Percutaneous full-endoscopic C2 ganglionectomy for the treatment of intractable occipital neuralgia: technical feasibility and preliminary results

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Research Article

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Abstract

Background

The treatment algorithm for occipital neuralgia follows the ladder principle. For intractable occipital neuralgia, C2 ganglionectomy can be an effective treatment option. Advancements in percutaneous endoscopic spinal surgery make it possible to accomplish C2 ganglionectomy using a full-endoscopic uniportal surgical technique.

Methods

From March 2020 to April 2021, three consecutive patients with intractable occipital neuralgia underwent percutaneous full-endoscopic C2 ganglionectomy. All the patients were followed up for more than 12 months. Previous treatment measures, operative time, blood loss, length of hospital stay, and complications were recorded. Finally, the visual analog score (VAS) and Headache Disability Inventory (HDI) were utilized to evaluate surgical efficacy.

Results

All the patients were successfully treated with a percutaneous full-endoscopic C2 ganglionectomy. Both the postoperative VAS scores and the HDI scores improved after C2 ganglionectomy in the three patients. There was no CSF leakage, incision infection, neck deformity, or other complications.

Conclusions

Patients suffering from intractable occipital neuralgia may benefit from percutaneous endoscopic C2 ganglionectomy, which possesses several advantages such as excellent visualization and reducing surgery-related trauma, blood loss, and length of stay.

Background

According to the guidelines of the International Classification of Headache Disorders, 3rd Edition (ICHD–3), occipital neuralgia (ON) refers to a headache characterized by paroxysmal shooting, stabbing, or sharp pain in the distribution of the greater, lesser, or third occipital nerves [1]. Other features of ON are associated with the following: trigger points at the emergence of the great occipital nerve, tenderness over the affected nerves, and can be temporarily blocked by local anesthetics [2].

In patients with occipital neuralgia, physical therapy and drug pharmacologic treatment play primary roles in managing the symptoms [2]. For patients with refractory symptoms, further treatment options

may include nerve block, pulsed radiofrequency, etc. However, surgical intervention may be indicated for patients unresponsive to the above-mentioned treatment modalities [2, 3].

Previous pieces of literature have established that C2 ganglionectomy is an effective option for intractable occipital neuralgia [4–6]. However, the traditional microsurgery of C2 ganglionectomy requires a wide incision and significant paraspinous muscle dissection for adequate visualization, resulting in iatrogenic occipital neck pain and a slow recovery. It is one of the reasons why this technique is not frequently employed by surgeons. Percutaneous full-endoscopic C2 ganglionectomy has been developed to overcome these shortcomings [7], in which C2 ganglionectomy can be executed through the slim working channel under continuous irrigation (Fig. 1). Moreover, the skin incision is less than 8 mm. In this article, we presented three cases with intractable occipital neuralgia who underwent percutaneous full-endoscopic C2 ganglionectomy. All the cases were followed up for more than 1 year. The report aimed to validate the feasibility of this technique, as well as describe several operative nuances and pearls based on our experience.

Methods

The study was approved by the ethics committee of Zhongshan Hospital, Fudan University (Institutional Review Board approval number B2020-319R) in accordance with the guidelines of the 1964 Declaration of Helsinki. The patients signed informed consent forms before the operation. Clinical data used or analyzed in this study are available from the corresponding author upon reasonable request.

Patients

All three patients were referred for the treatment of their medically intractable occipital neuralgia. ON was diagnosed according to the criteria put forth by the International Classification of Headache Disorders, 3rd Edition (ICHD–3) [1]. The three patients had previously undergone a series of treatments in other hospitals, including medical management, physical therapy, Chinese Acupuncture, local anesthetic injection, and pulsed radiofrequency (PRF), all of which failed to provide long-term pain relief. The detailed medical history, physical examination, and imaging records (including cervical radiographs, 3D-CT reconstructions, and magnetic resonance images) were collected and thoroughly analyzed. No structural lesion was visualized on imaging in any of the patients in the series that was thought to be the source of occipital neuralgia. It is worth mentioning that there was no specific cause of ON in case 1 and case 2, while case 3 was caused by head trauma. Case 3 also developed facial spasms in addition to ON following the traumatic event. Prior to C2 ganglionectomy, all three patients underwent a diagnostic C2 ganglion block performed with 0.5% lidocaine. Pain relief of 90% or more and lasting for at least 30 mins were considered a positive response.

Endoscopic Instruments

The endoscopic surgical system TESSYS (Joimax GmbH, Karlsruhe, Germany), including an endoscope, endoscopic sheaths, nucleus pulposus clamps, basket forceps, scissors, etc. were employed to perform

the surgery. A radiofrequency probe (Trigger-FlexR Bipolar System, Elliquence LLC, Baldwin, New York) was utilized to control bleeding and for ablation.

Operative Technique

The operation was executed under general anesthesia. The patient was placed in the prone position with the head fixed in a Mayfield skull clamp. The operator stood on the ipsilateral side of the pathology [7]. After the patient was routinely sterilized and draped, an 18-gauge spinal needle was used to puncture under anteroposterior and lateral fluoroscopy. In the final position, the needle tip was in contact with the inferior articular process of the C2. The needle was then replaced by a guidewire that passed through it. Next, a 7-mm stab incision was made in the skin centered on the guidewire. Afterward, the soft tissue was progressively expanded by soft tissue dilators, and a working cannula was inserted along the dilator. The position of the working cannula was confirmed by anteroposterior and lateral fluoroscopy (Fig. 2A, B). Next, the endoscopic surgical system was introduced through the cannula, and all the subsequent operational steps were performed under the monitoring of high-definition endoscopic visualization.

After the soft tissue was cleared and ablated using a radiofrequency probe, the inferior articular process and pedicle of C2 were identified. With the assistance of the “pedicle guidance technique” [7], the lateral atlantoaxial articulation was identified. The C2 ganglion was positioned posterior to the atlantoaxial joint cavity (Fig. 3A).

The C2 nerve root and ganglion were explored and carefully dissected with the radiofrequency probe. Afterward, basket forceps or scissors were used to partially excise the distal end of the ganglion (Fig. 3B, 4A). The connection could keep the ganglion from drifting in the water. Next, the proximal end of the ganglion was coagulated and completely cut off (Fig. 3C, 4B). Finally, nucleus pulposus forceps were employed to grasp the ganglion and drag it out (Fig. 3D, 3E, 4C). The bipolar radiofrequency probe was used to control bleeding and ablate nerve stumps. No drainage was placed, and the incision was closed with 2 stitches (Fig. 3F).

Evaluation of Surgical Outcomes

Operative time, blood loss, length of hospital stay, and complications were recorded and investigated. Pain was preoperatively and postoperatively evaluated using the visual analog scale (VAS) that rated each patient's pain from 0 to 10. The Headache Disability Inventory (HDI) assessed the patient’s daily activities and participation in society. Surgical outcome data for all patients were acquired by an investigator not involved in this study. The postoperative scores (on day 1 and at 3, 6, and 12 months) were compared to the preoperative baseline.

Results

Demographic Results

Between March 2020 and April 2021, three consecutive patients with intractable occipital neuralgia were successfully treated with percutaneous full-endoscopic ganglionectomy. Table 1 outlines the baseline demographics of the patients, as well as surgical site, operative time, blood loss, and length of hospital stay.

Table 1. Demographic findings of the 3 cases

Characteristic \ Case		Case1	Case2	Case3
Gender		Female	Female	Male
Age (yr)		83	61	50
Etiology		Not clear	Not clear	Trauma history
Duration of symptoms (months)		24	384	3
Previous treatment measures	Medication	✓	✓	✓
	Chinese acupuncture	✓	✓	
	Physical therapy	✓	✓	✓
	Nerve blocks	✓	✓	✓
	Pulsed radiofrequency	✓	✓	✓
Side of the surgery		Left	Left	Left
Operation time (min)		60	50	50
Blood loss (mL)		5	5	5
Hospital stay (day)		2	2	3

Clinical Results

The VAS scores for each of the patients at the time of admission were 9/8/9, respectively. 1 day following the surgery, the VAS scores were 3/2/2, respectively. At 1-month, 3-month, 6-month, and 12-month follow-ups, the VAS scores were 2/1/3, 3/2/3, 2/2/3, and 1/2/4, respectively. Case 1 had a VAS score of 1 and had been followed for more than 20 months (Fig. 5). The HDI score improved after C2 ganglionectomy in all three patients. In case 3, facial spasm did not improve following the surgery, although occipital neuralgia was relieved. The patient stated that facial spasms severely negatively impacted his mood and made him feel anxious; this was also why his HDI score was relatively poor at the last follow-up (Fig. 6).

Surgical Complications

There was no CSF leakage, incision infection, neck deformity, or other complications other than transient occipital numbness in cases 1 and 3. The numbness was mild, marginally affected daily life, and gradually disappeared over the course of a week without any specific treatment.

Representative case illustration

A 61-yr-old female complained of left-sided stabbing pain in her neck and occipital region over the past 16 years. The pain was exacerbated when her head was in a dorsiflexed position. She was unable to lay her head on a pillow at night owing to the pain. Besides, she had no prior history of head and cervical spine injuries. Physical examination revealed left C1-2 paravertebral tenderness and referred pain to the left occipital and retro-orbital areas. Cervical dynamic X-ray radiographs were unremarkable. Cervical magnetic resonance imaging (MRI) and computed tomography (CT) showed no evidence of cervical disc herniation, space-occupying lesions, or cervical spondylolisthesis (Fig. 7). She was initiated on pharmacological treatment and physical therapy, but the pain was not alleviated. She also received a local anesthetic injection six times and pulsed radiofrequency (PRF) treatment twice in another hospital without satisfactory results. She had been experiencing progressively worsening pain for the last year and rated the pain as 8/10 on the visual analog scale (VAS) when she was admitted to the hospital.

Prior to surgery, she underwent a diagnostic C2 ganglion block, which provided significant pain relief (Fig. 8). Afterward, she underwent percutaneous full-endoscopic C2 ganglionectomy in our hospital. After the procedure, the pain notably decreased to a VAS score of 2/10. At the 1,3,6, and 12-month follow-ups, the VAS score was 1, 2, 2, and 2, respectively. No complications such as infection at the incision site, neck deformity, or movement disorders were observed during the study.

Discussion

According to the definition of the International Headache Society (IHS) 3rd Edition (ICHD–3), the diagnostic criteria for occipital neuralgia (ON)are as follows: A. Paroxysmal stabbing pain, with or without persistent aching between paroxysms, in the distribution of the greater, lesser, and/or third occipital nerve B. Tenderness over the affected nerve C. Pain is eased temporarily by a local anesthetic block of the nerve [1]. Hence, the diagnosis of ON mainly relies on the scope, properties, and characteristics of the pain. ON usually originates from the suboccipital region and spreads throughout the back of the head. However, the pain can involve other areas, such as the retro-orbital area, due to the

convergence of the nucleus trigeminus pars caudalis [8, 9]. This pain is usually unilateral, but bilateral exits. During physical examination, tenderness is typically detected by palpation. Besides, Tinel and pillow signs may be evoked [2]. All three cases in our series fulfilled these characteristics. For example, the pain extended to the periorbital region in case 2. Tinel and pillow signs were apparent during her physical examination.

The treatment algorithm for ON should generally be in accordance with the ladder principle. That is to say, conservative therapy involving physical and drug medications should initially be considered. Patients with symptoms refractory to conservative therapy may be treated with percutaneous anesthetic injections with/without steroid medications. However, most C2 nerve blocks commonly provide short-term pain relief, with approximately 15–36% sustaining extended relief for several months [10]. In our series, the three patients had undergone C2 nerve blocks more than once in other hospitals without a satisfactory duration of pain relief. The procedure can be performed either distally at the nuchal line (commonly used method) or proximally between C1 and C2, where the C2 ganglion is located [11]. We recommend the latter before C2 ganglionectomy, given that it can more accurately reflect the effect of C2 ganglionectomy in theory. All three patients underwent C2 ganglion block, and the results were favorable.

Pulsed radiofrequency (PRF) as a treatment for occipital neuralgia has yielded promising clinical results, although evidence validating its long-term effects is lacking [12–14]. This minimally invasive surgical option may be considered for patients failing conservative and injection therapies. As a minimally invasive percutaneous technique, PRF therapy exposes the nerve to high-voltage radiofrequency pulses, which is speculated to induce an inhibitory electrical field around the sensory nerves, disrupting pain transmission and potentiation with none to minimal neurodestruction [15]. Herein, the three patients had undergone PRF in other hospitals before hospitalization in our hospital. Among them, case 2 had the longest period of pain relief, lasting around 3 months.

Occipital nerve stimulation (ONS) is an alternative treatment option for patients with medically refractory ON [16–17]. The Congress of Neurological Surgeons 2015 Guideline recommends the use of ONS for the treatment of intractable occipital neuralgia (level III) [18]. ONS affects the subcutaneous insertion of electrodes in the C1/C2 regions of the posterior cervical spine. It is attractive as a non-destructive, reversible therapeutic approach with long-term efficacy. However, several potential complications related to ONS include lead migration, electrode fractures, hardware erosions, disconnections, and sepsis [2, 19, 20]. Moreover, some other factors limit its application, such as high medical costs, lack of medical insurance coverage in some regions, and the patient's religious belief that implants are not acceptable in their body. In our three cases, we recommended ONS to all the patients and thoroughly outlined the pros and cons of this technique. Case 1 did not consent to ONS because of her religious belief, and the other two cases did not choose this approach given its exorbitant cost.

Patients with symptoms refractory to the above medical procedures may be subjected to increasingly invasive surgical modalities. The ideal surgical management remains to be elucidated. Alternative surgical methods include occipital nerve decompression [21], neuroablation [12, 13, 22], partial rhizotomy

[20], peripheral neurolysis [24], and C2 to C3 root decompression [25]. However, these procedures are commonly associated with pain recurrence from aberrant postoperative neuronal regeneration and neuroma formation [2, 5, 23, 25].

C2 dorsal root ganglionectomy was developed to address the failures of the above procedures [4–6, 25]. For patients with intractable ON, C2 dorsal root ganglionectomy is a viable surgical option. Theoretically, the removal of the primary sensory neurons residing in the C2 ganglion is more conducive to preventing pain recurrence [2, 5, 25–26]. Previous pieces of literature have established that this procedure is successful in treating the majority of intractable ON.

However, traditional C2 ganglionectomy is performed under microscopy [4, 5, 25], which necessitates a wide incision due to the deep location of the C2 ganglion. The incision of the skin usually extends from the occiput to the spinous process of C3 or C4. A large amount of paraspinal muscle is stripped from the bone to expose the C1 posterior arch and C2 dorsal element [4, 5], which may result in surgery-induced neck pain owing to intraoperative iatrogenic damage and postoperative scar of muscle tissues. Occasionally, surgery-induced neck pain can often be difficult to distinguish from preoperative occipital neuralgia in patients, influencing the evaluation of the effectiveness of surgery. In addition, brisk bleeding from an exuberant venous plexus enfolding the C-2 ganglion is commonly encountered during open surgery. It usually requires meticulous and extensive electrocoagulation, which also induces damage to adjacent nerve branches.

These shortcomings may be overcome by virtue of the properties of the percutaneous full-endoscopic surgery technique. The skin incision is less than 8 mm, without the need to strip the paravertebral muscles. Through the slim working cannula, the camera's eye with 30 view angles can be inserted millimeters away from the C2 ganglion, delivering high-definition images for the operator on a monitor. All actions are executed through the working channel under excellent endoscopic visualization without requiring a wide wound exposure. Hence, postoperative surgery-induced neck pain can be alleviated. Moreover, the surgical procedure is performed under continuous irrigation. The pressure of the irrigation fluid can help minimize bleeding from the venous plexus enfolding the C2 ganglion. Extensive cauterization is not required, reducing injury to adjacent nerve branches. In our case series, all the patients experienced a satisfactory therapeutic response. The VAS scores were maintained at low levels after a 1-year follow-up, further corroborating the validity and merits of percutaneous full-endoscopic C2 dorsal root ganglionectomy.

There are two key steps in percutaneous full-endoscopic C2 ganglionectomy. The first is to locate and confirm the location of the C2 ganglion, and the second is to effectively and safely excise the ganglion under endoscopy. In our previous study, we introduced the "pedicle guidance technique" to accomplish the first objective [7], that is, by following the superior edge of the C2 pedicle, the operator can reach the lateral atlantoaxial articulation, behind which the C2 ganglion is located. The ganglion can be identified as a large bulge about 4 mm away from the dural sac and proximal to the C2 primary rami (Fig. 3A). Endoscopic spine surgery is characterized by one hand and one instrument, unlike microscopy surgery.

Understanding the characteristic mentioned above is critical to accomplishing the second goal. Therefore, when firstly cutting the distal end of the ganglion, the distal end should not be completely excised. The reserved partial connection can maintain the ganglion in situ (Fig. 3B). Otherwise, the ganglion will float in the water flow, leading to challenges in accurate proximal resection.

Our experience has proved that percutaneous full-endoscopic C2 ganglionectomy can be successfully accomplished. It converted an open surgery into a percutaneous puncture surgery. This will make it easier for surgeons to choose among the alternative surgical options for ON while also increasing “Return on Investment”. The primary limitations of this study were its retrospective nature, the limited sample size, and its relatively short follow-up period. Despite the aforementioned limitations, we believe that our description of the operative nuances about endoscopic C2 ganglionectomy and the introduction of clinical features about ON cases will assist in clinical decision-making. In the future, a larger number of cases and more strictly designed experiments are needed to corroborate that the “returns” have definitely been increased.

Conclusion

C2 ganglionectomy can be accomplished successfully using a full-endoscopic uniportal surgical technique under continuous irrigation. Percutaneous full-endoscopic C2 ganglionectomy has several benefits, such as excellent visualization, reducing surgery-related trauma, blood loss, and length of stay. Patients suffering from intractable occipital neuralgia can get favorable pain relief following a C2 ganglionectomy.

List Of Abbreviations

CSF, cerebrospinal fluid; CT, computed tomography; VAS, visual analog score; HDI, headache disability inventory; ICHD–3, international classification of headache disorders-3rd edition; IHS, International Headache Society; MRI, magnetic resonance imaging; ON, occipital neuralgia; ONS, occipital nerve stimulation; PRF, pulsed radiofrequency

Declarations

Acknowledgments

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Authors' contributions

C.L and Y.J contributed equally to this work. C.L, Y.J, Q.W, F.X, L.Y, Y.M, T.L, L.Y, S.L, and Y.Y contributed to drafting of the manuscript and critical revision of the manuscript for important intellectual concepts. L.Y and S.L prepared figures 1-8. The authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and analysed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

The study was approved by the ethics committee of Zhongshan Hospital, Fudan University (Institutional Review Board approval number B2020-319R) in accordance with the guidelines of the 1964 Declaration of Helsinki. The patients signed informed consent forms before the operation.

Consent for publication

Not applicable.

Competing interests

none

Authors' information

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Figures

Figure 1

Illumination for percutaneous full-endoscopic C2 ganglionectomy.

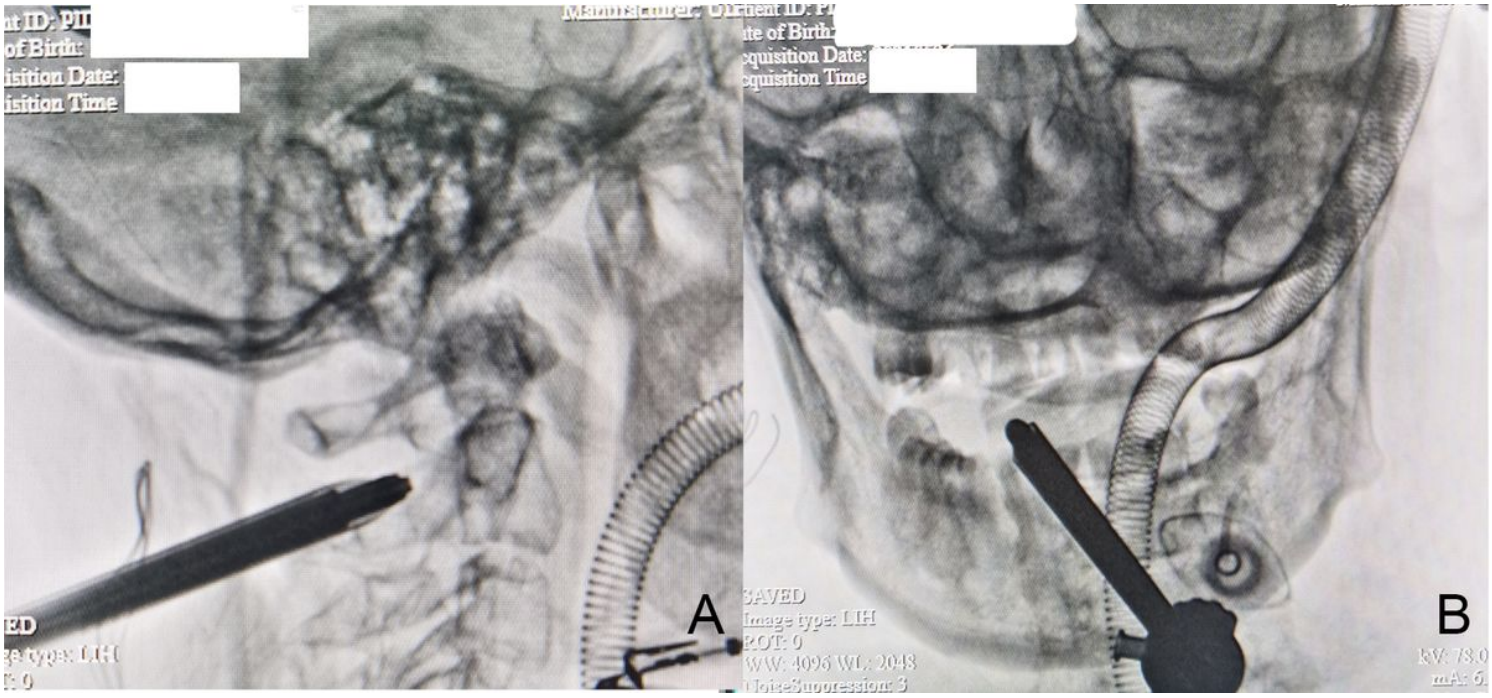

Figure 2

Lateral (A) and anteroposterior (B) fluoroscopy confirmed the position of the working cannula.

Figure 3

Percutaneous full-endoscopic C2 ganglionectomy. A, Endoscopic view of the C2 ganglion, dural sac, and C2 pedicle (a, represents the C2 ganglion; b, represents the dural sac between the lamina of C1 and C2; c, represents the C2 pedicle). B, partially resecting the distal end of the ganglion with basket forceps. C, excising the proximal end of the ganglion with a scissor (a, represents the C2 ganglion). D, Endoscopic view of the C2 ganglion. The proximal end of the ganglion was completely severed (a, represents the C2 ganglion). E, grasping the C2 ganglion and dragging it out. F, The skin incision was closed with 2 stitches.

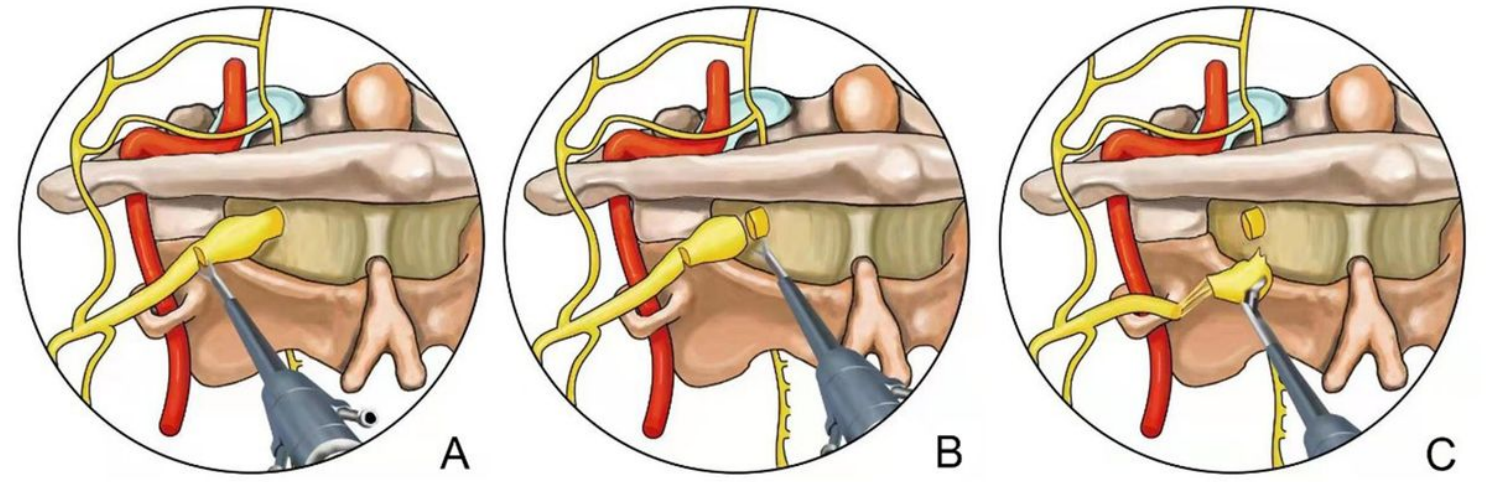

Figure 4

Schematic diagram illustrating the sequential steps for the endoscopic removal of the C2 ganglion. A, the distal end was firstly partially cut. There was still a little connection that kept the ganglion from drifting. B, The proximal end of the ganglion was completely resected. C, Nucleus pulposus forceps were used to grasp the ganglion and drag it out.

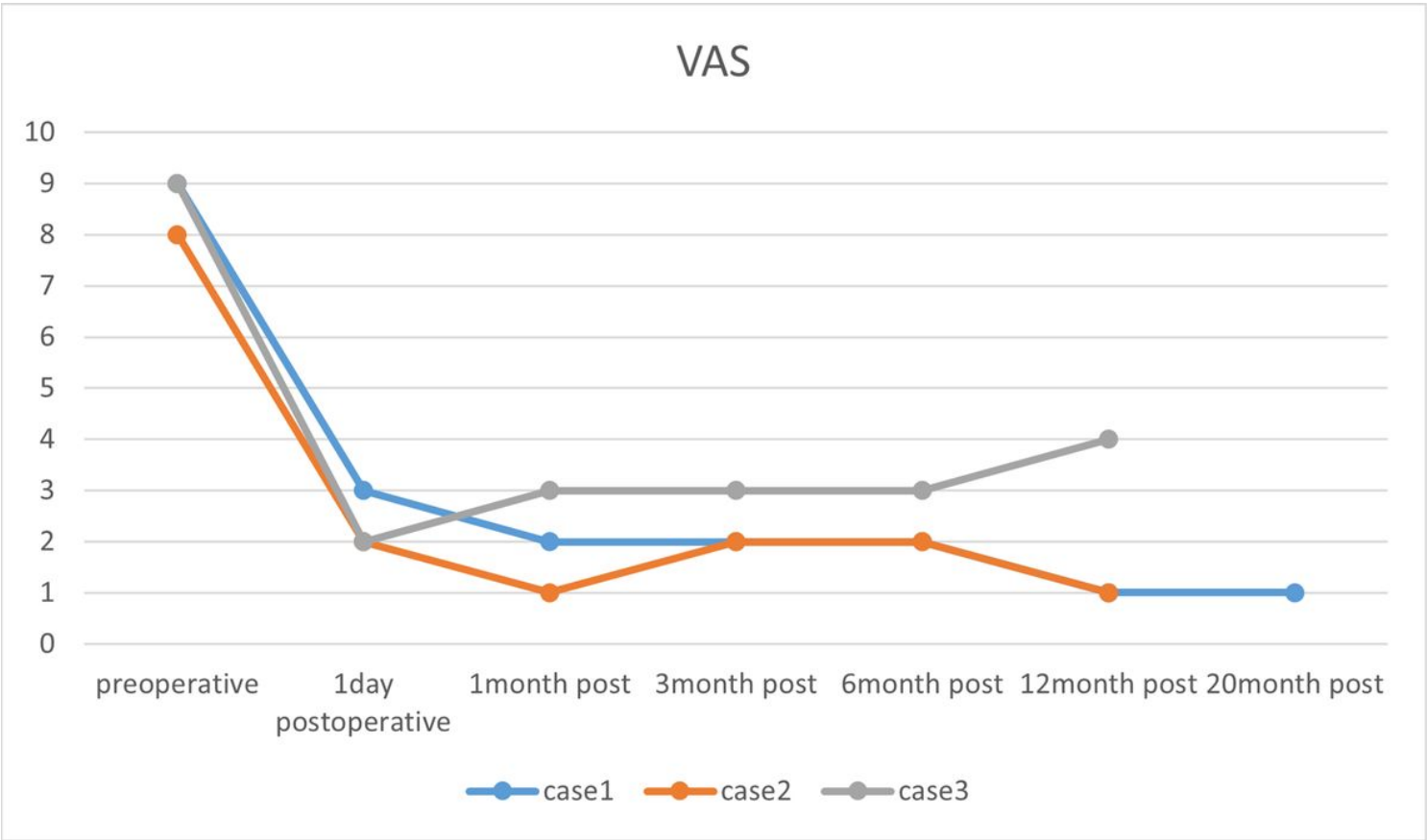

Figure 5

Diagram depicting pre- and postoperative alterations in the VAS score of the three patients.

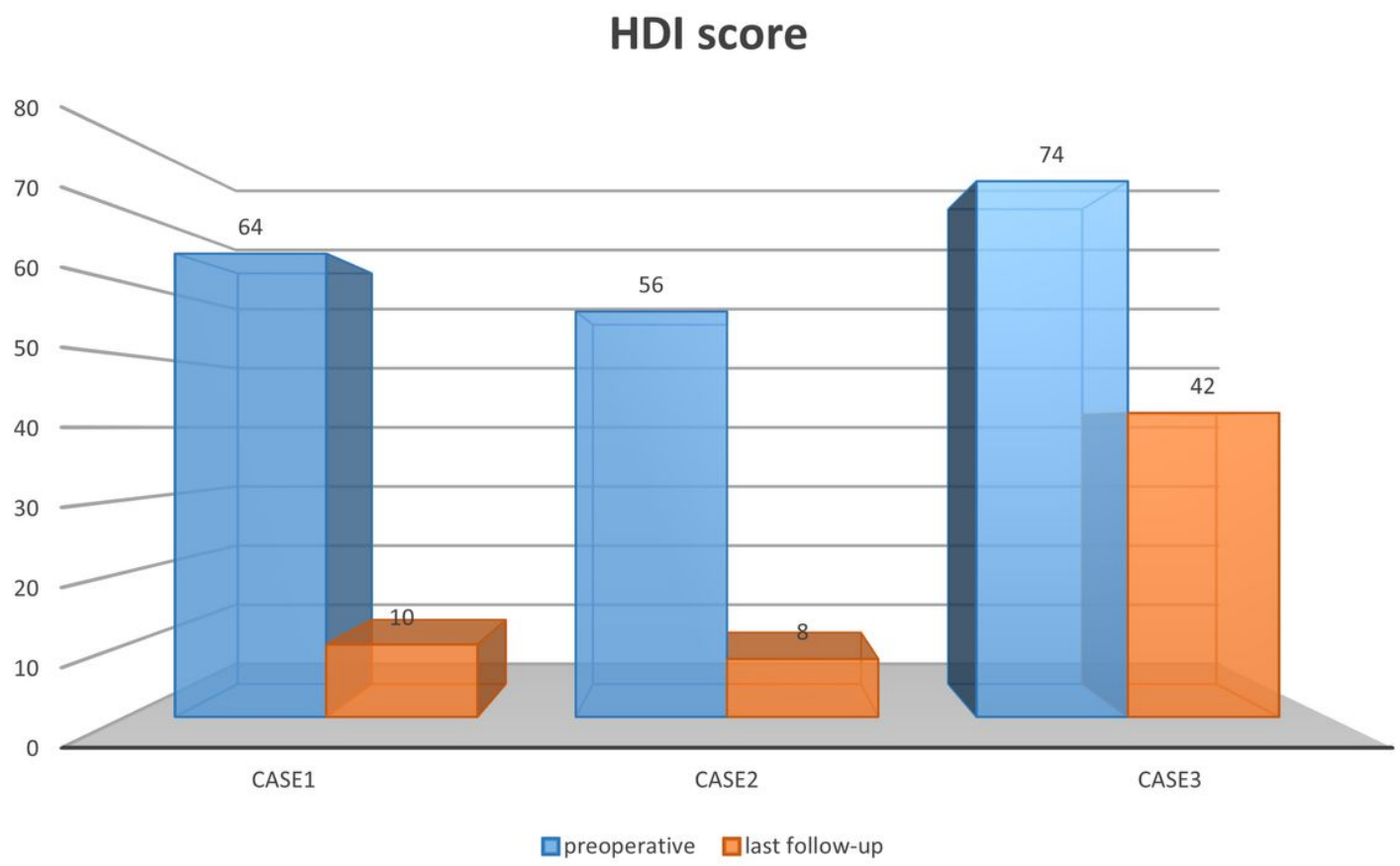

Figure 6

HDI scores in quality of life measures before the procedure and up to the last follow-up after the procedure. A reduction of ≥16 points in total HDI was utilized as a measure of treatment success.

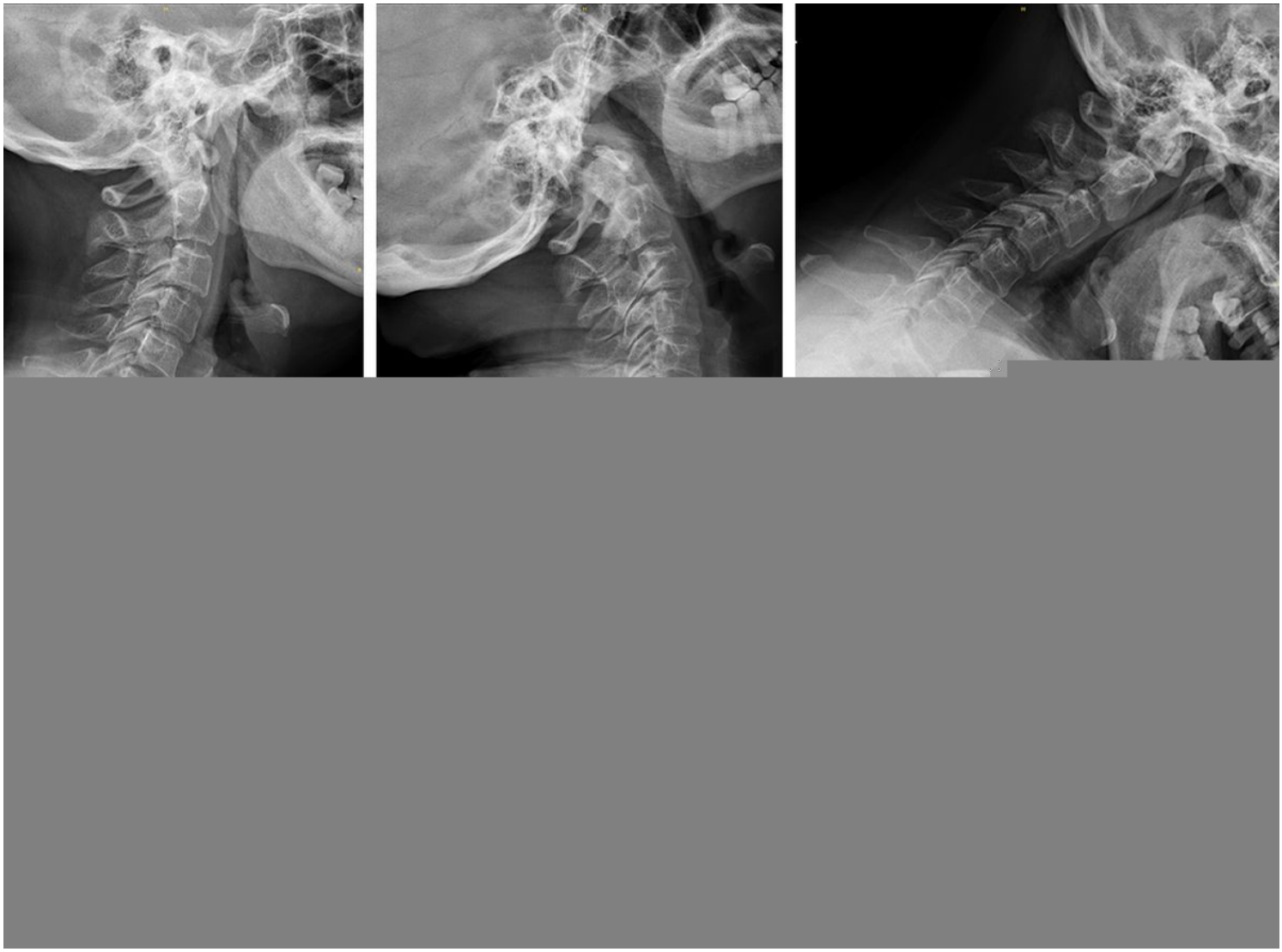

Figure 7

Preoperative imaging results of case 2: A, B, and C: Dynamic lateral cervical radiographs displaying no signs of instability. D, and E: Cervical magnetic resonance imaging (MRI) and computed tomography (CT) exhibiting no prominent structural lesions that could be the cause of occipital neuralgia. F: Cervical 3D CTA showing a typical VA course extending from C2 to C1 transverse foramen and wriggles across the vertebral artery groove on the superior surface of the posterior arch of C1 into the dura.

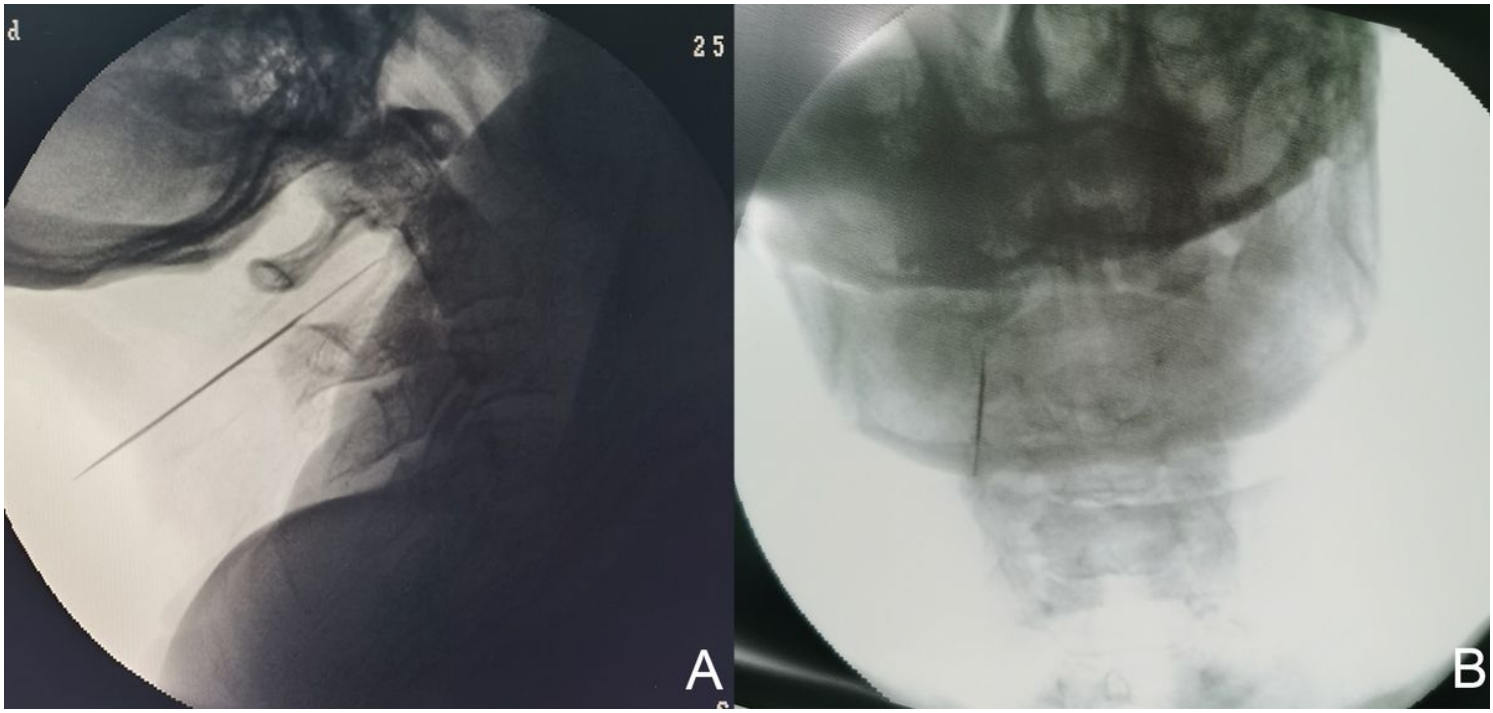

Figure 8

Illustrations showing the diagnostic C2 ganglion block prior to the surgery. A: lateral fluoroscopy. B: anteroposterior fluoroscopy.