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The efficacy of C1/2 arthrodesis with C2 root resection at symptomatic side for occipital neuralgia from atlantoaxial osteoarthritis

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This article aimed to assess the efficacy of surgical treatments, particularly arthrodesis with C2 nerve transection, in patients with atlantoaxial osteoarthritis (AAOA). Data of patients with AAOA who underwent surgical treatment between January 2020 and November 2023 were retrospectively collected. Of 11 patients with AAOA, nine underwent fusion surgery, and two underwent decompression surgery. C2 root transection on the symptomatic side was performed in all fusion cases. C2 root transection was not performed during decompression. All patients experienced immediate and persistent pain relief after surgery. Four patients who underwent fusion surgery reported a complete disappearance of pain. Four patients who underwent fusion surgery reported some numbness of the scalp, but it did not affect the quality of daily activities or cause significant discomfort. One patient who underwent decompression surgery reported residual tingling sensation and hyperesthesia of the scalp. We concluded that C1-2 fusion surgery is an excellent surgical treatment option and should be primarily considered for patients with refractory AAOA. C2 root transection on the symptomatic side can completely alleviate pain at the expense of minor scalp anesthesia. Decompressive-only surgery is not recommended but can be considered in strictly selected patients.

Keywords Atlantoaxial osteoarthritis, Occipital neuralgia, C2 root transection, Atlantoaxial fusion

Occipital neuralgia resulting from atlantoaxial osteoarthritis (AAOA) is a rare degenerative disease that can be devastating to a patient's quality of life if not diagnosed correctly. Since its first official report in 1984 by Ehni et al.[1], AAOA has been recognized as an important disease entity among the various causes of occipital neuralgia. The pain originates from compression of the C2 root by the degenerated atlantoaxial facet joint, which forms the main component of the greater occipital nerve. Patients typically present with sharp, shooting occipital pain that aggravates with neck rotation to the symptomatic side[2]. Physical examination usually reveals tenderness along the distribution of the greater occipital nerve, and temporal alleviation of symptoms is observed after anesthetic and steroid injections at the C1/2 facet joint[3]. Computerized tomography (CT) and open-mouth odontoid radiography are useful for detecting asymmetric AAOA, and magnetic resonance imaging (MRI) shows compression of the C2 root and nearby bone marrow edema.

As it is relatively rare, it can be misdiagnosed as a common headache or manifestation of subaxial spondylosis[4]. The majority of patients' symptoms resolve after conservative treatment, such as oral analgesics, physical therapy, or steroid injection[3]. However, some patients show refractory pain from severe degenerative arthrosis of the C1-2 facet, and surgical intervention might be the only curative treatment for them. Therefore, early diagnosis is important to reduce unnecessary periods of severe refractory pain.

Atlantoaxial fusion surgery has shown excellent clinical outcomes for refractory occipital neuralgic pain from AAOA[5–9]. Fusion surgery may be an effective treatment by eliminating the micromovement of the degenerated C1-2 facet and dynamic compression of the C2 root[3]. However, other additional pathogeneses of occipital neuralgia associated with C1/2 foraminal stenosis and direct mechanical compression by a bony spur or ossicle should be considered. Our surgical procedures consisted of C2 root transection on the symptomatic side as well as C1/2 arthrodesis. C2 root transection during instrumentation can provide favorable outcomes

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and a safe surgical corridor without definite complications[10–13]. It can be assumed that this technique may also be effective for AAOA patients with occipital neuralgia and can aid in complete recovery of pain. Here, we report our experience with 11 patients with AAOA presenting with occipital neuralgia who underwent surgical treatment and focus on the efficacy of the combination of C1-2 arthrodesis and C2 root transection.

Methods

This retrospective study was performed in a single institution in patients who underwent surgical treatment for occipital neuralgia due to atlantoaxial osteoarthritis (AAOA). This study was approved by Institutional Review Board of Gangnam Severance Hospital, Yonsei University Health System, which waived the requirement for informed consent. All methods in this research were performed in accordance with relevant guidelines/regulations.

Data of patients with AAOA who underwent surgical treatment between January 2020 and November 2023 were retrospectively collected. The inclusion criteria were: presence of typical occipital neuralgic pain, refractory pain persisting for at least 6 months after conservative treatment, and a radiologic diagnosis of atlantoaxial osteoarthritis. Typical occipital neuralgic pain was regarded as a sharp, shooting pain radiating from the neck to the occipital or retroauricular area and aggravated with axial neck rotation to the symptomatic side. Radiologic diagnoses included signs of asymmetric sclerotic and degenerative changes in the C1/2 facet joint on CT and X-ray imaging, bone marrow edema near the C1/2 facet joint on STIR or fat suppression MRI, or C2 root compression in the C1/2 foraminal space on MRI. The exclusion criteria were the presence of myelopathy and a definite history of infection, trauma, or malignancy.

Decompression or fusion surgeries were performed. Fusion surgery was performed using the Harms-Goel technique, which involved lateral mass screw fixation for the atlas and pedicle screw or lamina screw fixation for the axis. A pedicle or lamina screw for the axis was selected depending on the pathway of the vertebral artery. The C2 root on the symptomatic side was always transected during fusion surgery. Interfacet graft insertion was used in some patients to increase the disc space and facilitate the fusion process. Inter-laminar allograft bone chips were applied to all patients at the end of fusion surgery. The representative case of fusion surgery is illustrated in Fig. 1. For decompression surgery, we performed a partial or total C1 laminectomy with a C2 partial upper cut laminectomy and foraminotomy of the symptomatic side. The representative case of decompressive surgery is illustrated in Fig. 2.

Baseline demographic factors, such as patient age and sex, direction of symptoms, and duration of symptoms before the first clinical visit, were recorded. Cervical instability was assessed using dynamic radiography. Various surgical techniques, including instrumentation, bone grafting, and C2 root transection were recorded. Medical or surgery-related complications during the follow-up period were also recorded. The clinical outcomes were described based on the patients' self-reports at the last outpatient follow-up.

Results

Eleven patients with AAOA were retrospectively reviewed. Patient descriptions are listed in Table 1. Seven patients presented with left occipital neuralgic pain, while four presented with right-sided pain. All patients showed persistent and refractory symptoms despite conservative treatment for at least one year, with one patient showing symptoms lasting for 20 years. Three patients had combined cervical instability on dynamic radiography, all of whom underwent fusion surgery.

Nine patients underwent fusion surgery, and two patients underwent decompression surgery. C2 root transection on the symptomatic side was performed in all fusion cases. After C2 root transection, insertion of the interfacet graft was performed in five patients due to the severely collapsed disc space. C2 root transection was not performed during decompression. One patient had a complication of vertebral artery injury that occurred during C2 pedicle screw insertion.

All patients experienced immediate and persistent pain relief after surgery. None of the patients experienced mechanical instrument-related complications during the follow-up. Four patients who underwent fusion surgery reported a complete disappearance of pain. Four patients who underwent fusion surgery reported decreased sensation on the scalp, but it did not affect the quality of daily activities or cause significant discomfort. One patient who underwent decompression surgery reported residual tingling sensation and hyperesthesia of the scalp.

Case illustration

Case 6

A 66-year-old female patient presented with severe left occipital neuralgia that persisted for 5 years. Various conservative treatments including medication or injection failed to relieve the symptoms. Physical examination showed typical characteristic of occipital neuralgia, with pain radiating from neck to occipital and retro auricular area. The pain aggravated with head rotation to left, limiting the patient form neck motion in daily activities. The open mouth X-ray image and the coronal view of CT image showed apparent degenerative change of left C1/2 facet joint. The CT image also revealed a bony spur and calcified ossicles of left C1/2 facet joint that is causing the root compression. The MRI image also showed impingement of left C2 root between C1/2 foraminal space (Fig. 1).

Under the diagnosis of left occipital neuralgia due to left C1/2 osteoarthritis and C2 root compression, the patient underwent a C1-2 fusion surgery. The fusion procedure was performed with bilateral lateral mass screw fixation for the atlas, and right pedicle screw and left lamina screw fixation for the axis. Left C2 lamina screw was chosen due to the high riding left dominant vertebral artery. Severe C2 root compression by the bony spur and calcified ossicles between C1/2 facet joint was noted, and the left C2 root was transected proximal to the

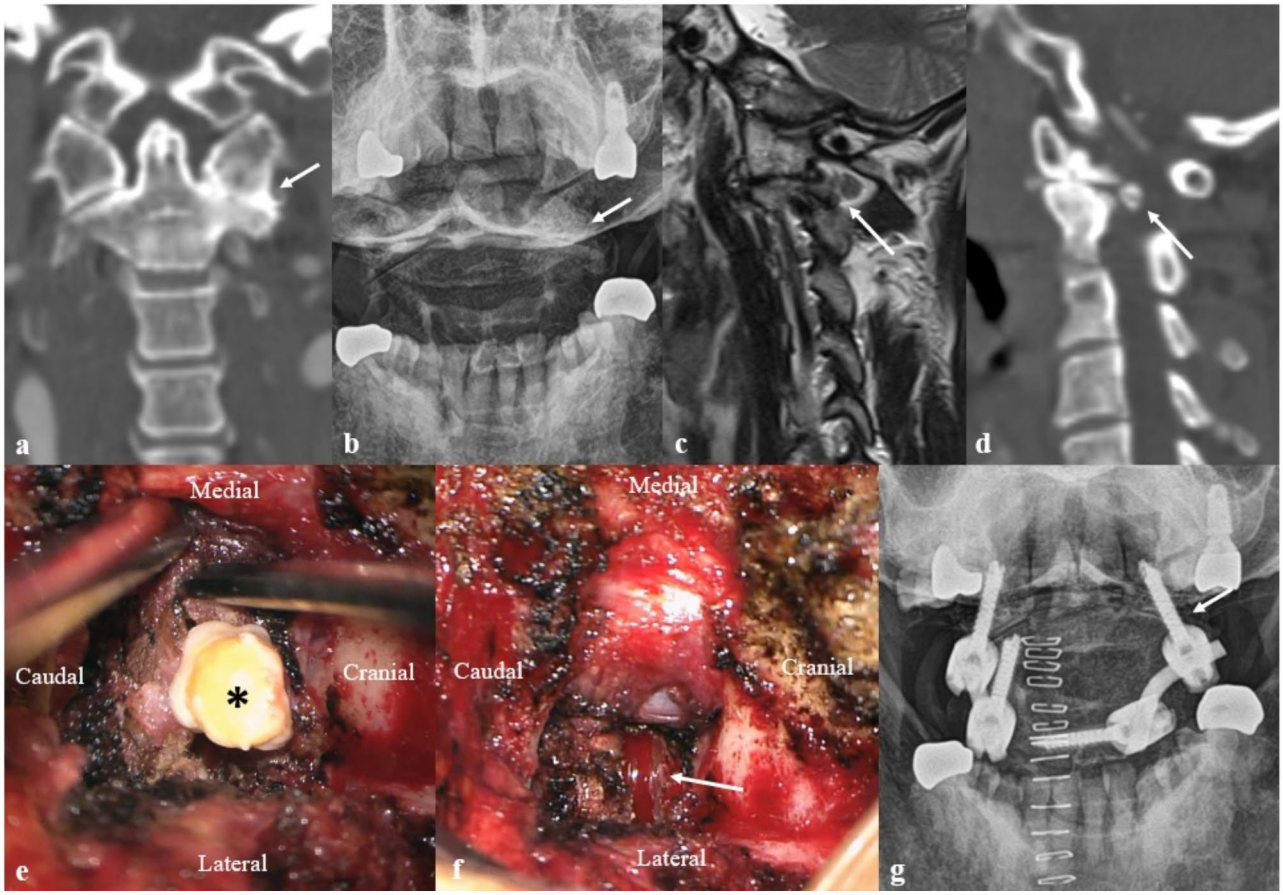

Fig. 1. Representative case of patient with atlantoaxial osteoarthritis with severe left occipital neuralgic pain who underwent C1-2 fusion surgery. (**a**,**b**) Open mouth X-ray image and computerized tomography (CT) image showed asymmetric sclerotic degeneration of left C1/2 facet joint. (**c**,**d**) Sagittal view of magnetic resonance imaging (MRI) and CT showed impingement of the left C2 root between left C1/2 foraminal space by bony spur and calcified ossicle. (**e**) The isolated left C2 nerve ganglion (*) is being removed after the C2 nerve root was cut proximal and distal to the ganglion. (**f**) After the C2 root transection, the fully exposed facet interspace was drilled for preparation of inter-facet allobone graft insertion. (**g**) After the arthrodesis surgery and inter-facet graft insertion, restoration of space between left C1/2 facet joint can be observed in post-operative X-ray.

ganglion. The allobone graft was inserted between left C1/2 facet joint space to elevate disc space and facilitate fusion process. Posterolateral fusion with allobone chips were also performed. Postoperative X-ray image showed stable fixation of C1/2 joint and restoration of space between left C1/2 facet joint (Fig. 1).

The patient reported immediate and complete relief of symptom after the surgery. The patient was taken under routine post-operative care and was discharged without any significant complication after full stitch-out of the wound, which was 9 days after the surgery. The patient was followed up until 9 months after the surgery at the out-patient clinic and reported persistent and complete relief of symptoms. However, the patient reported some decreased scalp sensation along left occipital nerve distribution, but it did not affect her daily activities or cause significant discomfort.

Case 10

A 70-year-old female presented with severe left occipital neuralgia. The duration of symptoms was 3 years and did not respond to any kind of conservative treatment. Physical examination showed typical presentation of occipital neuralgia, with pain aggravating with neck rotation to symptomatic side. CT image showed asymmetric degenerative change and fused status of left C1/2 facet joint and prominent bony spur causing left C1/2 foraminal stenosis. The MRI image also showed severe asymmetric left C1/2 foraminal stenosis and C2 root compression (Fig. 2).

Under the impression of left occipital neuralgia due to left C1/2 osteoarthritis with bony spur and left C2 root compression, the patient underwent a decompression surgery. The rationale for the choice of decompression surgery was (1) the already fused status of left C1/2 facet, which was evident in CT image, and (2) the prominent bony spur in C1/2 left foramen which was the main cause of pain and a definite target for decompression. For the decompression procedure, left C1/2 foraminotomy with bony spur removal was performed. The release of

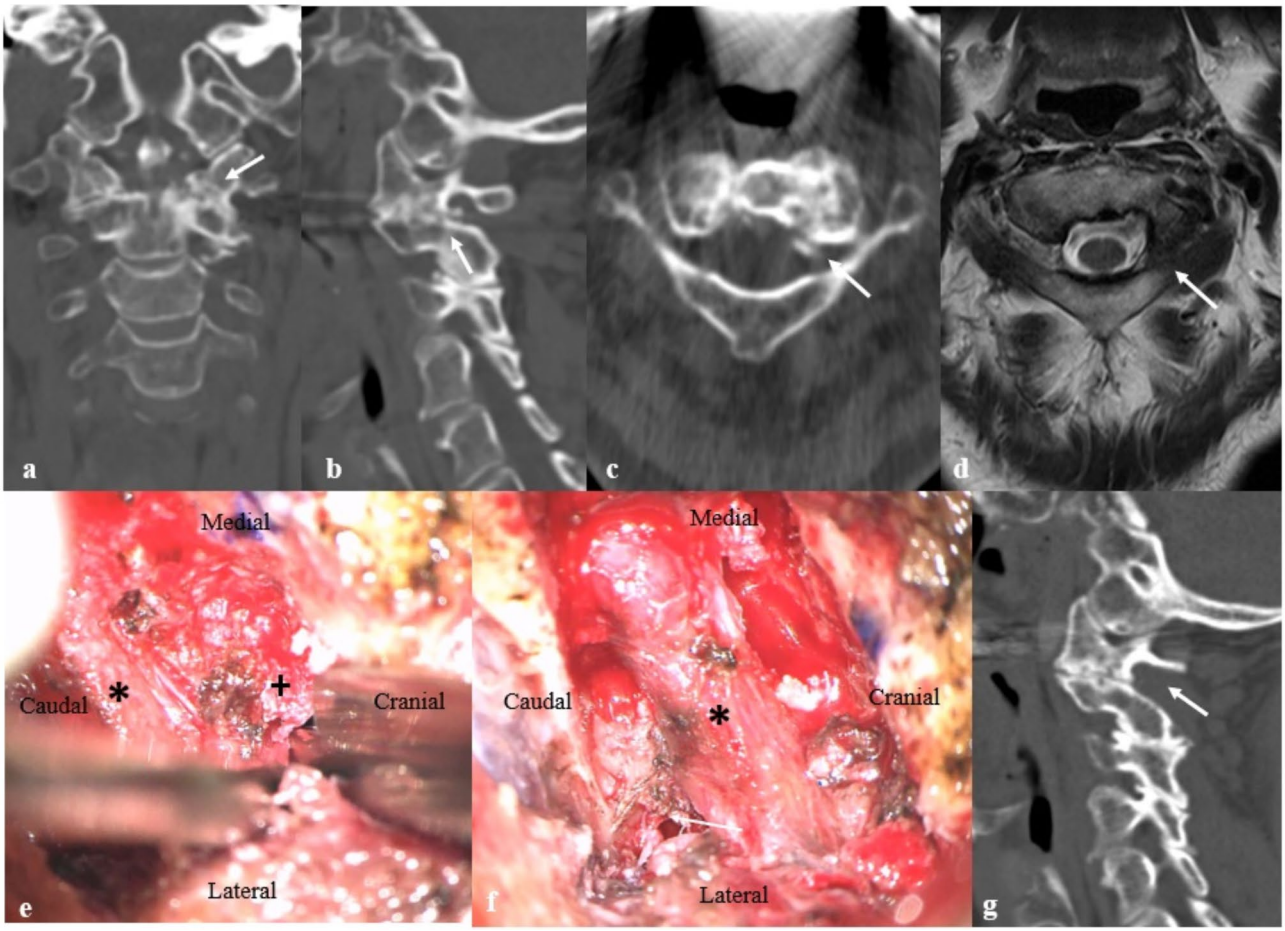

Fig. 2. Representative case of patient with atlantoaxial osteoarthritis with severe left occipital neuralgic pain who underwent decompression surgery. (**a**,**b**) Asymmetric degenerative change and fused status of left C1/2 facet joint can be seen on computerized tomography (CT) coronal and sagittal image. (**c**,**d**) The axial image of CT and magnetic resonance imaging (MRI) showing asymmetric left foraminal stenosis and severe left C2 root compression by bony spur. (**e**) During the decompression surgery, bony spur(+) compressing the C2 root(*) was noted and removed. (**f**) The left C2 root(*) was completely decompressed after the surgery. (**g**) Removed bony spur and decompressed status of C1/2 left foramen can be observed in post-operative CT.

left C2 root was confirmed, which had been compressed severely by the bony spur. The post-operative CT image showed removed status of the bony spur and decompressed C1/2 left foraminal space (Fig. 2).

The patient reported immediate relief of symptoms after the surgery. The patient was taken under routine post-operative care and was discharged without any significant complication after half stitch-out of the wound, which was 6 days after the surgery. The patient was followed up 1 month and 8 months after the surgery at the out-patient clinic. The patient reported improvement of the symptoms, but also reported some mild degree of residual tingling sensation and hyperesthesia on scalp.

Discussion

The intractable form of occipital neuralgic pain from AAOA that requires surgical intervention is the subject of only a few reports. References[1,5–7,14–16] Although there is still debate on the optimal surgical technique, fusion surgery has proven to be an effective method providing immediate and persistent pain relief[5–9]. According to a systematic review, up to 92.6% of AAOA patients after C1-2 fusion surgery reported improvement of pain[9]. Various fusion techniques have been described in the literature, including Gallie's wiring technique, Halifax interlaminar clamp, Margeryl's trans-articular screw fixation, and Harm's screw fixation and rod connection. This superior clinical outcome was achieved regardless of the fusion technique, with no significant difference between various types of technique[9]. As the AAOA patient's pain originates from the dynamic nerve root irritation by the degenerated C1/2 joint including bony spur and calcified granules, it is thought that fusion surgery effectively alleviates the pain by eliminating the micromotion of the degenerated C1/2 joint[3]. Since the introduction by Harms and Goel in 2001[17], screw fixation of C1 lateral mass and C2 pedicle with rod connection is currently the most widely used fixation technique with favorable fusion outcomes and relatively less complications. Thus, the most recent reports on AAOA, including ours, utilized this fusion method with favorable clinical outcomes.

Patient number	Age & gender	Left or right	Duration of symptom (years)	Cervical instability	Fusion or decompression	Instrumentation	Graft	C2 root transection	Follow up periods (months)	VAS (preop)	VAS (postop)	Patient-reported outcome	Complication (surgical)	Complication (clinical)
1	74 F	Right	2	No	Fusion	C1 bilateral lateral mass & C2 Rt pedicle & Lt laminar screw	Right interfacet allobone graft	Yes - Right	1	10	1	Improved pain		
2	81 M	Left	3	Yes	Fusion	C1 bilateral lateral mass & C2 Rt pedicle & Lt laminar screw	Bilateral interfacet allobone graft	Yes - Bilateral	6	9	0	Pain disappeared completely		Scalp sensory decrease
3	56 F	Left	2	Yes	Fusion	C1 bilateral lateral mass & C2 bilateral pedicle screw	Bilateral interfacet allobone graft	Yes - Bilateral	3	10	0	Pain disappeared completely		Scalp sensory decrease
4	59 F	Right	3	No	Decompression			No	1	8	1	Improved pain		
5	78 M	Right	20	No	Fusion	C1 bilateral lateral mass & C2 Rt lamina & Lt pedicle screw	Right interfacet allobone graft	Yes - Bilateral	3	8	2	Improved pain	Vertebral artery injury	
6	65 F	Left	5	No	Fusion	C1 bilateral lateral mass & C2 Rt pedicle & Lt laminar screw		Yes - Left	10	8	0	Pain disappeared completely		Scalp sensory decrease
7	67 M	Left	2	No	Fusion	C1 bilateral lateral mass & C2 Rt pedicle & Lt laminar screw		Yes - Left	4	9	0	Pain disappeared completely		
8	80 F	Left	1	No	Fusion	C1 bilateral lateral mass & C2 Rt pedicle Lt laminar screw		Yes - Left	3	10	1	Improved pain		Scalp sensory decrease
9	62 F	Left	15	Yes	Fusion	Suboccipital plate & C1 bilateral lateral mass & C2 bilateral pedicle screw	Left interfacet allobone graft	Yes - Left	3	9	2	Improved pain		
10	70 F	Left	3	No	Decompression			No	8	8	4	Improved pain		Scalp tingling sensation and hyperesthesia
11	72 M	Right	1	No	Fusion	C1 bilateral lateral mass & C2 Rt pedicle & Lt laminar screw		Yes - Right	1	10	2	Improved pain		

Table 1. Description of patients.

In our study, adjunct C2 root transection on the symptomatic side was performed in all cases of C1-2 fusion surgery. Goel et al. first officially introduced routine bilateral sectioning of the C2 root ganglion during C1-2 fixation surgery to facilitate screw fixation, joint decortication, and arthrodesis procedure[18,19]. C2 root transection during C1-2 fixation surgery has proven to be a valid treatment option for various high cervical diseases without definite complications[10–13]. Excellent fusion rates and favorable patient satisfaction have been reported after C1-2 instrumented arthrodesis with C2 neurectomy[10]. Furthermore, Dewan et al. reported that C2 root transection had no negative consequences during C1-2 stabilization[11]. One of the most common complications is the decreased sensation on the scalp skin area along the distribution of the C2 root[12,13]. However, most studies reported that those complications did not induce significant discomfort to the patients. Our study agrees with this, with four patients who underwent arthrodesis with C2 root transection reporting decreased sensation on the scalp but without significant discomfort. Although it is reasonable to conclude that C2 root transection during arthrodesis can provide favorable outcomes without definite complications, whether it should be preferred over C2 root preservation remains controversial. C2 root transection may not be required if adequate screw fixation is achieved without definite mechanical root compression or decompression. Moreover, destruction of the normal anatomy should be avoided if possible.

In spite of the controversies, we suggest that transection of the C2 root can provide several surgical and clinical advantages, especially in patients with AAOA[3,10,19]. First, the C2 root in most patients with AAOA who require surgical treatment is severely compressed by degenerated bony spurs and calcified soft tissues around the C1/2 facet joint. Removal of these degenerated structures while preserving the C2 root can be surgically challenging, and C2 root sacrifice may be inevitable for complete removal of the degenerated structures. In addition, the C2 root is mostly denatured owing to prolonged compressive injury over a significant amount of time. This implies that decompression of the C2 root can induce residual symptoms due to chronic neuropathic pain, which can be avoided if C2 root transection is performed. Moreover, the C2 root transection effectively exposes the C1/2 joint space, enabling the surgeons to decorticate the joint surface and insert additional bone grafts into the joint space with ease. In cases of severe asymmetric C1/2 arthritis, insertion of an interfacet graft may be crucial for the restoration of the collapsed facet joint space and regaining symmetric balance (Fig. 1). The inserted bone graft can also facilitate fusion process between the C1/2 facet joint after surgery. Since micromotion of the degenerated C1/2 joint plays an important role in the pathophysiology of AAOA, complete fusion and elimination of micromotion are crucial for optimal clinical outcomes. Additionally, C2 root transection enables surgeons to effectively coagulate the large venous plexus adjacent to the C2 root, leading to significantly less intraoperative blood loss. Finally, complications of occipital neuralgic pain due to mechanical nerve compression by an adjacent fixated screw, which is seldom reported as a cause of persistent neuralgia after C1-2 fusion with preservation of the C2 root, can be completely avoided. Thus, C2 root transection may be preferred, particularly in patients with AAOA, if the surgeon aims to achieve a perfect fusion rate and complete elimination of neuralgic pain after instrumentation.

During C2 root transection, the anatomical location and transection technique are important for optimal clinical results. The clinical results of C2 root transection may be controversial due to the inconsistent and poor description of surgical techniques in the literature[20]. Neuronal sprouting or axonal regeneration may occur after incomplete or inadequate C2 neurectomy and can lead to persistent and intractable neuralgic pain[21,22]. In our institution, the C2 ganglion was first identified and coagulated by bipolar cautery at both proximal and dorsal to the ganglion. Stepwise cutting was performed using sharp scissors or a blade and the isolated C2 ganglion was removed (Fig. 1). As the dorsal root ganglion plays an important role in the transmission of painful sensations and can exhibit regenerative capacity, complete ganglionectomy should be performed during C2 transection[23].

Decompressive treatment without additional instrumentation may be a valid option for selected patients. Fujiwara et al. reported 3 cases of AAOA that were successfully treated with microscopic posterior foraminotomy[24]. In our study, two patients underwent decompressive surgery with preservation of the C2 root. One patient had an already fused status of the C1/2 facet joint, which was identified on CT imaging (Fig. 2), and a bony spur was noted to be the main cause of C2 root compression and a definite target for decompression. Another patient had a fused status from the clivus to C2, which was identified on CT imaging, and no instability was noted on dynamic radiography. The most important indication for decompressive surgery in these two patients was the already fused status of the high cervical area, thus making an additional fusion process unnecessary. Because the micromotion of the degenerated C1/2 joint and dynamic compression of the nerve root are the main pathophysiologies of AAOA[3], it is important to note that immobilization of the degenerated joint is crucial for the complete alleviation of pain. Although both patients reported improved symptoms compared with the preoperative baseline, one patient reported complications of a residual tingling sensation and some degree of scalp hyperesthesia. This may be due to excessive C2 root manipulation during decompression and persistent dynamic compression by residual micromotion of the C1/2 facet joint. Therefore, fusion surgery should be considered for the surgical treatment of patients with AAOA; however, decompressive surgery may be considered for the selected patients who already have fused joints and are medically intolerable to aggressive surgery.

Our study has some limitations. First, only a limited amount of data was available in our study. However, refractory occipital neuralgia due to AAOA is relatively rare, particularly in patients requiring surgical intervention. Most of the literature reports only a few similar cases, and our number of patients is similar to that of these previous studies. We believe that this is the first study to focus on fusion surgery with C2 root transection for AAOA. Second, our data were collected retrospectively, and various biases cannot be ruled out during data collection. However, all patients were enrolled in a single institution and performed by a single surgeon; therefore, variations in the surgical procedures between each institution and surgeon could be eliminated. In addition, a control group that underwent fusion surgery without transection of the C2 root was not established. Therefore, the accurate beneficial impact of C2 neurectomy could not be assessed in this study. Finally, the follow-up period in our study was relatively short. This is because we did not recommend additional clinical visits after the patient

was confirmed to have persistent pain relief with a stable fusion status. However, long-term follow-up data are required to assess the various long-term complications and fusion rates. Further comparative studies with larger sample sizes and long-term follow-ups are needed to accurately assess the beneficial impact of arthrodesis with C2 root transection in patients with AAOA.

Conclusion

Occipital neuralgia due to AAOA is an important disease entity that can be diagnosed using appropriate imaging assessment and physical examination. C1-2 fusion surgery is an excellent surgical treatment option and should be considered for patients with refractory AAOA. C2 root transection on the symptomatic side during C1-2 fusion can completely alleviate pain at the expense of minor scalp anesthesia. Decompressive-only surgery is not recommended but can be considered in strictly selected patients.

Data availability

All data generated or analyzed during this study are included in this published article.

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Author contributions

Dongkyu Kim: Data curation/Visualization/Writing - original draft. Keun Su Kim: Conceptualization/Supervision/Writing - review & editing.

Declarations

Competing interests

The authors declare no competing interests.

Ethics declarations

This retrospective study was approved by Institutional Review Board of Gangnam Severance Hospital, Yonsei University Health System, which waived the requirement for informed consent.

Additional information

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