabalin even at final follow-up. In one of these two patients, an epidural block was tried, but was not effective.

Occipital neuralgia in the preservation group

In the preservation group, 14 patients had unilateral (7) or bilateral (7) occipital neuralgia preoperatively, whereas

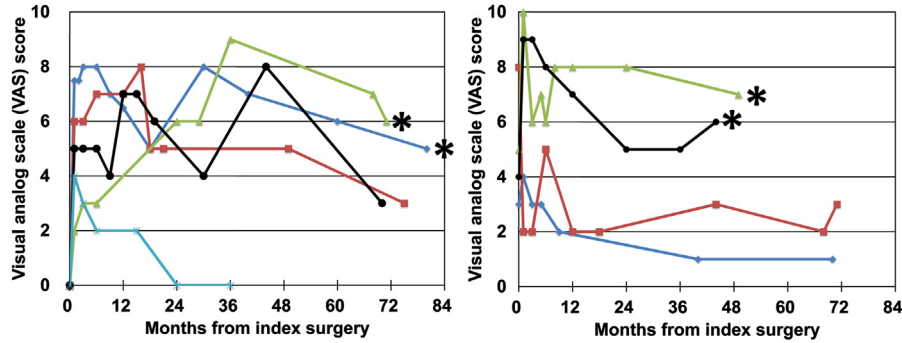


Fig. 2. The change in the intensity of neuralgic pain over time in all patients with preoperative and/or postoperative occipital neuralgia in the transection group is shown. (Left) The visual analog scale (VAS) scores of five patients who did not have preoperative occipital neuralgia but developed new neuralgia after surgery. Two patients (asterisks) required almost daily medication until the final follow-up. (Right) The VAS scores of four patients who had preoperative occipital neuralgia are shown. Two patients (asterisks) required almost daily medication until the final follow-up.

the other 27 did not. Among the 27 patients without preoperative neuralgia, four (15%) developed new neuralgia postoperatively, including two patients with unilateral and two patients with bilateral symptoms (Fig. 3, Left), with a VAS score of 2, 2, 3, and 5, respectively, at 1 month postoperatively. All four patients had complete resolution of neuralgic pain within 1 year after surgery.

Among the 14 patients who had preoperative occipital neuralgia with a mean VAS score 5.4 (range, 3 to 8), nine had complete resolution within 6 months of surgery (Fig. 3, Right). The other five had alleviation of pain compared with their preoperative status. At final follow-up ranging from 12 to 37 months postoperatively, the VAS score ranged from 1 to 4, and only one patient (with VAS of 4 at 15 months after surgery) was on medication, taking nonsteroidal anti-inflammatory drugs intermittently.

Comparison of occipital neuralgia between the transection and preservation groups

As described, in the transection group, seven (29%) of the 24 patients experienced increased neuralgic pain at 1 month after surgery, either with new or aggravated occipital neuralgia. Even at final follow-up ranging from 44 to

80 months, six had increased neuralgic pain and four required almost daily medication. On the other hand, in the preservation group, only four (10%) of 41 patients had increased neuralgic pain at 1 month after surgery, and no patients had increased neuralgic pain at or after 1 year after surgery. Table 4 summarizes the percentages of patients with increased neuralgic pain in the two groups. The rate of increased neuralgic pain was significantly higher in the transection group than in the preservation group at 3, 6, 12, and 24 months postoperatively ($p=.031, .031, .000, \text{ and } .022$, respectively), although it was not significantly different at 1 month postoperatively ($p=.083$). In addition, as shown in Table 5, the average VAS score for occipital neuralgia, which was not significantly different between the two groups preoperatively ($p=.125$), was significantly higher in the transection group than in the preservation group at final follow-up ($p=.003$).

Comparison of occipital neuralgia rates based on C2 nerve root transection method

Among the four patients in whom the C2 nerve root was transected using a knife or Metzenbaum scissors followed by hemostasis, one had increased neuralgic pain. Among

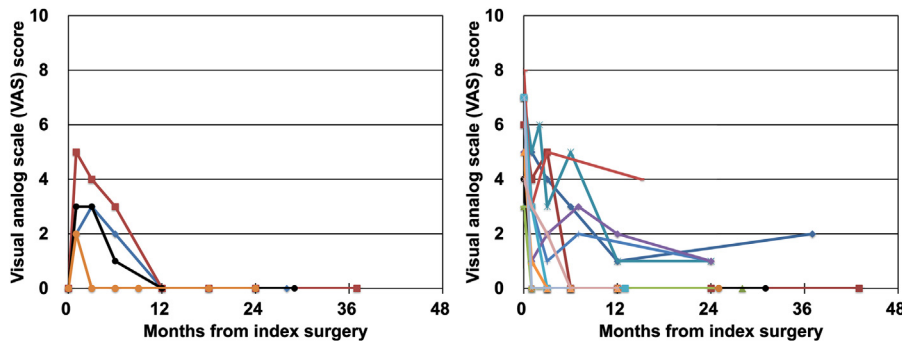


Fig. 3. The change in the intensity of neuralgic pain over time in all patients with preoperative and/or postoperative occipital neuralgia in the preservation group is shown. (Left) The visual analog scale (VAS) score of four patients who did not have preoperative occipital neuralgia but developed new neuralgia after surgery is shown. (Right) The VAS score of 14 patients who had preoperative occipital neuralgia.

Table 4

Patients with increased neuralgic pain postoperatively, either because of newly developed occipital neuralgia or aggravation of preexisting occipital neuralgia

| Months | Group | | p Value* |
|--------|---------------------|----------------------|----------|
| | Transection n/N (%) | Preservation n/N (%) | |
| 1 | 7/24 (29) | 4/41 (10) | .083 |
| 3 | 7/24 (29) | 3/41 (7) | .031 |
| 6 | 7/24 (29) | 3/41 (7) | .031 |
| 12 | 7/24 (29) | 0/41 (0) | <.001 |
| 24 | 6/24 (25) | 0/23 (0) | .022 |

* Fisher's exact test.

the 10 patients in whom the C2 nerve root was transected using a monopolar electrocautery with a sharp needle tip, three had increased neuralgic pain. Among the 10 patients in whom the nerve root was cauterized using bipolar electrocautery before transection, three had increased neuralgic pain postoperatively. There was no significant difference in the frequency of increased neuralgic pain among the three transection methods ($p=1.000$).

Location of occipital neuralgia and anesthesia

The location of occipital neuralgia was not confined to the occipital area. Typical paresthetic neuralgic pain characterized as stabbing or burning was located in the vertical, retroauricular, and/or retromandibular as well as occipital zones, as illustrated in Fig. 4, Top and Bottom. Not all patients with occipital neuralgia had neuralgic pain in the occipital zone. For example, among the nine patients who had occipital neuralgia at 1 month after surgery in the transection group, only four patients had pain in the occipital zone (Fig. 4, Bottom). In the transection group, 21 patients had completely anesthetic areas in the occipital zone postoperatively, whereas the other three did not. In the preservation group, none had completely anesthetic areas postoperatively.

Discussion

Although routine transection of the C2 nerve root during atlantoaxial segmental screw fixation has advantages including improved visualization of the C1 screw insertion site, better preparation and decortication of the bone graft

Table 5

VAS score for patients with occipital neuralgia

| VAS score for occipital neuralgia | Group | | p Value* |
|-----------------------------------|---------------------|----------------------|----------|
| | Transection Mean±SD | Preservation Mean±SD | |
| Preoperative | 0.8±2.1 | 1.9±2.8 | .125 |
| At final follow-up | 1.4±2.3 | 0.2±0.7 | .003 |

SD, standard deviation, VAS, visual analog scale.

* Student *t* test is used.

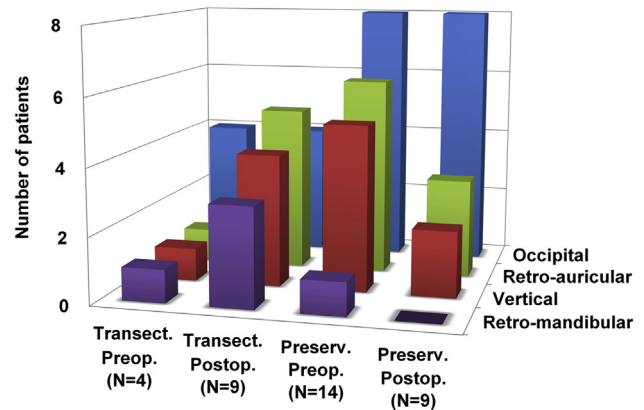
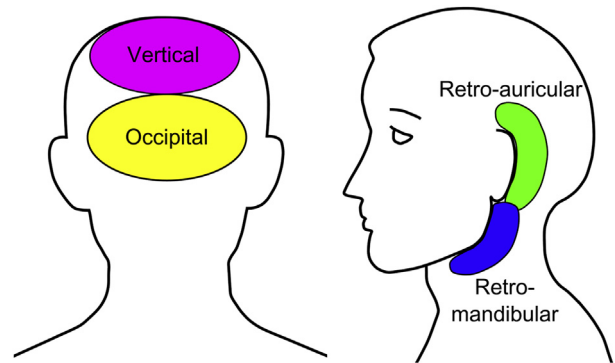


Fig. 4. The location of occipital neuralgia is illustrated. (Top) The painful areas can be divided into four typical zones. (Bottom) A bar graph shows the location of the preoperative and postoperative occipital neuralgia in the two groups.

recipient site, and decreased bleeding, outcomes of C2 nerve root transection, particularly related to sensory sequelae are controversial [1–7,20]. Although there have been several contradictory reports on the advantages and disadvantages of C2 nerve root transection [1–7,20], we were unable to find, in a MEDLINE search, any prospective comparative studies specifically analyzing patient-derived sensory outcomes of C2 nerve root transection. Given the disparate reports on this topic, we elected to perform a prospective comparative study focusing on patient-derived sensory outcomes following C1–C2 fusion with or without intentional C2 nerve root transection.

Key findings of the current study

The results of the current study suggest that C2 nerve root transection is not a benign procedure for more than a quarter of the patients and that outcomes related to occipital neuralgia are better with C2 root preservation than with its sacrifice. More specifically, seven (29%) out of the 24 patients in the transection group experienced increased neuralgic pain at 1 month after surgery, compared with their preoperative status, either with newly developed or aggravated occipital neuralgia. Four of these patients required almost daily medications, even after achieving

solid fusions and continuing until their final follow-up (44 and 80 months). In contrast, in the preservation group, only four (10%) of 41 patients had increased neuralgic pain at 1 month after surgery compared with their preoperative status, and at ≥ 1 year, no patient had increased neuralgic pain. The difference in prevalence of increased neuralgic pain between the two groups was significant at all time points beyond the first month. In addition, the pain intensity was significantly higher in the transection group.

As expected, the preoperative and postoperative NDI score, VAS score for neck pain, and JOA score and recovery rate, none of which reflect occipital neuralgia, were not significantly different between the two groups. Additionally, bone union rate, which might be thought to be better in the preservation group, because C2 nerve root transection provides better bone graft recipient beds, was not significantly different between the two groups, either.

Comparison with relevant literature on the outcomes of C2 nerve root transection

Although one article recommended against routine transection of C2 nerve root [7], most of the previous studies on the pros and cons of C2 nerve root transection have been favorable toward the procedure, reporting only a small number of patients with postoperative neuropathic pain adversely affecting the quality of life [1–6]. These findings and opinions are in stark contrast to the results of the current study. There are several possible reasons for the contradicting results between the previous reports and the current study. First of all, the study designs of the previous reports were different, providing lower class evidence (class III) than the current study (class II). All of the previous studies or articles [1–7] were retrospective, whereas the current study is prospective. In addition, only three of those retrospective analyses [2–4] focused on the outcomes of C2 root transection. Furthermore, except for those three studies [2–4], it is unclear whether the data were patient-reported or surgeon-derived; in contrast, the current study analyzed patient-derived outcome data including a pain score. Only by asking the person most affected by the transection, namely the patient, can we understand the true impact of this procedure. Moreover, none of the previous studies have scrutinized the change in outcomes during the follow-up period as in the current study, which analyzed detailed changes with time. The shortcomings of all of the previous studies on this topic are well-known: recently, in a detailed systematic review of the studies on the outcomes of C2 nerve root transection, Elliot et al. stated that although neuropathic pain appeared to be nearly absent following C2 nerve root transection, the conclusion was based on class III evidence and the true incidence of postoperative C2 dysfunction was likely underestimated [21]. We agree with the assessment by Herzog and Groff [22], who commented on the article by Elliott et al., and stated that the inherent

weakness of the review article was its reliance on the poor quality of the evidence available at the time of its writing.

Second, in most of the previous reports, it is not clear which of the sensory deficits or symptoms is associated with the C2 nerve root transection [1–3,5–7]. It is described in only one report, in which the authors defined occipital neuralgia as the presence of pain in the suboccipital region only [4]. On the contrary, in the current study, occipital neuralgia was located not only in the occipital zone but also in the vertical, retroauricular, and/or retromandibular zones (Fig. 4, Top and Bottom). As an extreme example, among the nine patients who had neuralgic pain at 1 month after surgery in the transection group, only four had pain in the occipital zone; in the other five patients, neuralgic pain was located in nonoccipital zones (Fig. 4, Bottom). In previous reports, neuralgic pain in nonoccipital zones might have been ignored and not recognized as being related to the transection, thus underestimating the incidence of postoperative occipital neuralgia. Neuralgic pain in those zones might be thought to be unrelated to C2 neuralgia because they typically are in the trigeminal or adjacent C3 dermatomal areas. However, it is well known that the painful areas of occipital neuralgia frequently include those areas [23–25].

Interestingly, none of the previous favorable reports following C2 nerve root transection [1–6] provided reasonable explanations for why the transection was inconsequential, in comparison to transection of other nerve roots, which carries a risk of neuralgic pain. For example, postoperative neuralgic pain has been described following thoracic nerve root transection during total en bloc spondylectomy or after injuries to either peripheral or central nervous system such as limb amputation and traumatic spinal cord injury [26,27]. Based on this evidence, we might infer that it is natural that some patients who undergo transection of the C2 nerve root may have unfavorable results, as noted by McCormick and Kaiser [7].

Are the sensory outcomes of C2 nerve root preservation better than transection?

When attempting to answer whether C2 nerve root transection or preservation is associated with better clinical outcomes, one has to determine which method has a higher incidence, pain intensity, and duration of postoperative occipital neuralgia. Although several authors have reported that some patients complain of postoperative dysesthesia or neuralgia with C2 root preservation [8–12], most cases are transient. To the best of our knowledge, only one report has compared the results of C2 nerve root transection and preservation [3]. However, this study unfortunately had only five patients in the preservation group and is retrospective in design. Elliot et al., in their review article, concluded that C2 nerve root preservation and retraction for C1 screw placement may result in a higher incidence of neuropathic pain than transection [21]. However, it should be remembered

that this review article was based the available class III evidence [21,22], as described previously. In the current prospective comparative study, increased neuralgic pain (either with aggravated or newly developed neuralgia) was less common, less intense, and less persistent in the preservation group than in the transection group.

Limitations of this study

As with any study, there are several limitations with ours. First of all, the current study is not randomized. There may have been subtle biases in doing the preservation group second. However, we know of no way that one could adversely influence the outcomes of the transection group or improve the outcomes of the preservation group, assuming that a surgeon would actually try to. It is also possible that a few years of additional experience may have contributed to decreased C2 nerve irritation in the preservation group. However, the first group all had their roots transected. This is quite easy to do and takes little experience. A few years' additional experience is not likely to have helped in transecting the root. Therefore even if the two groups had been randomized and operated over the same period, it is likely that our findings would remain unchanged.

Second, the starting point for the C1 fixation varied between the two groups. Standard lateral mass screws were used for most patients (96%) in the transection group. In the preservation group, the starting point of the lateral mass screw was placed more cranially, at the level of the posterior arch of C1. We used these posterior arch screws in the preservation group to keep them more cranial to the C2 nerve root than standard lateral mass screws. We preferred standard C1 lateral mass screws in the transection group because, after root transection, they were easier to place than screws that started in the posterior arch. Because posterior arch screws require less retraction of the C2 nerve root during their placement and remain farther away from the C2 nerve root after surgery, there is less chance of root irritation compared with lateral mass screws. Therefore this study demonstrates that better postoperative outcomes related to occipital neuralgia can be achieved with C2 root preservation using C1 lateral mass screws starting at the posterior arch than with C2 root transection using of C1 standard lateral mass screws.

Finally, the method of root transection may be related to the occurrence of occipital neuralgia following C2 nerve root transection. First, the level of transection in relation to the dorsal root ganglion might influence the occurrence of the postoperative neuralgia. Second, transection methods might influence the occurrence of occipital neuralgia. Although the results of three different transection methods were not significantly different (Table 4), the numbers of patients in the three groups were small. Finally, ligation of the stump of the C2 nerve root might minimize the occurrence of postoperative neuralgia. As far as we know, no study has

analyzed the effect of those surgical techniques describe here on the incidence, pain intensity, and/or duration of postoperative occipital neuralgia. Prospective comparative studies with a sufficient number of cases are required.

Despite these limitations, we believe that this study has unique strengths. We specifically focused on postoperative occipital neuralgia. We analyzed prospectively collected and, in addition, patient-derived data rather than retrospectively collected surgeon-derived data. We performed a comparative analysis with relatively large numbers of patients in both groups. We assessed the change of the severity of pain over time, particularly with a relatively long follow-up for those in the transection group.

Conclusions

To our knowledge, this is the only prospective comparative study to specifically analyze the patient-derived sensory outcomes over time in patients with intentional C2 nerve root transection during atlantoaxial segmental screw fixation compared with those without C2 nerve root transection. Our results demonstrate that C2 nerve root transection is not a benign procedure and more than a quarter of the patients experience increased neuralgic pain following C2 nerve root transection. In addition, prevalence and pain intensity of postoperative neuralgia was significantly higher after C2 nerve root transection than its preservation. Based on the results of this study, we recommend against routine C2 nerve root transection when performing atlantoaxial segmental screw fixation. We understand that there are some situations where transection of the C2 nerve root may be helpful or necessary, including some pathologic or congenital conditions. However, for most degenerative conditions, surgeons should be aware that routine C2 nerve root transections may result in postoperatively increased neuralgia in up to 29% of patients.

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