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CLINICAL CASE REPORT

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Post-Traumatic Torticollis Due to Odontoid Fracture in a Patient With Diffuse Idiopathic Skeletal Hyperostosis

A Case Report

Shotaro Tsuji, MD, Shinichi Inoue, MD, Toshiya Tachibana, MD, Keishi Maruo, MD, Fumihiro Arizumi, MD, and Shinichi Yoshiya, MD

Abstract: Descriptive case report.

To report a rare case of post-traumatic torticollis by odontoid fracture in a patient with diffuse idiopathic skeletal hyperostosis (DISH).

Cervical fractures in DISH can result from minor trauma, and a delay in presentation often prevents their timely diagnosis. Cervical fractures in patients with spinal DISH usually occur in extension injuries, and almost always occur in the lower cervical spine. Reports of odontoid fractures with torticollis in patients with spinal DISH are rare.

A 73-year-old man with DISH presented with severe neck pain and a cervical deformity presenting as torticollis without neurological deficits. He gave a history of a fall while riding a bicycle at a low speed 3 months ago. X-ray showed torticollis in the right side, and computed tomography (CT) showed a type-II odontoid fracture and subluxation at the C1-2 level.

We performed a staged treatment because this patient had severe neck pain associated with a chronic course. Initially, the fracture dislocation was reduced under general anesthesia and was stabilized with a halo vest. We then performed posterior occipitocervical in situ fusion after confirming the correction of the cervical deformity by CT. The patient showed significant amelioration of neck symptoms postoperatively, and bony fusion was achieved 1 year after surgery.

For post-traumatic torticollis due to an odontoid fracture, plain CT is useful for diagnosis and posterior occipitocervical in situ fusion following correction and immobilization with a halo vest is a safe and an effective treatment.

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Abbreviations: CT = computed tomography, DISH = diffuse idiopathic skeletal hyperostosis.

INTRODUCTION

In 1950, Forestier and Lagier[1] described a peculiar pattern of axial new bone formation characterized by flowing ossification along the anterior aspect of the vertebral column, which they termed ankylosing hyperostosis of the spine (also known as Forestier disease). In 1976, Resnik et al[2] described diffuse idiopathic skeletal hyperostosis (DISH) as a common disease that leads to ossification of ligaments and processes enthuses of the spine as well as peripheral skeleton. Cervical fractures in DISH can result from minor trauma and a delay in presentation often prevents the timely diagnosis of fractures. Fractures in DISH-affected spines usually occur in extension injuries, and almost always occur in the lower cervical spine.[3–8] Reports of odontoid fractures with in DISH are rare. No reports have described an odontoid fracture causing severe torticollis in a DISH patient, to our knowledge. We present a rare case of post-traumatic torticollis by odontoid fracture in a patient with DISH.

CONSENT

In our case, the patient signed related informed consent for the publication of clinical data and images.

CASE REPORT

The review board of our institute approved this study, and an appropriate written informed consent was obtained from the patient. The study was authorized by the local ethical committee and was performed in accordance with the Ethical standards of the 1964 Declaration of Helsinki as revised in 2000.

A 73-year-old man with DISH presented with severe neck pain and torticollis without neurological deficits. Three months ago, he sustained a fall while riding a bicycle at a low speed receiving a bruise on the left side of his head. Despite undergoing cervical spine radiographs immediately after the injury at the emergency room of the nearby general hospital, his cervical fracture was not diagnosed. He continued to have severe neck pain and torticollis for 3 months and subsequently consulted an orthopedic surgeon at a nearby hospital. A fracture of his cervical spine was suspected by radiographic examinations and he was referred to our department. The right torticollis with severe pain had increased, but his neurological examination was normal (Figure 1). His past medical history included diabetes mellitus and hypertension. Plain radiographs of the cervical spine showed torticollis on the right side, but no specific fracture of the cervical spine was identified (Figure 2). Cervical computed tomography (CT) revealed a type-II odontoid fracture, destruction of right superior articular surface of axis, and subluxation of left atlantoaxial joint (Figure 3). In addition, on cervical CT, we found flowing ossification not only on the ventral cervical spine from the thoracic spine but also between the margin of foramen magnum and the tip of dens. First, we planned to reduce the fracture/dislocation under general anesthesia, because of severe neck pain with motion. We performed the reduction of the fracture

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From the Department of Orthopaedic Surgery, Hyogo College of Medicine, Nishinomiya, Japan
Correspondence: Shinichi Inoue, Department of Orthopaedic Surgery, Hyogo College of Medicine, Nishinomiya, Japan, 1-1 Mukogawa-cho, Nishinomiya, Hyogo 663-8501, Japan (e-mail: inoshin@hyo-med.ac.jp).
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FIGURE 1. Preoperative photograph of the cervical spine shows severe torticollis.

dislocation carefully, followed by rigid external immobilization with a halo vest. His neck pain and deformity were ameliorated after the reduction without worsening myelopathy symptoms, respiratory disturbance, or dysphagia. CT following immobilization showed corrective repositioning of the C1-2 facets (Figure 4). After 1 week, surgery consisting of Occiput-C6 posterior in situ fusion using iliac bone graft was performed, followed by immobilization in a semi-rigid collar for 8 weeks (Figure 5). No complications occurred during the intraoperative or perioperative periods. Neck pain and torticollis resolved completely without any supportive treatment after surgery (Figure 6). At the 1-year follow-up, his symptoms had not recurred, and CT showed a union of odontoid fracture and bone bridging between the occiput and C2 (Figure 7).

DISCUSSION

Odontoid factures are rarely observed in patients with DISH. In this case, a lateral bending injury caused an atypical C2 facture with subluxation resulting in severe neck symptoms that lasted for 3 months because of an initial misdiagnosis. To our knowledge, no reports have described an odontoid fracture causing severe torticollis in a DISH patient. Some authors have reported that spinal fractures are most frequently cervical in DISH patients, and the most common mechanism of injury is low-energy trauma.[3–8] Caron[6] reported in 122 patients with ankylosing spinal disorders that the majority of fractures were located in the cervical spine, with C5-7 being most commonly affected. Bransford et al[7] and Westerveld et al[8] reported that the incidences of C1-2 fractures in patients with ankylosing spinal disorders are 7% and 13%, respectively. However, Meyer et al[9] and Bransford et al[7] reported no case with C1-2 fracture in their series of cervical spine trauma in DISH. Westerveld et al[5] reported that delayed diagnosis of fractures in with ankylosing spinal disorders often occur; in addition, they reported that all delayed diagnoses and in DISH patients were caused by physician misdiagnosis, possibly because of low awareness of this condition. In the present case, a rare C2 fracture was suspected 3 months after the initial trauma, and it manifested as neck pain without neurological deficits. Some reports recommended screening of the entire spinal column with advanced neuroimaging, such as CT, because of the well-documented difficulties in

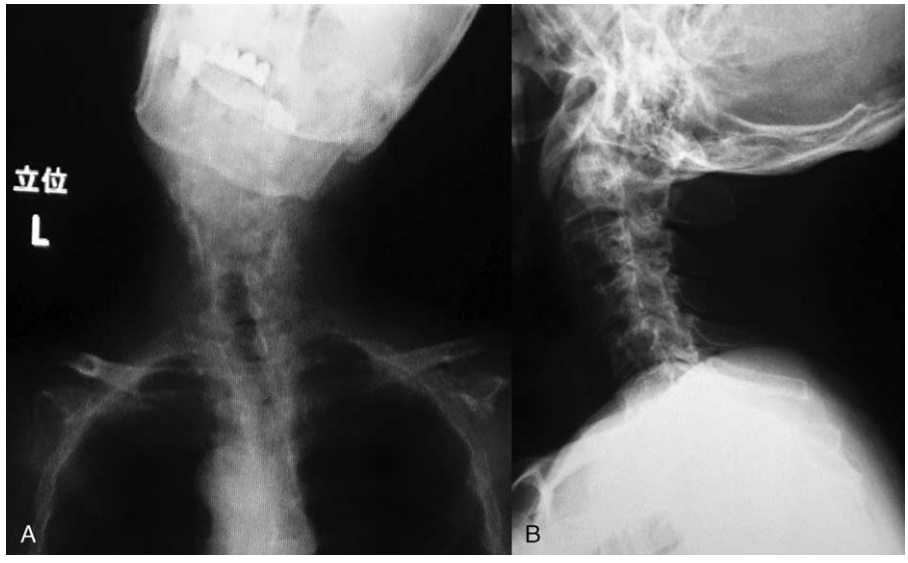

FIGURE 2. A: Preoperative anteroposterior radiograph of the cervical spine torticollis on the right side. B: Postoperative lateral radiograph of the cervical spine. No fractures of the cervical spine were identified.

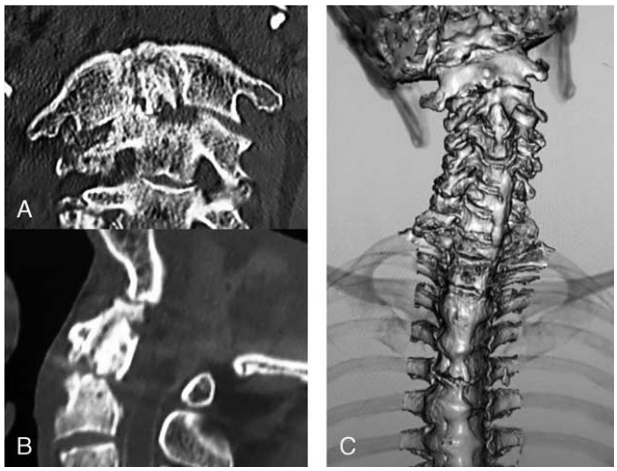

FIGURE 3. Preoperative computed tomography (CT) scan of the cervical spine. A: Coronal CT shows an odontoid fracture, destruction of right superior articular surface of axis, and subluxation of left atlantoaxial joint. B. Sagittal CT shows a type-II odontoid fracture, and continued ossification between the margin of foramen magnum, and the tip of dens. C: Three-dimensional CT shows flowing ossification ventral to the spine, extending to the cervical spine from the thoracic spine.

radiographic visualization of the spine in DISH. As a result in this case, CT diagnosed an odontoid fracture 3 months after his accident.

Treatment for a patient with DISH with post-traumatic torticollis by odontoid fracture is largely unknown. In general, C1-2 fusion by posterior approach is recommended for chronic odontoid fracture without ankylosing spinal disorders, but may have limited indications for patients with ankylosing spinal disorders. In 1991, Miller et al[10] reported 2 patients with ankylosing spondylitis with odontoid fractures who underwent atlantooccipital fusion. Paley[3] reported on 1 patient with DISH and late myelopathy due to odontoid fracture who underwent C1-2 fusion. However, it is unclear whether their cases had severe cervical deformity. In this case, it was necessary to confirm the reduction for cervical deformity by manipulation under general anesthesia successfully, because his odontoid fracture had been injured for over 3 months. If this patient would have had a rigid cervical deformity, it would have required a more invasive procedure, such as a reduction by the anterior transoral approach or an osteotomy by the posterior approach. Wang et al[11] reported on a case of a patient with a type-II odontoid fracture with irreducible dislocation that

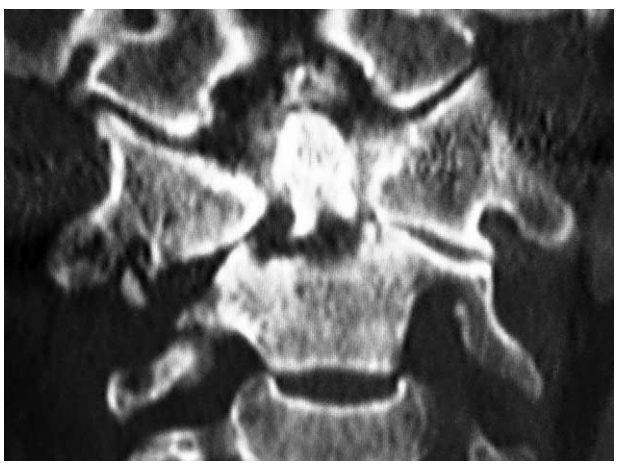

FIGURE 4. Coronal computed tomography scan after correction and stabilization with a halo vest under general anesthesia shows the reduction of C1-2 facets.

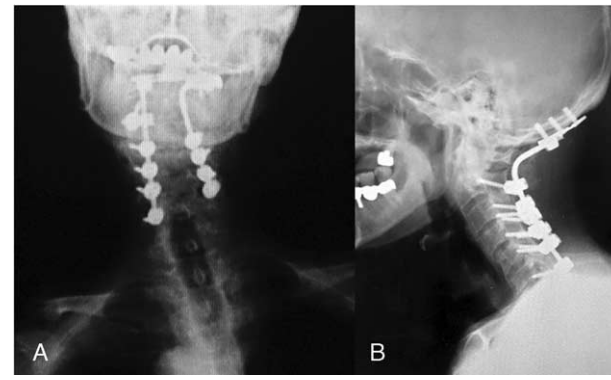

FIGURE 5. (A) Anteroposterior and (B) lateral postoperative radiographs after posterior occipitocervical in situ fusion.

FIGURE 6. Postoperative photograph shows an improvement of torticollis.

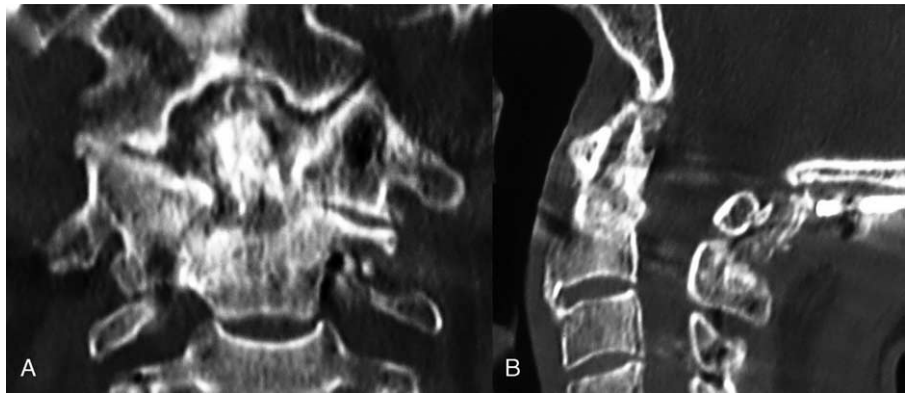

FIGURE 7. (A) Coronal and (B) sagittal postoperative computed tomography scans demonstrated a union of odontoid fracture and bone bridging between the occiput and C2.

was treated with intraoperative transoral open reduction with anterior–posterior fixation. However, this anterior transoral approach is challenging and operative complexity. Grundy and Gill[12] reported 1 case of osteotomy of the upper spine deformity with ankylosing spondylitis. Their technique may be beneficial when an anterior approach is technically difficult.

In the present case of a DISH patient with severe torticollis, posterior in situ occipitocervical fusion after reduction under general anesthesia was safe and effective. Despite Paley et al's[3] report of a DISH patient with odontoid fracture undergoing C1-2 fusion, we considered treatment for this case by C1-2 fusion insufficient because of several reasons. First, it may be difficult to have sufficient anchoring at C2 because preoperative CT demonstrated not only odontoid fracture but also destruction of right axis. Second, because our patient's cervical spine was fused, it acted as a long lever arm. Therefore, we believed that more rigid fixation approaches, such as occipitocervical fusion, was essential for maintaining reduction of severe torticollis. However, postoperative dysphagia after occipitocervical fusion must be considered. Izeki et al[13] described that reduction of anterior atlantoaxial subluxation during occipitocervical fusion may cause airway stenosis leading to postoperative dysphagia. In this case, our patient's severe neck pain with torticollis ameliorated considerably by occipitocervical fusion without any intraoperative, perioperative, or postoperative complications.

CONCLUSION

We present a rare case of post-traumatic torticollis by odontoid fracture in a patient with DISH. Our case demonstrated that plain CT was useful for diagnostic imaging, and posterior occipitocervical in situ fusion after the correction and immobilization with a halo vest under general anesthesia was a safe and an effective treatment.

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