CASE REPORT
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Occipital Neuralgia after Occipital Cervical Fusion to Treat an Unstable Jefferson Fracture

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In this report we describe a patient with an unstable Jefferson fracture who was treated by occipitocervical fusion and later reported sustained postoperative occipital neuralgia. A 70-year-old male was admitted to our center with a Jefferson fracture induced by a car accident. Preoperative lateral X-ray revealed an atlanto-dens interval of 4.8 mm and a C1 canal anterior-posterior diameter of 19.94 mm. We performed fusion surgery from the occiput to C5 without decompression of C1. The patient reported sustained continuous pain throughout the following year despite strong analgesics. The pain dermatome was located mainly in the great occipital nerve territory and posterior neck. Magnetic resonance images revealed no evidence of cord compression, however a C1 lamina compressed dural sac and C2 root compression could not be excluded. We performed bilateral C2 root decompression via a C1 laminectomy. After decompression, bilateral C2 root redundancy was identified by palpation. After decompression surgery, pain was reduced. This case indicates that occipital neuralgia, suggesting the need for diagnostic block, should be considered in the differential diagnosis of patients with sustained occipital headache after occipitocervical fusion surgery.

Key Words: Spinal Fracture · Cervical Atlas · Nerve root compression

INTRODUCTION

The pathogenesis of occipital neuralgia is unclear, with diagnosis dependent on diagnostic block. Although there have been several studies of the cause of occipital neuralgia, none has reported occipital neuralgia caused by direct compression of the C2 nerve root after treatment of a Jefferson fracture[4,9]. Here, we describe a patient with an unstable Jefferson fracture who was treated by occipitocervical fusion and later reported sustained postoperative occipital neuralgia.

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CASE REPORT

A 70-year-old male was admitted to our center with a Jefferson fracture caused by a car accident. The patient reported neck pain with an intensity of 7-8 on a visual analog scale ranging from 0-10, without any neurological deficit other than combined injury. A preoperative open mouth view showed a 7.26 mm lateral displacement of the C1 to the C2 lateral mass. Preoperative lateral X-ray also revealed an atlanto-dens interval of 4.39 mm and a C1 canal anterior-posterior diameter of 20.20 mm. Preoperative magnetic resonance imaging (MRI) and computed tomography showed fractures of the bilateral anterior arch and the left side posterior arch of C1, and injury to the transverse ligament. Although there was no cord compression or abnormal cord signal change, mild dural sac compression of the left lateral side was observed (Fig. 1).

We decided to perform fusion surgery from the occiput to C5 without decompression of C1. The C1 lamina was used as a fusion bed with an iliac bone block from the occiput and C2, and further bone fusion from the occiput to C5 was per

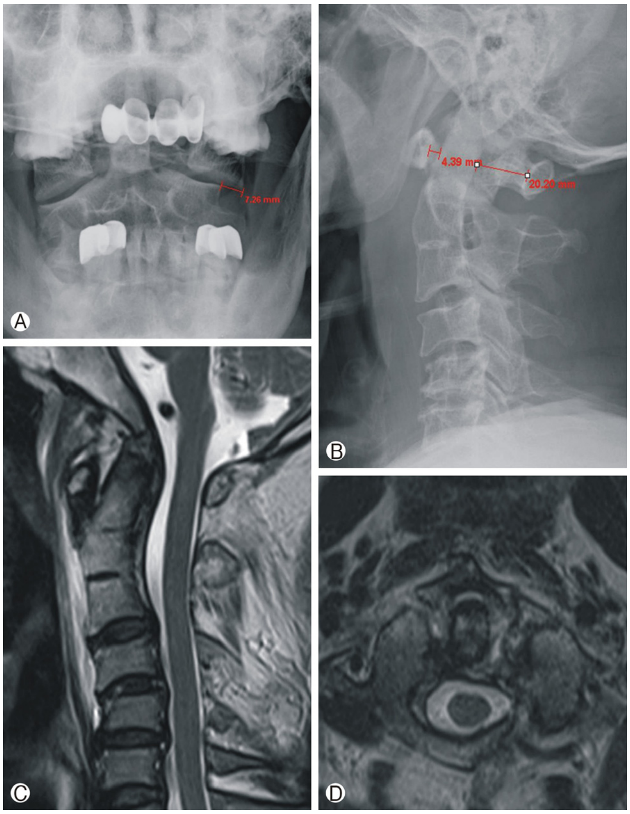

Fig. 1. (A) Preoperative open mouth view, showing a 7.26 mm displacement of the left C1 lateral mass to the C2 lateral mass. (B) Preoperative lateral X-ray showing a 4.39 mm atlanto-dens interval and a C1 canal diameter of 20.20 mm. (C) Preoperative T2-weighted midline magnetic resonance sagittal image showing an absence of cord compression. (D) Preoperative T2-weighted magnetic resonance axial image at C1-C2 level revealing a Jefferson fracture, injury to the transverse ligament, and dural compression of the left side fractured lamina.

formed using iliac bone chips. Postoperative lateral X-rays showed an atlanto-dens interval of 3.12 mm and a C1 canal anterior-posterior diameter of 15.13 mm (Fig. 2). There were no complications during or after surgery. The patient had normal neurological status, but reported neck pain and occipital headache, which we initially believed was part of the normal postoperative course; however, the patient reported sustained continuous pain for 1 year, despite strong analgesics. The pain dermatome was located mainly in the great occipital nerve territory and posterior neck (Fig. 3). Although MRI revealed no evidence of cord compression, a C1 lamina compressed dural sac and C2 root compression could not be excluded (Fig. 4). From the pain dermatome and C1 lamina-induced dural compression we suspected C2 compression. A diagnostic bilateral C2 root block resulted in mild pain improvement; therefore, we decided to decompress the canal and bilateral

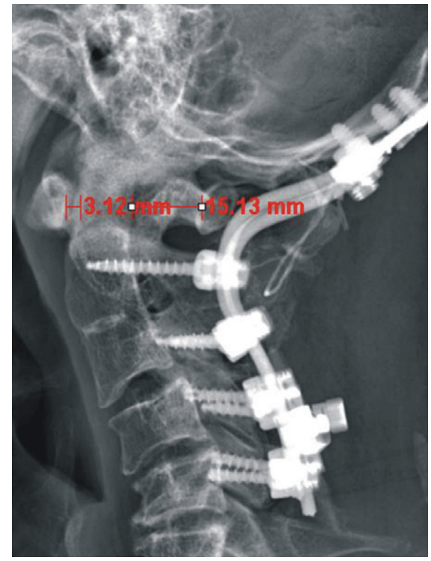

Fig. 2. Immediate postoperative lateral X-ray showing a 3.12 mm atlanto-dens interval and a C1 canal diameter of 15.13 mm. Iliac bone block with wiring between the occiput and C2 is also visible.

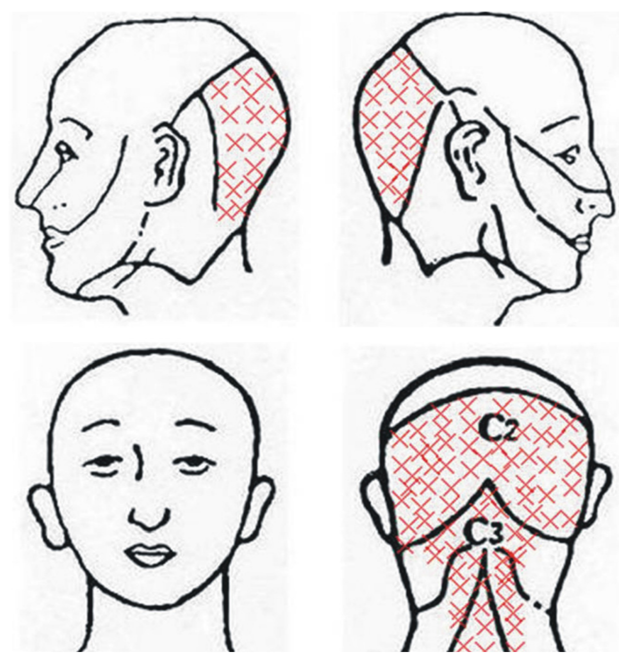

Fig. 3. Postoperative pain was located mainly in the C2 dermatome.

C2 roots surgically. During the operation, we found that the previous implantation of bone chips had resulted in firm bone stability and rigid bone fusion; however, we also observed a bone block resorption from the occiput to the C2 as well as replacement by thick fibrous tissue. Removal of the latter showed that the C1 posterior arch had severely compressed

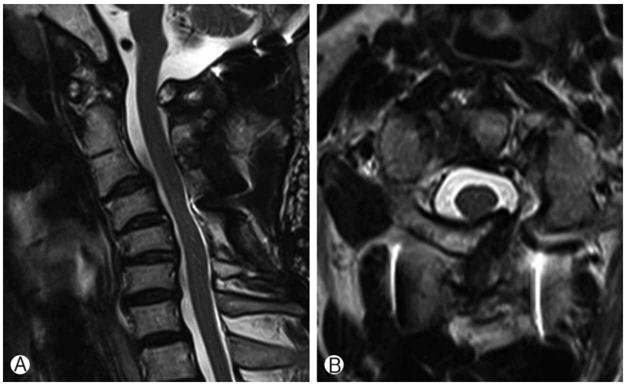

Fig. 4. (A) One year post-operative T2-weighted midline magnetic resonance sagittal image showing contact between the C1 posterior arch and cord, indicating narrowing of the canal compared to the preoperative magnetic resonance image. (B) One year post-operative T2-weighted magnetic resonance axial image at the C1-C2 level showing C1 laminar compression of the dura compared to the preoperative magnetic resonance image.

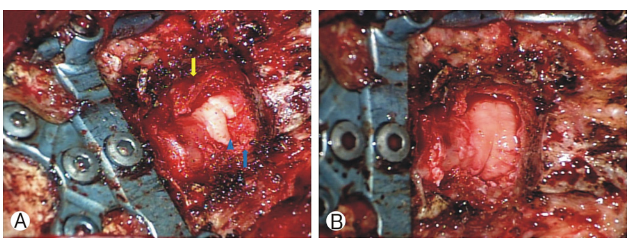

Fig. 5. (A) The C1 lamina (blue arrow) was partially removed. Dural compression of the C1 lamina was observed (blue arrowhead). The remaining C1 lamina also compressed the lateral side of the dura (yellow arrow). (B) Dura release after decompression. Bilateral root redundancy was identified by palpation.

the dural sac. We therefore performed bilateral C2 nerve root decompression followed by central canal decompression. After identifying bilateral C2 root redundancy by palpation, we completed the surgery (Fig. 5). One month later, the patient reported improvement of the occipital and neck pain (intensity of 1-2 on a visual analog scale).

DISCUSSION

Although occipitocervical fusion has been used to treat unstable Jefferson fractures, advances in surgical techniques have resulted in the preservation of normal cervical motion[6,11]. We therefore thought that our patient could be treated successfully by C1-C2 fusion or bilateral C1 fixation and fusion. Our findings, however, suggest that uninstrumented C1 lamina may result in an iatrogenic dural sac and bilateral C2 nerve compression.

Occipital neuralgia has many causes including C1 lateral mass screw irritation and C2 nerveroot retraction during C1-C2 fusion[1,2,4,5,7]. However, direct compression of the C2 nerve rootafter treatment of a Jefferson fracture has not previously been described. Occipital neuralgia in our patient may have been caused by direct dural compression or bilateral C2 root compression by the C1 posterior arch. The canal diameter decreased from 20.20 mm before surgery to 15.13 mm after the surgery, despite an improvement in the atlanto-dens interval. We attempted to compress the iliac bone block to the C1 posterior arch with a wire to achieve good fusion; however, this maneuver resulted in compression of the dura by the C1 posterior arch. Another possible cause of root compression was the rotation of the C1 posterior arch caused by the surgical position. If we had instrumented only C1 and C2, this might not have occurred because the cross link might have pulled each C1 lateral mass towards the center, resulting in a more posterior location of the C1 posterior arch. However, the surgical option we chose resulted in more anterior displacement of the C1 posterior arch caused by an iliac bone block. Iliac bone fusion mass resorption may be related to instability in the C1 posterior arch and an immobile structure without instrumentation.

Surgical indications for occipitocervical fusion include atlanto-occipital dissociation, occipital condyle fractures, and rheumatoid arthritis[3]. When C1 instrumentation is impossible, a post-instrumentation change in canal distance should be considered if the C1 posterior arch could be preserved.

The C2 nerve root is susceptible to injury or entrapment that can be treated by several surgical and diagnostic procedures[8,10]. Diagnostic block seems to be the most important tool for detecting the offending lesion.

In summary, this case showed pain in the typical dermatome of the C2 nerve root and we identified a possible cause as C2 nerve compression with diagnostic block. This case indicates that, after occipitocervical fusion, postoperative occipital neuralgia should be considered in possible differential diagnosis as a complication.

CONCLUSION

We have described a patient who, after occipitocervical fusion without C1 laminectomy, experienced occipital neuralgia associated with central and lateral compression of the C1 posterior arch. An intraoperative change in canal diameter may be a cause of such a complication should be considered. Occi- pital neuralgia, suggesting the need for diagnostic block, should be considered in the differential diagnosis of patients who complain of sustained occipital headache after this type of the surgery.

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