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Case Report

Dominant Vertebral Artery Injury during Posterior Atlantoaxial Transarticular Screw Fixation in a Juvenile Rheumatoid Arthritis Patient with Atlantoaxial Subluxation

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Many authors have reported on iatrogenic vertebral artery (VA) injury, but, to our knowledge, this is the first report of a dominant VA injury with compensatory blood flow from the hypoplastic VA. A 23–year-old woman with juvenile rheumatoid arthritis and atlantoaxial subluxation sustained injury to her dominant VA after occipitocervical fusion using transarticular screws. This did not result in lethal consequences due to compensation from her hypoplastic contralateral VA. Postoperative angiography, however, illustrated occlusion of the dominant left side, while the hypoplastic VA of the right side was enlarged. The patient experienced vertigo and loss of consciousness several times during rehabilitation. At the 4-year follow-up exam, bony fusion was observed, with no neurological deficits or correction loss. She had had no episodes of unconsciousness and no recurrence of any symptoms over the previous 3 years.

Key words: atlantoaxial subluxation, vertebral artery injury, transarticular screw, rheumatoid arthritis

Rheumatoid arthritis (RA) is a chronic, progressive, systemic autoimmune inflammatory disease affecting synovial joints. The cervical spine comprises multiple synovial structures, including apophyseal, uncovertebral, and atlanto-odontoid joints in a relatively small area, which makes it vulnerable to rheumatoid inflammation [1, 2]. Rheumatoid inflammation can lead to cervical spine deformities such as atlantoaxial instability because of softening of the bone or destruction of the facet joint [3]. The most common cervical abnormality associated with RA is atlantoaxial subluxation (AAS) [4, 5]. Furthermore, degenerative malalignment of vertebrae could compress the traversing vertebral arteries, leading to anomalous vertebral artery (VA) and cerebral ischemia [6].

In recent years, posterior atlantoaxial transarticular screw fixation, first described by Magerl and Seemann [7], has been widely used because of its biomechanical strength, minimal postoperative correction loss, and high fusion rates for treating AAS [8, 9]. The great potential risk of this procedure, however, is VA injury that may lead to massive hemorrhage, brain stem infarction, and even death [8, 10, 11, 12–14]. If there is unilateral VA hypoplasia or occlusion, injury to the dominant VA will cause lethal complications as a result of insufficient blood flow to the brain stem. The optimal technique must be selected carefully by considering all factors in the cervical spine and balancing safety with biomechanical strength, while being fully aware of the patient's unique condition.

We present the case of a woman who has bony

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structural changes and a vascular anomaly due to juvenile rheumatoid arthritis (JRA); her right VA is hypoplastic and the contralateral, dominant VA was accidentally injured. Fortunately, in this case, the neck pain of the patient was relieved, and her hypoplastic, right VA widened, allowing it to compensate with sufficient blood flow. Many authors have reported on VA injury [3, 6, 8, 10–13], but, to our knowledge, this is the first report of a dominant VA injury with compensatory blood flow from the hypoplastic VA. This report also stresses the value of carefully selecting the optimal technique for these types of patients.

Case Report

A 23–year-old woman with JRA visited our hospital in 2005 because of severe neck pain. Radiographs showed AAS. According to Ranawat's criteria, she had grade 3 rheumatoid arthritis pain. She had mild cervical myelopathy with numbness of all four limbs. In her activities of daily living, she was dependent upon wheel chair activity. Although she could stand, she could not walk because of arthritis in all four limbs. Radiographs showed severe instability at the atlantoaxial joint. The atlantodental interval, measured using lateral dynamic radiographs, was 10 mm at flexion and 0 mm at extension (Fig. 1). Preoperative CT myelography showed left side high-riding VA (Fig. 2), and preoperative MR angiography demonstrated right side VA hypoplasia (Fig. 3). The patient underwent posterior occipito-axial fusion using atlantoaxial transarticular screws bilaterally, inserted with the help of an image intensifier. Bone grafting material came from the iliac crest. During screw insertion, massive bleeding was not observed.

Surgery relieved the patient's neck pain; however, she experienced vertigo and loss of consciousness several times during rehabilitation. We performed catheter angiography, which showed occlusion of the dominant VA, due to screw misplacement (Figs. 4, 5, and 6). Fortunately, the hypoplastic VA on her right side increased in size and blood carrying capacity. She was treated post-operatively with oral anticoagulant therapy.

At the 4-year post-surgical follow-up exam, bony fusion was observed (Fig. 7), with no residual neurological deficits or correction loss. She had not experienced any vertigo or loss of consciousness, and there was no recurrence of any symptoms during the previous 3 years.

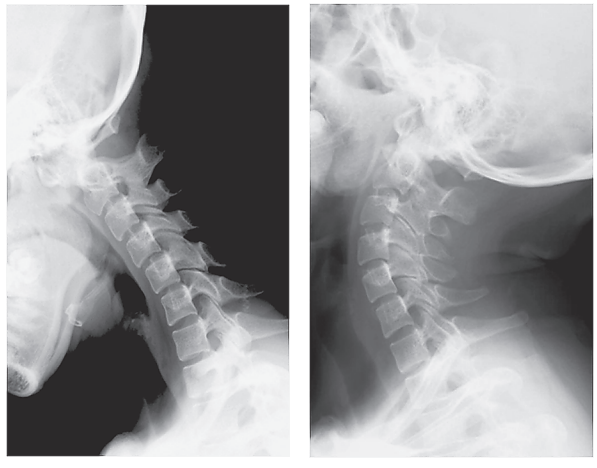

Fig. 1 Preoperative lateral radiographs. Lateral, dynamic radiographs reveal a reducible atlantoaxial subluxation. The atlantodental interval was 10 mm at flexion (A) and 0 mm at extension (B).

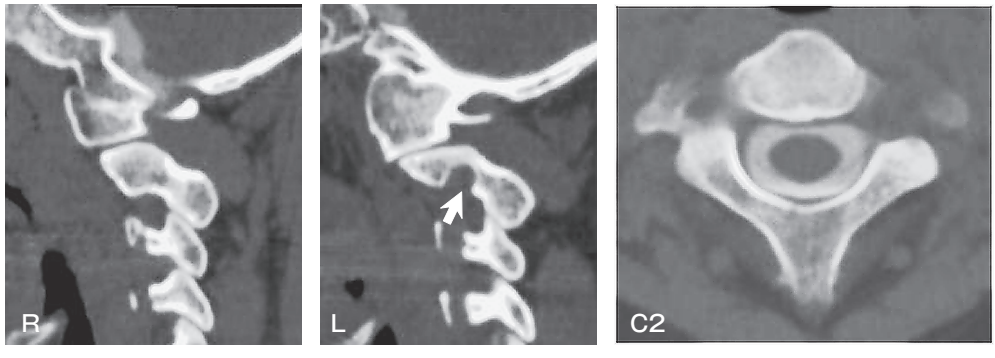

Fig. 2 Preoperative CT myelography. Preoperative CT myelography showed a left side high-riding vertebral artery (white arrow).

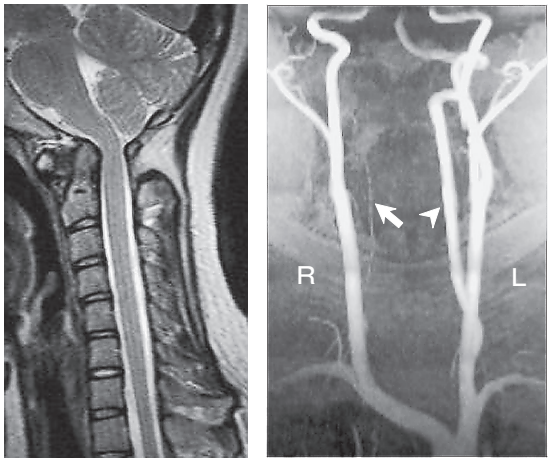

Fig. 3 Preoperative MR angiography. The left side vertebral artery (white arrowhead) is dominant, and the right side vertebral artery (white arrow) is hypoplastic but still communicating with the basilar artery.

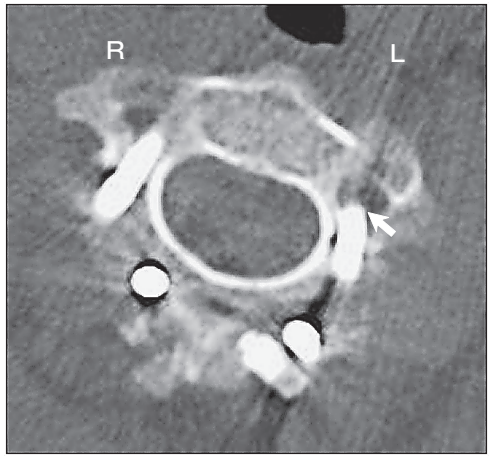

Fig. 4 Postoperative CT. The white arrow shows screw misplacement on the left side.

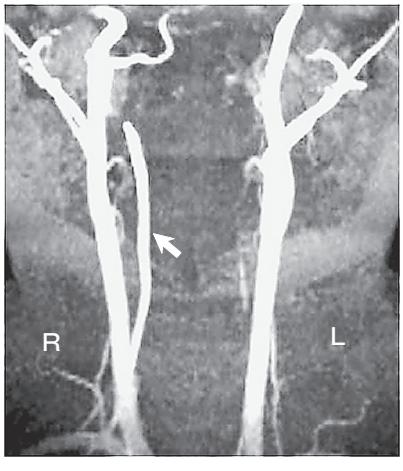

Fig. 5 Postoperative MR angiography. MR angiography shows occlusion of the left side vertebral artery, while the right side vertebral artery (white arrow) is enlarged.

Fig. 6 Postoperative catheter angiography. Catheter angiography shows an increase in size of the hypoplastic vertebral artery and occlusion of the dominant vertebral artery.

Discussion

In RA patients, the cervical spine is sometimes affected, and AAS is the most common type of instability experienced by these patients [1, 4, 5]. Surgical fixation for instability of the upper cervical spine is performed using transarticular or pedicle screws because of their biomechanical stability. However, the use of these screws carries a high risk of neurovascular complications [8, 10–14]. Furthermore, due to a high-riding VA [3, 13] or occlusion of the unilateral VA, surgery on rheumatoid cervical spines also entails considerable risks. Takahashi *et al.* [15] reported that the rate of unilateral VA occlusion was 23.8% in patients with RA. Simultaneously, due to erosion of the anatomical structures, bony landmarks are not reliable in RA patients. When the VA runs though abnormal anatomical structures, screw insertion has the potential for vascular injury.

In the patient described here, the dominant VA was accidentally injured. Fortunately, her hypoplastic VA widened, allowing it to compensate with sufficient blood flow. The mechanism of this phenomenon is unknown,

Fig. 7 Radiograph at the final follow-up.

though we propose 2 theories to explain it. The first theory is that the hypoplastic VA widened as a result of collateral blood circulation, and the second theory is that the VA widened as a result of reduction of AAS. VA injury is a major complication of cervical reconstruction surgery using transarticular screws [8, 13, 20]. Two studies reported that the rates of VA injury and misplaced screws during use of the transarticular screwing technique were 0% to 8.2% [8, 10, 14, 18, 19] and 0% to 15% [14, 18, 19], respectively.

Neo *et al.* [13] recently surveyed more than 5600 operations and described the incidence of VA injury using posterior atlantoaxial transarticular screw fixation as 1.3%. Wright and Lauryssen [11] surveyed 2949 atlantoaxial transarticular screws in 1318 patients and reported that the VA injury rate was 2.4%, while that of suspected injuries was 1.7%. In addition, the VA and bone structure at the craniocervical junction are complicated by having some unexpected variations, as reported by Duan *et al.* [20]. The VA makes a sharp, lateral, arc-shaped turn going though the transverse foramen of C2 [20, 21], which limits the space available for insertion of the transarticular screw. Therefore, it is imperative to select the optimal surgical technique, balancing safety and biomechanical strength to avoid inadvertent VA injury.

As experience with these cases broadens, improving patient safety and biomechanical strength simultaneously is indeed of importance. Several authors have reported the usefulness of a navigation system for pedicle screw insertion [15, 22]; however, many reports have confirmed that 100% accuracy is not guaranteed in transarticular screw placement, even with all the advanced elements in place, such as proficient surgeons [11, 13, 14, 18, 19] and a computer-assisted navigation system [15, 22]. We believe that using laminar screws in an RA patient with unilateral occlusion or asymmetry for atlantoaxial fusion is a good and relatively safe choice. Several authors have reported on the usefulness of C2 laminar screws because of their safety for avoiding VA injury and for their biomechanical strength [17, 23, 24]. Patients who clearly have a dominant VA, as in our case study, are good candidates for the laminar screw technique. Although the incidence of VA injury is low, we also believe that it is inexcusable for a surgical procedure to claim a life, irrespective of how low the incidence of death during the procedure may be, and we remain committed to improving the safety of this procedure.

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