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Case Report

# A pediatric case of axial rotary fixation associated with severe head trauma requiring emergency craniotomy for hematoma removal

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ABSTRACT

*Background:* Atlantoaxial rotatory fixation (AARF) causes the atlantoaxial joint to be fixed in a rotated position, resulting in painful torticollis. We report a case of pediatric AARF associated with severe head trauma requiring emergency craniotomy and was treated with conservative treatment.

*Case presentation:* A 10-year-old boy was struck by a van while walking across the street. Upon admission to our trauma care center, his Glasgow Coma Scale score was 11 points (E3V3M5), pupils were 4 mm bilateral regular circles, and other vital signs were stable. Plain computed tomography (CT) revealed left acute epidural hematoma, traumatic subarachnoid hemorrhage, cerebral contusion, pneumoencephalopathy, and rightward deviation of the axial vertebra. We performed an emergency craniotomy due to an enlarged hematoma on a repeat head CT scan and decreased level of consciousness. Based on imaging studies, rightward deviation of the axial vertebra was diagnosed as AARF; however, since the patient was already on ventilatory management and no physical findings were obtained, conservative treatment with cervical collar fixation was started. His condition improved, and he was extubated on day 3, released from the cervical collar on day 10, discharged from the hospital on day 17, and followed-up until day 32.

*Conclusions:* AARF is often caused by minor trauma or inflammation in children; however, we experienced a case complicated by severe head trauma, which was treated conservatively and showed a good clinical progress. Since AARF treatment depends on the length of time from onset, early diagnosis, in trauma care, carefully assessing factors other than major trauma, will lead to improved prognosis.

## Introduction

Atlantoaxial rotatory fixation (AARF) causes the atlantoaxial joint to be fixed in a rotated position, resulting in painful torticollis. It is more common in children and is thought to be triggered by minor trauma or upper respiratory tract inflammation. There have been

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no reports of pediatric cases of AARF associated with severe head trauma. We encountered a case of AARF associated with severe head trauma requiring emergency craniotomy that was treated conservatively.

## Case presentation

A 10-year-old boy with no specific medical history or medications, including collagen or genetic diseases, was struck by a van traveling at 30 km/h while walking across the street. Upon emergency medical service contact, the patient was aroused via stimulation and showed signs of restlessness. Although movement of the extremities was observed, a subcutaneous hematoma was also observed from the left occiput to the temporal region of the head with impaired consciousness, and the patient was transported to our trauma treatment center.

Upon admission to our hospital, his Glasgow Coma Score (GCS) was 11 (E3V3M5), pupils were bilateral regular circles of 4 mm, light reflexes were bilateral rapid, and there was no gross tetraplegia. Blood pressure was 132/89 mm Hg, heart rate was 94 beats/min, respiratory rate was 19 breaths/min, $SpO_2$ was 100 % (room air), and body temperature was 36.2 °C. Respiratory sounds were normal and did not differ from side to side, no inspiratory sounds or subcutaneous emphysema were noted, and focused assessment with sonography for trauma was negative. No obvious active hemorrhage was observed on the body surface. Plain computed tomography (CT) revealed left acute epidural hematoma, traumatic subarachnoid hemorrhage, cerebral contusion, left sphenoid fracture, left temporal bone fracture, pneumoencephalopathy, left zygomatic arch fracture, and rightward deviation of the axial vertebra (Fig. 1). His injury severity score, revised trauma score, survival probability, and trauma and injury severity score were 17, 6.904, 0.9717, and 0.0283, respectively. Arterial blood gas and laboratory test results are shown in Table 1.

On the same day, a repeat CT scan of the head showed an enlarged hematoma, and the patient's level of consciousness deteriorated to GCS E2V2M5 (Fig. 2). The patient was diagnosed with atlantoaxial rotatory fixation (AARF) with Fielding type I for deviation of the axial vertebra, and the atlantodental interval (ADI) was <3 mm.[1] However, no physical findings were obtained, and the patient was treated conservatively with cervical collar immobilization until his level of consciousness improved. After craniotomy to remove the hematoma, the patient's condition stabilized, and he was extubated on the third day (Fig. 3). Thereafter, the hematoma did not increase and the patient's level of consciousness gradually improved. On the 10th day, there was no cervical pain or torticollis and no problem in cervical movement, thus, we removed the cervical collar fixation and follow-up observation was initiated. On the 17th day, his general condition stabilized, and he was discharged from the hospital. No further symptoms or abnormalities on imaging tests were found, and we decided to close the medical examination on the 15th day after his discharge.

## Discussion

We present a case of a pediatric patient diagnosed with AARF complicated by severe head trauma requiring emergency craniotomy and was treated conservatively. In children, AARF may develop with minor trauma or inflammation due to less bony support of the atlantoaxial joint and a looser joint capsule than that in adults. This is the first report of AARF with severe head trauma. Moreover, head and upper cervical spine injuries are often associated with each other, and the definitive diagnosis of AARF may be delayed because there is little or no paralysis at the time of injury or the findings are inconclusive.

The characteristic symptoms of AARF include limited cervical rotation, pain on movement, and torticollis. The blood flow between the veins and lymphatic vessels leads to the venous plexus around the pharynx and dens of the axis, and inflammation of these tissues leads to fragility of the articular capsule and transverse ligament, resulting in AARF.[2] The atlantoaxial joint is responsible for approximately 50 % of the rotational motion of the cervical spine, and children have smaller supra-annular articular processes and

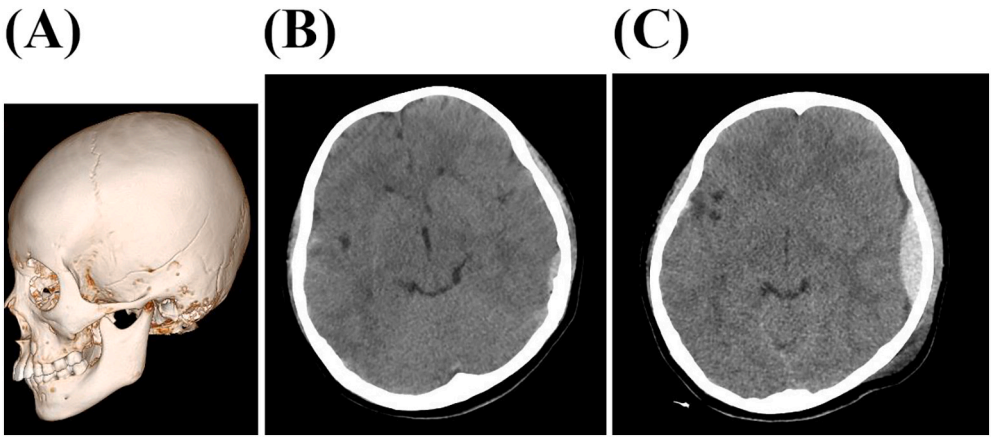


**Fig. 1.** Plain head CT images on hospital arrival and admission.
(A) 3D reconstruction of the skull revealing left temporal bone fracture. (B) Head CT on hospital arrival showing subarachnoid hemorrhage and left acute epidural hematoma. (C) Repeated head CT on admission showing an enlarged left acute epidural hematoma.

**Table 1**
Laboratory findings at the time of admission.

| | | |
|---|---|---|
| *Biochemistry* | | |
| Total protein | 7.5 | g/dL |
| Albumin | 5.0 | g/dL |
| Creatinine kinase | 222 | IU/L |
| AST | 31 | IU/L |
| ALT | 19 | IU/L |
| LDH | 366 | IU/L |
| ALP | 365 | IU/L |
| γ-GTP | 11 | IU/L |
| Amylase | 132 | IU/L |
| Total bilirubin | 0.7 | mg/dL |
| Direct bilirubin | <0.1 | mg/dL |
| Creatinine | 0.37 | mg/dL |
| BUN | 12.1 | mg/dL |
| Sodium | 136 | mEq/L |
| Potassium | 3.1 | mEq/L |
| Chloride | 102 | mEq/L |
| Magnesium | 2.1 | mg/dL |
| Calcium | 9.2 | mg/dL |
| Glucose | 122 | mg/dL |
| HbA1c | 5.0 | % |
| CRP | 0.03 | mg/dL |
| *Complete blood count* | | |
| White blood cells | 14,810 | /μL |
| Red blood cells | $5.78 \times 10^6$ | /μL |
| Hemoglobin | 15.5 | dL |
| Hematocrit | 44.6 | % |
| Platelet | $366 \times 10^3$ | μL |
| *Coagulation status* | | |
| APTT | 29.2 | s |
| PT-INR | 1.04 | |
| Fibrinogen | 193 | mg/dL |
| FDP | 57.0 | μg/dL |
| D-dimer | 17.5 | μg/dL |
| *Venous blood gas* | | |
| $F_IO_2$ | 0.21 | |
| pH | 7.386 | |
| $PaCO_2$ | 39.7 | mm Hg |
| $PaO_2$ | 39.1 | mm Hg |
| $HCO_3^-$ | 23.8 | mmol/L |
| Base excess | −1.1 | |
| Lactate | 35 | mg/dL |
| Anion gap | 14.2 | |

Abbreviations: AST; aspartate aminotransferase, ALT; alanine aminotransferase, LDH; lactic acid dehydrogenase, ALP; alkaline phosphatase, Γ-GTP; Γ-glutamyl transpeptidase, BUN; blood urea nitrogen, HbA1c; hemoglobin A1c, CRP; C-reactive protein, APTT; activated partial thromboplastin time, PT-INR; prothrombin time-international normalized ratio, FDP; fibrin degradation product, FiO2; fraction of inspiratory oxygen.

steeper tilts than adults. A meniscus-like synovial fold is present between the annular axis vertebrae, impeding repositioning.[3]

Accordingly, this disease is most common in early childhood to school age,[4] and is usually triggered by minor trauma; upper respiratory tract infection; or dental, oral, or otorhinolaryngological surgery,[5] while it can also occur without triggers. However, to the best of our knowledge, there have been no reports of cases triggered by severe head traumas. In severe head trauma, the initial treatment is often performed by emergency or neurosurgery physicians, resulting in a delayed or missed opportunity for orthopedic consultation. In the latter case, the fact that AARF itself is not recognized is assumed to be one of the reasons for this. In addition, it is possible that high-energy trauma, as in this case, could lead to axial process fracture rather than AARF.[6,7]

Imaging studies are useful for diagnosis, and cervical spine simple X-rays are used to examine the atlantoaxial joint; however, examination may be difficult due to pain, and CT is more useful for diagnosis. The treatment plan depends on the phase of the disease. Although there is no clear definition of acute and chronic phases, it has been reported that the success rate of conservative treatment decreases when diagnosis is delayed by more than one month.[8,9] In the acute phase with no neurological symptoms within 1 week of onset, conservative treatment, such as administration of anti-inflammatory analgesics, cervical collar fixation, and Glisson traction, is generally effective, and the prognosis is often good.

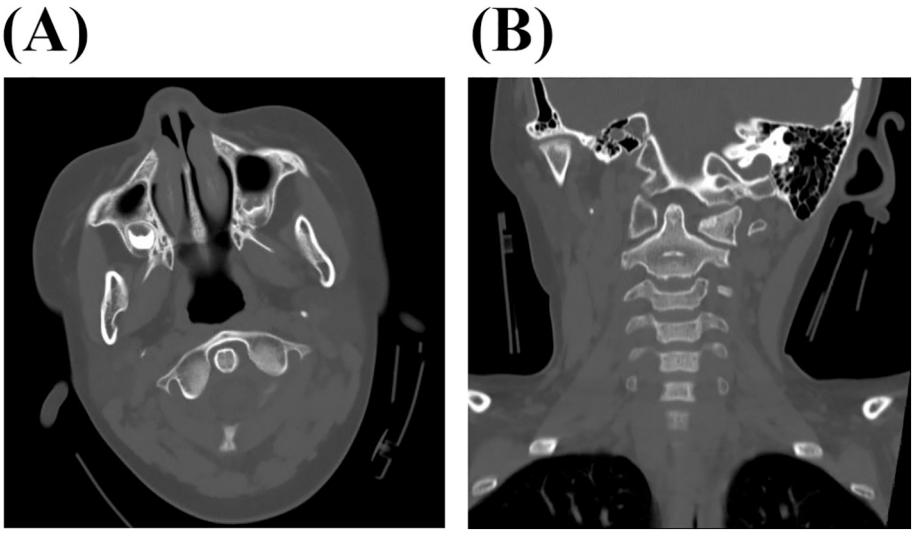


**Fig. 2.** Cervical spine CT images on hospital arrival.
CT images revealed rightward deviation of the axial vertebra; (A) axial sectional image and (B) coronal sectional image. The ADI was <3 mm; therefore, the diagnosis was AARF with Fielding Type I.
Abbreviations: ADI; atlanto-dental interval, AARF; atlantoaxial rotatory fixation.

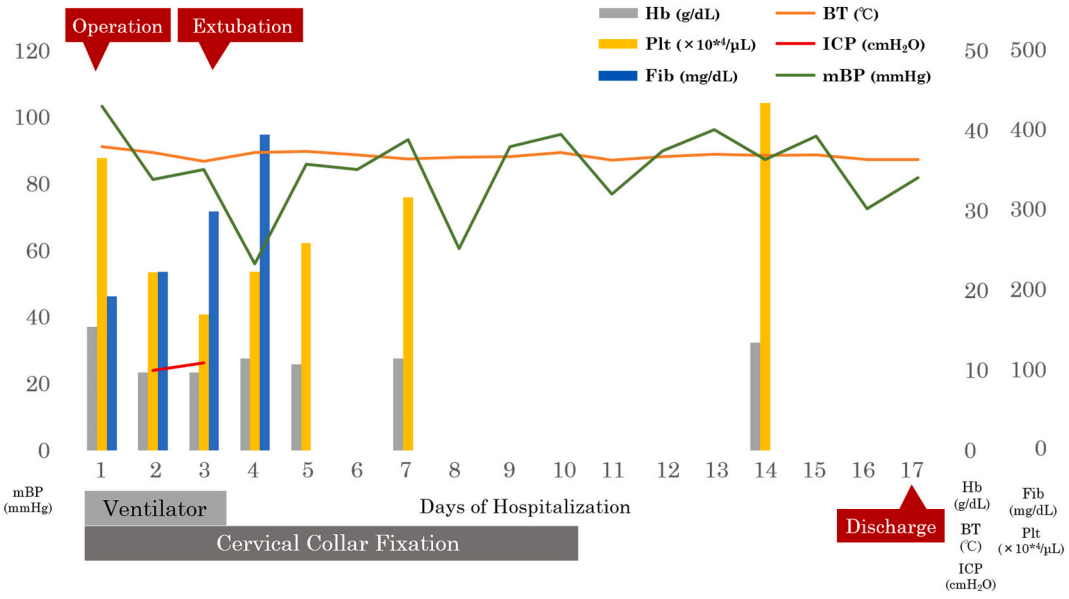


**Fig. 3.** Summary of the clinical course.
Abbreviations: Hb, hemoglobin; Plt, platelet count; Fib, fibrinogen; BT, body temperature; ICP, intracranial pressure; mBP, mean blood pressure.

However, since AARF can occur secondary to upper respiratory tract infection, patients do not always visit an orthopedic surgeon first, and the definitive diagnosis is consequently delayed. Ishii et al. advocated conservative treatment with noninvasive manual repair under general anesthesia followed by halo vest fixation as a strategy for the treatment of chronic AARF that avoids surgical treatment, with remodeling of the C2 facet deformity as the index of remodeling.[10,11] Although the number of chronic cases requiring surgery has been decreasing due to this remodeling therapy, some cases with difficulty in repair or recurrence still require surgical intervention.

As in this case, when a patient is unconscious and the injury site differs from the primary site, definitive diagnosis may be delayed, and the treatment plan may be more invasive or complicated. In this case, immediate total-body CT scanning was performed to identify the patient as an acute case, and conservative treatment was adopted with a good prognosis. AARF should be considered even in cases of severe trauma in pediatric patients with difficulty obtaining symptoms or physical findings.

## Conclusion

AARF is often caused by minor trauma or inflammation in children; however, we experienced a case complicated by severe head trauma, which was treated conservatively and showed good clinical progress. Since the treatment of AARF depends on the length of time from disease onset, the more time passes, the more refractory the disease becomes and the more invasive the treatment method,[12] careful observation and examination in addition to the primary focus in severe trauma care will allow for prompt treatment and improved prognosis.

## Ethics approval and consent to participate

In Japan, approval from an ethics committee is not required to report these cases. This case was reported in accordance with ethical guidelines for medical and health research involving humans established by the Japanese government.

## Consent for publication

Written informed consent was obtained from the patient's legal guardians for the publication of this case report and accompanying images. A copy of the consent form is available for review from the Editor-in-Chief.

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## CRediT authorship contribution statement

Treating of patient: T Minamiyama, RK, H Okada, YM, T Miyake, H Okamoto, NK, SN, and SO.
Study concept and design: TM, RK, and HO.
Drafting and critical revision of the manuscript: TM and RK.
Approval of the final manuscript: TM, RK, HO, YM, TM, NK, and SO.

## Declaration of competing interest

None.

## Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

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