Clinical Case Report

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Symptomatic posterior fossa and supratentorial subdural hygromas as a rare complication following transarticular screw fixation with posterior wiring for atlantoaxial instability

A case report

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Abstract

Rationale: Atlantoaxial transarticular screw fixation has been an effective and appealing method for inducing fusion of the C1-C2 complex. This technique is usually performed with Gallie fusion. In performing Gallie fusion using sublaminar wiring, a major concern is the risk of dural tear associated with passing sublaminar wires through the epidural space. We present the first report on symptomatic symptomatic subdural hygroma (SDH) due to transarticular screw fixation with posterior wiring.

Patients concerns: A 50-year-old man had sustained dens fracture 20 years ago and presented with severe neck pain following a recent traffic accident. The images showed atlantoaxial instability due to nonunion of the dens fracture and the patient underwent transarticular screw fixation with posterior sublaminar wiring using Gallie technique. When the U-shaped wire was passed under the arch of C1 from inferior to superior, a dural tear and cerebrospinal fluid (CSF) leak occurred. The site of dural tear was repaired by direct application of sutures. The patient was discharged in good condition. Fifteen day after surgery, the patient was readmitted with a history of a progressive headache associated with vomiting and vertigo.

Diagnosis: Brain CT and MRI showed bilateral posterior fossa and a right-sided supratentorial SDH.

Interventions: The patient underwent right occipital burr hole and evacuation of posterior fossa SDH due to deteriorating neurological status.

Outcomes: The patient's condition gradually improved after the operation and became asymptomatic at 3-year follow-up.

Lessons: Posterior fossa and supratentorial SDH could occur resulting from any intraoperative dural tear and CSF leakage during posterior cervical spinal surgery. Symptomatic SDH after posterior cervical spinal surgery should be cautiously assessed and treated.

Level of Evidence: 5

Abbreviations: CSF = cerebrospinal fluid, CT = computed tomography, MRI = magnetic resonance imaging, SDH = subdural hygroma.

Keywords: cerebrospinal leakage, posterior spinal surgery, subdural hygroma

1. Introduction

Atlantoaxial instability can result from trauma, congenital malformation, tumor, or inflammation and is best treated by reduction and fusion of the C1-C2 complex. Among the various operative techniques for the C1-C2 stabilization, atlantoaxial transarticular screw fixation have been provided excellent stability and contributed to a high fusion rate. This technique is usually performed with Gallie fusion if the posterior arch of the atlas is intact.[1–3] In performing Gallie fusion using sublaminar wiring, a major concern is the risk of dural tear or neurologic injury associated with passing sublaminar wires through the epidural space.

Symptomatic posterior fossa and supratentorial subdural hygroma (SDH) following transarticular screw fixation with sublaminar wiring for atlantoaxial instability has never been reported in the literature. The authors present the first case report of symptomatic SDH resulting from intraoperative dural tears during passage of sublaminar wire and discuss the management issues. The patient has provided informed consent for publication of the case.

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I-BH and UYC contributed equally to this work.

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2. Case report

A 50-year-old man was admitted with severe neck pain following pedestrian accident. Dynamic plain radiographs showed atlantoaxial instability due to nonunion of dens fracture (Fig. 1). The patient had been diagnosed with dens fracture after motor vehicle accident 20 years before, but the patient did not undergo surgery at that time. With his recent severe neck pain following traffic accident, the patient underwent transarticular screw fixation with sublaminar C1 wiring due to severe neck pain (Fig. 2). An attempt was made to avoid a dural tear when sublaminar wire was passed under the arch of C1 by advancing the smooth U-shaped loop first and by gentle epidural dissection prior to passing. But the dura was accidentally punctured, resulting in a cerebrospinal fluid (CSF) leakage. The leak was primarily repaired with 5–0 prolene sutures. The patient was asymptomatic in the postoperative period and was discharged 2 weeks after the operation. Two weeks following discharge, the patient visited an outpatient clinic complaining of headache, and the patient was advised to return to home for rest and observation. However, the patient was readmitted with progressive headache, nausea, and vomiting soon after.

On examination, he had bilateral papilledema, which had been absent at the initial admission. Computed tomography (CT) and magnetic resonance imaging (MRI) of the brain showed bilateral posterior fossa SDH over the cerebellar surface and right frontotemporoparietal SDH with dilated third and lateral ventricles (Fig. 3). Soon after admission, the patient experienced frequent episodes of severe headache and then became obtunded, presumably from brainstem compression from the posterior fossa hygroma. The patient underwent an emergent right suboccipital burr hole and evacuation of the posterior fossa SDH (Fig. 3). On opening the dura, clear fluid drained under high pressure. The patient's neurological condition improved and his headache gradually improved after the operation. On follow-up CT 3 month after the operation, the subdural CSF collections in the both the supra- and infra-tentorial compartments had resolved

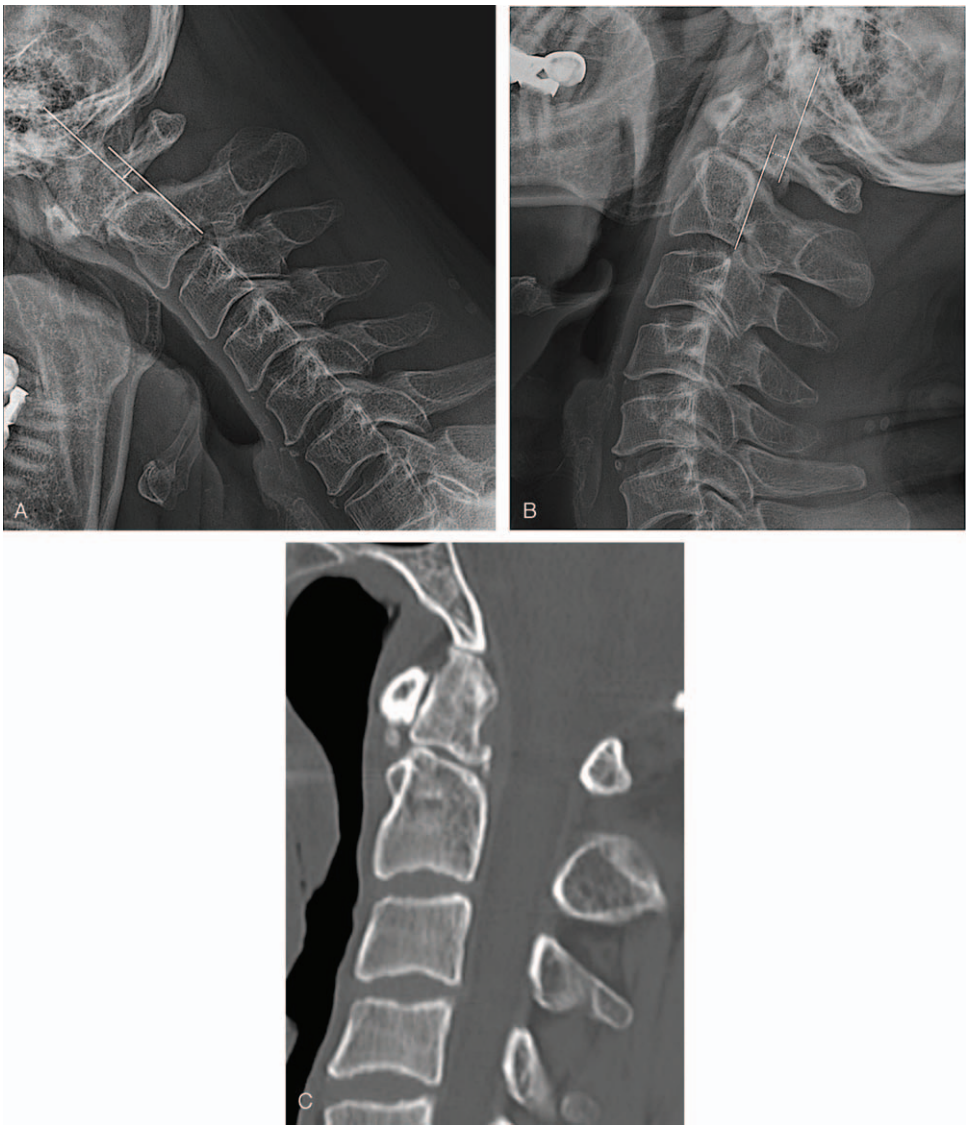

Figure 1. (A) Preoperative lateral flexion stress radiograph showing anterior displacement of nonunion fragment of odontoid process (white arrow) (B) Preoperative extension stress lateral radiograph showing posterior displacement of nonunion fragment of odontoid process (white dashed arrow) (C) Preoperative sagittal CT scan showing nonunion of odontoid process.

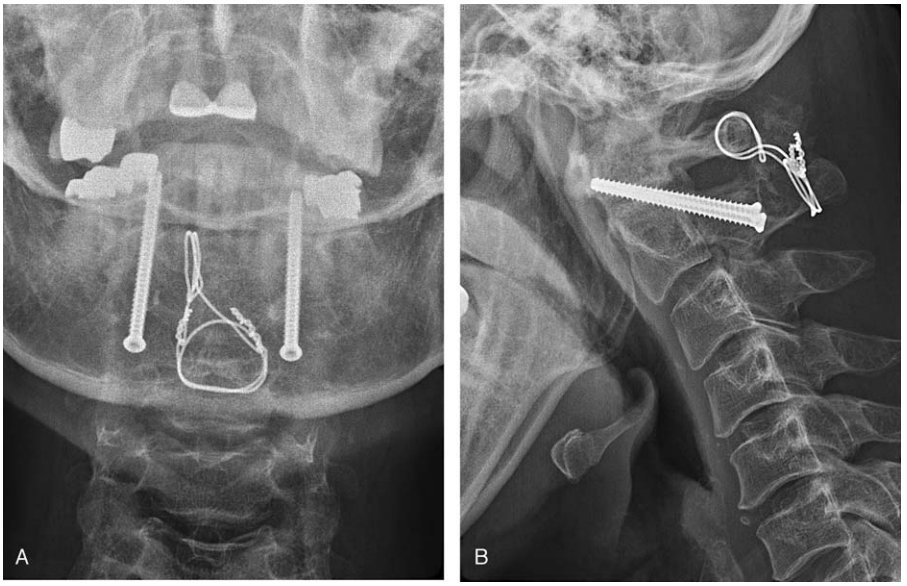

Figure 2. Postoperative anterior-posterior and lateral radiographs after posterior C1-C2 fusion.

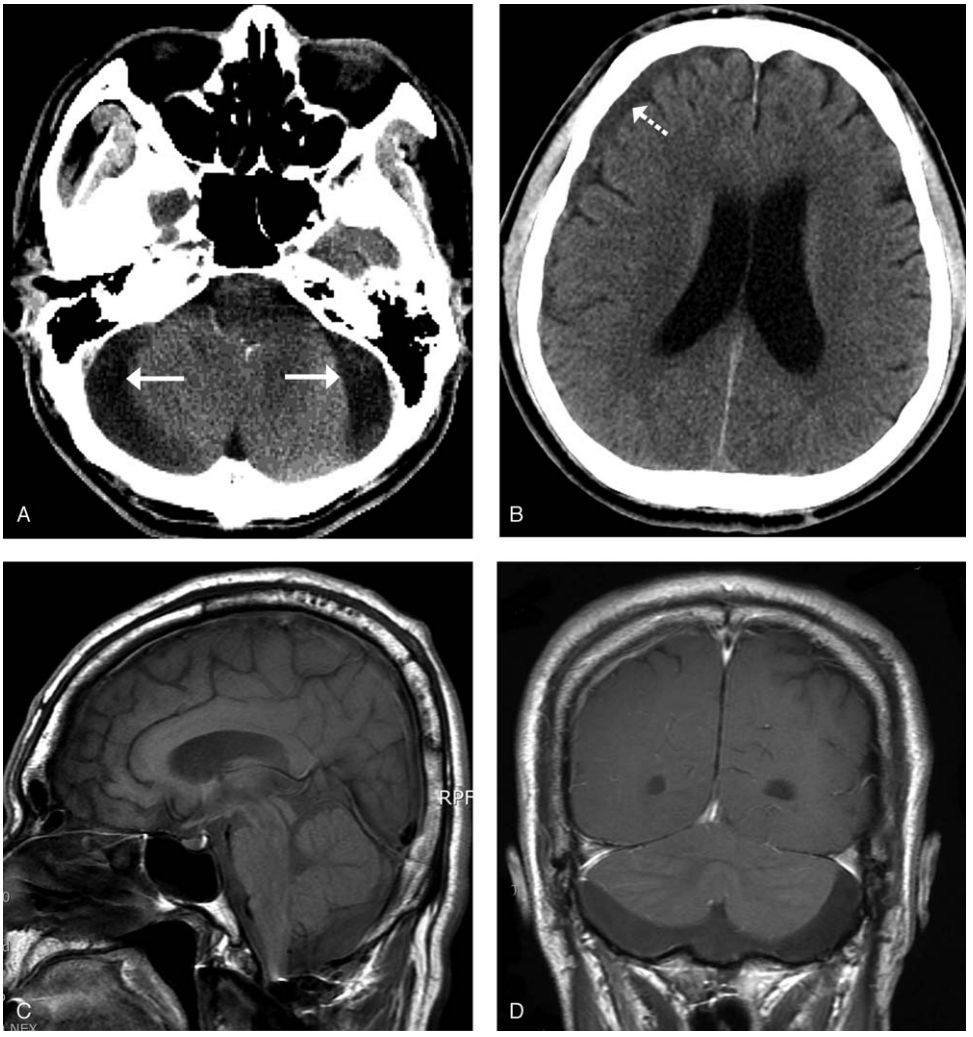

Figure 3. Computed tomographic and magnetic resonance images obtained 6 weeks after posterior C1-C2 fusion. (A) Axial CT scan disclosing fluid collection around cerebellum (white arrows). (B) Axial CT scan demonstrating left subdural fluid collection (white dashed arrow). (C,D) Saggital and coronal MRI images showing upward migration of cerebellum and abnormal fluid collection in the posterior cranial fossa.

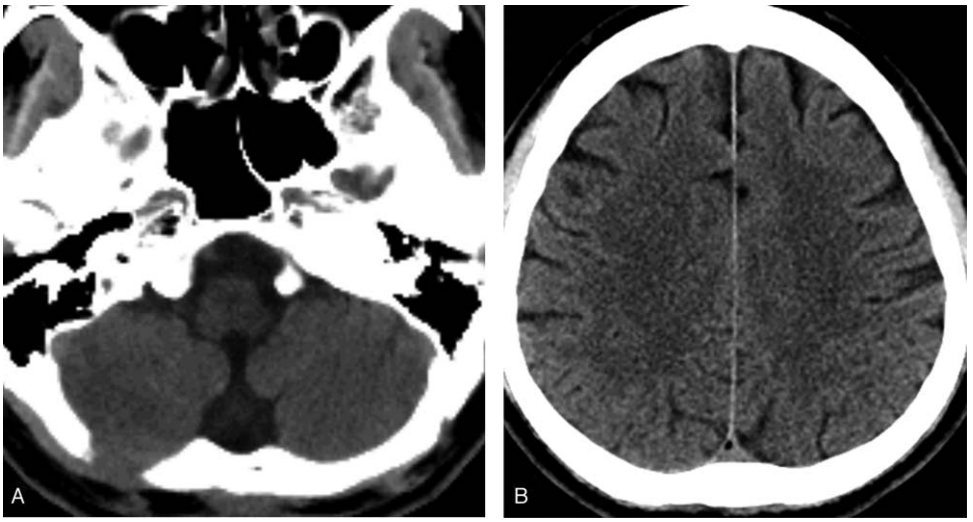

Figure 4. Computed tomographic images on the 3 month after the burr hole. Complete resolution of the abnormal fluid collection in infra (A) and supra (B) tentorial region.

completely (Fig. 4). The patient was asymptomatic at the 3-year follow-up.

3. Discussion

SDH, defined as an acute or chronic accumulation of CSF in the subdural space, has been reported to occur following foramen magnum decompression for the treatment of Chiari malformation-I.[4] Rarely, a few cases have been reported as a complication after posterior fossa tumor surgery.[5,6] However, symptomatic posterior fossa and supratentorial SDH following posterior cervical spine surgery has never been reported in the literature. Although the exact pathogenesis for the development of symptomatic posterior fossa and supratentorial SDH following foramen magnum decompression for the treatment of Chiari malformation-I remains uncertain, the probable cause could be a small defect in the arachnoid membrane, created during Chiari decompression surgery. The arachnoid defect creates a one-way slit valve-like mechanism through which CSF exits from the subarachnoid space to the subdural space without flow in the opposite direction. This subdural CSF can cause separation of the arachnoid and dura due to increased pressure in the subdural space. The CSF initially reaches the subdural space in the convexity, petrosal surface, and superior surface of the cerebellum. CSF then tracks to the supratentorial subdural space through the tentorial hiatus.[4,7] The subdural CSF collection can get absorbed slowly or cause rapid neurological deterioration, leading to coma due to compression of the brainstem and posterior fossa structures.

In the reported case, incidental durotomy occurred in the process of sublaminar wiring of C1. The durotomy was repaired by watertight suture, but a subdural hygroma developed in posterior cranial fossa and right supra tentorial region, postoperatively. Considering the pathomechanism and operation process, 2 injury mechanisms to the arachnoid membrane can be suggested. One possibility is that the arachnoid membrane defect remained open after suturing the overlying dura. An alternative explanation is that the arachnoid injury occurred by directed puncture of the suture needle during repair. Regardless of the exact cause of the arachnoid injury, it is plausible that the defect of arachnoid formed one-way slit valve. Additionally, CSF might have separated dura from arachnoid, enabling the formation of SDH tracking superiorly into the supratentorial region through tentorial hiatus. This SDH can develop non-communicating hydrocephalus causing pressure inward against cerebellar hemispheres, fourth ventricle and aqueduct of Sylvius.[8,9] This led to the classic neurological deterioration seen with other compressive pathology in the posterior fossa such as mass effect from tumors, ischemic, and hemorrhagic strokes and contusions.

If the patient did not have a large SDH with mass effect, and neurological deterioration, firstly, conservative management is recommended; with close monitoring in the hospital and repeat imaging. However, in our case, surgery was inevitable because the patient had progressive headache for about 1 month and neurologic deterioration due to increased intracranial pressure. Various surgical options have been reported in the literature for management of this. Suggested options include burr hole drainage, external ventricular drainage, ventriculo-peritoneal shunt, and re-exploration of the initial operation site. In deciding the treatment, co-existence of hydrocephalus should be considered. Kawaguchi et al described that the choice of treatment in the subdural hygroma depended on whether accompanying hydrocephalus existed.[10] If the patient had an obstructive hydrocephalus, external ventricular drainage or ventriculo-peritoneal shunt may be good options despite further risks of ventriculitis, epilepsy or shunt problems such as blockage or infection. However, obstructive hydrocephalus was not severe in our patient considering imaging studies. Bahl et al postulated that re-operation consisting of a wide arachnoidectomy is superior to CSF divergent procedures in subdural hygroma following decompression of the foramen magnum because only the re-operation can eliminate the causative defect; namely the arachnoidal slit-valve.[11] However, our surgery was not a simple decompression but a fusion with bone graft and wiring. It was not only more invasive to directly explore, but also technically difficult due to necessity of partial removal of the C1 arch for a wide arachnoidectomy and the disruption of the fusion construct and risk of need for a future occipital-cervical

fusion. Therefore, in our patient, burr hole drainage was a simple and effective procedure for the treatment of symptomatic SDH.

Pereira et al described that if CSF leak is evident following intraoperative arachnoid injury, wider opening of the arachnoid membrane is recommended to avoid the complication of SDH after decompression of the foramen magnum.[12] Because there were no reports of SDH following posterior C1-C2 fusion, it is not clear whether intraoperative wider opening of the arachnoid membrane can be performed after sublaminar wiring, but may be considered as an option during the index procedure.

Aside from cranial decompression surgery, upper posterior cervical spine surgery can also lead a SDH associating with injury of the arachnoid membrane. Especially, like the present case, there is always a concern and potential risk of dural tear or spinal cord injury during the passage of sublaminar wire.[3,13] If an intraoperative iatrogenic dural injury and postoperative SDH occurs, it is important to recognize this and treat it properly both intraoperatively and postoperatively. In general, the appropriate management of symptomatic SDH remains controversial. Through careful intraoperative examination of the arachnoid membrane, wider opening of the arachnoid can be attempted if a small arachnoid injury is detected. However, if the SDH developed postoperatively, treatment should be determined according to the clinical course and imaging studies. Conservative treatment can be attempted with mild clinical symptoms. But if the patient deteriorates or is at risk of significant brainstem compression then surgery should be performed based on existence of hydrocephalus.

4. Conclusions

Posterior fossa and supratentorial SDH could occur resulting from any intraoperative dural tear and CSF leakage during posterior cervical spinal surgery. Symptomatic SDH after posterior cervical spinal surgery should be cautiously assessed and treated.

Author contributions

Investigation: In-Bo Han, Dong-Eun Shin, Dae-sung Choi, Un Young Choi.
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Writing – review & editing: Alexander E. Ropper, Taekeun Ahn.
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