



Surgery for nontraumatic atlantoaxial rotatory fixation after COVID-19 in a 17-year-old girl: a case report

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Abstract

We herein describe a case of atlantoaxial rotatory fixation (AARF) following COVID-19 in a healthy, 17-year-old, female, high school student. After the COVID-19 infection improved, the patient experienced neck pain. After 1 week, the patient was unable to turn her neck. Her local doctor diagnosed the patient with AARF. She wore a cervical soft collar and underwent Glisson traction for 2 weeks. However, as the AARF did not improve, computed tomography (CT) was performed. Reconstructed CT showed that the morphology of the C1/2 facet joint was asymmetrical. We manually repaired her neck position under general anesthesia, and she wore a halo fixation for a month. However, the patient neck position dislocated slightly when the traction was released. Hence, we performed C1/2 posterior fusion and bone graft. She was discharged home 10 days postoperatively and was able to perform her daily activities without any problems. One year later, CT images showed no bony bridge at the C1/2 facet joint, and the morphology of the C1/2 facet joint did not change from that before surgery. As no remodeling is expected after the end of the growth phase, early surgical treatment of AARF could be considered if conservative treatment is ineffective.

Keywords Atlantoaxial rotatory fixation · COVID-19 · Atlantoaxial joint fixation · Young-adult · Torticollis · Grisel syndrome

Introduction

AARF commonly occurs in school-age children and is often associated with colds or minor injuries. Treatment involves resting the affected area with a cervical collar. Most cases improve with this treatment. In some cases, additional therapy, such as traction, may be necessary. However, cases requiring surgery are rare. Here, we present the case of a 17-year-old female who developed AARF without an

apparent trigger. She required surgical intervention due to her resistance to conservative treatment.

Case presentation

A healthy, 17-year-old, female, high school student was diagnosed with COVID-19 in 2023. She rested at home for 7 days and had no subjective symptoms. Shortly thereafter, she felt a pain in her neck as she was studying. She thought it was a stiff shoulder and continued to study, but after 1 week, she could not rotate her neck; thus, she went to her local doctor, who diagnosed her with atlantoaxial rotatory fixation (AARF) by X-ray and computed tomography (CT). She wore a cervical soft collar for 2 weeks, but her symptoms did not improve; hence, she was referred to our hospital. At the initial visit, she was in a typical cock-robin position, looking to the left. There were no obvious neurological abnormalities. The right sternocleidomastoid muscle was tight and tender. The cervical spine had the following ranges of motion: flexion, 60°; extension, 50°; right rotation, 15°; left rotation, 45°; the rotational motion was limited.

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X-ray images obtained at the initial presentation showed a cervical spine imbalance (Fig. 1a), with a left–right difference in the dental process in the open position (Fig. 1b) and an open atlantodental interval (6.75 mm) (Fig. 1c). Therefore, we determined that the AARF was type 3, according to the Fielding classification [1]. A plain CT clearly showed abnormal rotational fixation (Fig. 1e, f). Thus, we initiated Glisson traction at 4 kg. We continued the traction for 2 weeks, and the right sternocleidomastoid muscle tone and tenderness improved slightly, but the cock-robin position remained unchanged, as did the range of motion of the cervical spine. Initial CT showed mild degeneration of the right C1/2 joint, which we considered a possible etiology (Fig. 1g, h).

As the patient did not improve after more than a month of conservative treatment from onset, we attempted a halo fixation under general anesthesia. Although manual manipulation under general anesthesia was easy, her neck position dislocated slightly when the traction was released. We confirmed by intraoperative CT imaging that the patient's neck was successfully repositioned and immobilized using halo fixation. She wore a halo fixation for a month, but her symptoms did not improve. We also considered the possibility that the cause of her symptoms was morphologic abnormalities of the intervertebral joints; hence, we decided that conservative treatment was not sufficient and that surgical fusion was necessary. During the subsequent surgery, the cervical alignment remained corrected; therefore, no further reduction maneuver was performed. Preoperative

contrast-enhanced CT showed a high-riding vertebral artery in the left C2 (Fig. 2a–f); thus, we decided to use navigation for fusion, with bilateral lateral mass screws for C1, bilateral cervical pedicle screws for C2, and an iliac bone graft on the left side.

Postoperative imaging showed good repositioning, and she was able to look forward and was discharged 10 days postoperatively (Fig. 2g). At 12 months postoperatively, the cervical spine had the following ranges of motion: flexion, 60°; extension, 50°; left rotation, 50°; right rotation, 50°, which was mildly limited but did not interfere with her daily activities. CT images at 7 and 12 months after surgery showed the morphology such as bony bridge of the C1–2 facet joint did not change from that before surgery (Fig. 2h, i). Although previous reports have described morphological remodeling of the atlantoaxial joint after successful reduction, such remodeling was not observed in the present case. This case suggests that satisfactory clinical recovery can be achieved even without radiographic remodeling or fusion, provided that stable reduction is maintained.

Discussion

AARFs are known to occur after a common cold or minor trauma [1, 2]. AARF associated with inflammatory diseases of the neck is called Grisel syndrome [3]. The treatment for AARF is usually local rest with a cervical collar, but additional treatment such as traction therapy is necessary in

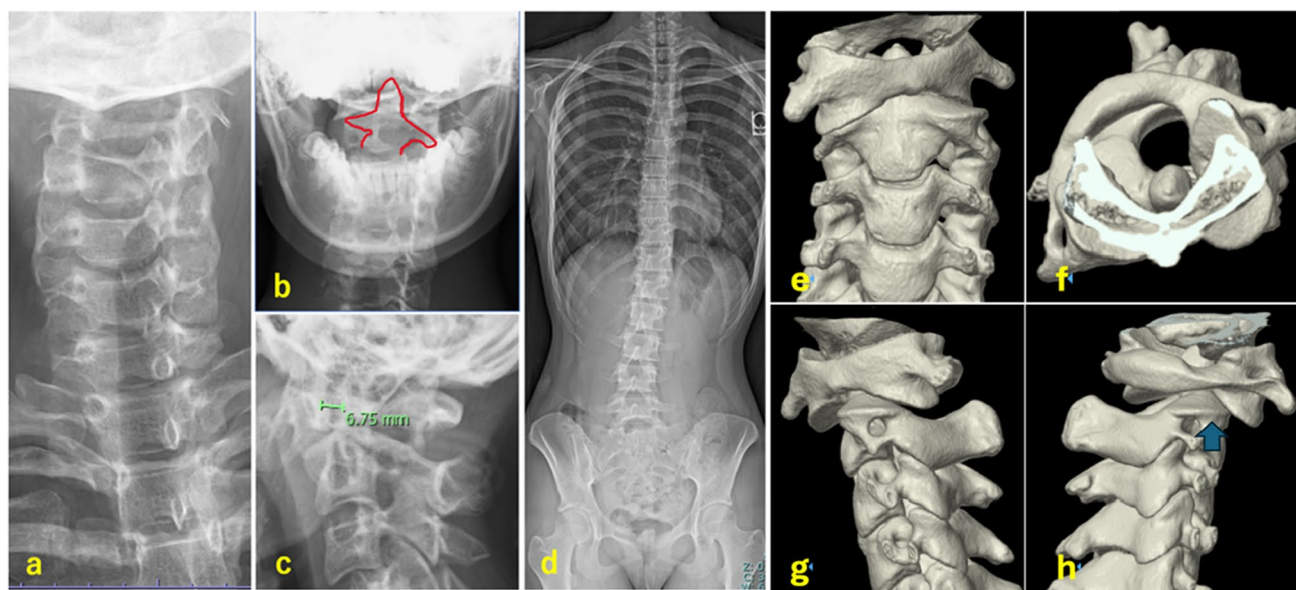
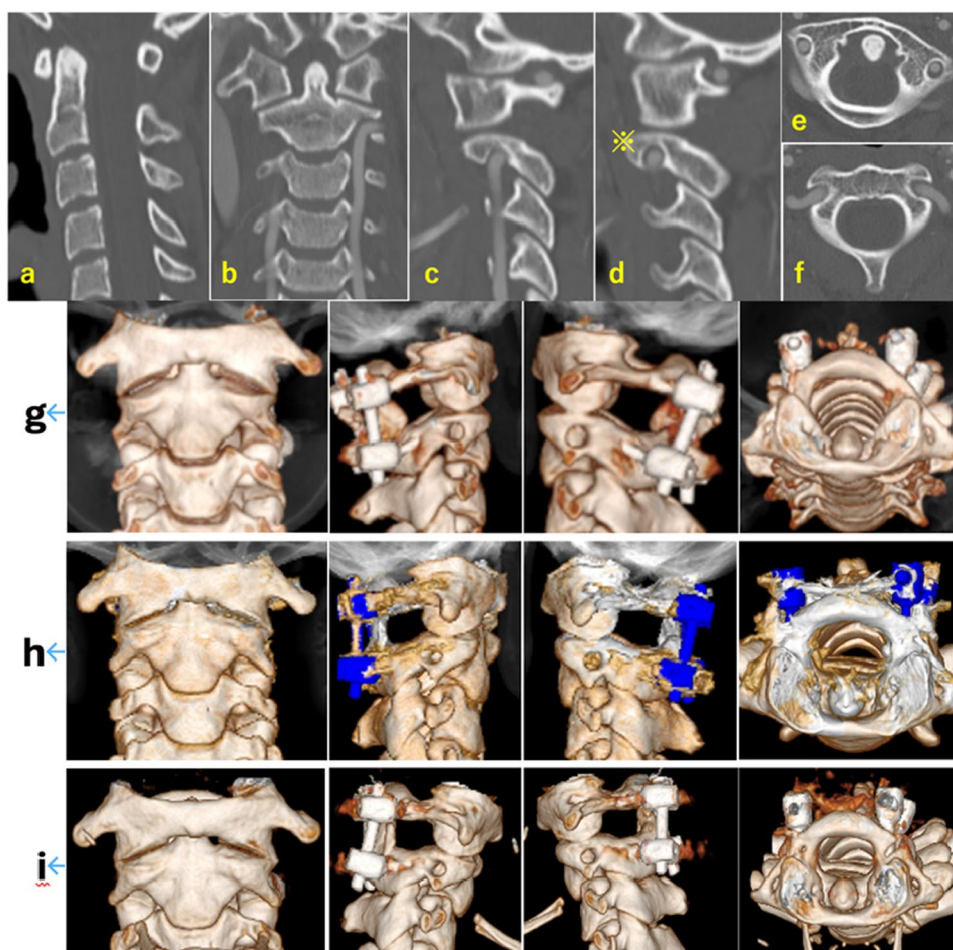


Fig. 1 a–d Preoperative plain radiographs. e–h Preoperative plain three-dimensional computed tomography. **a** Poor alignment of the spinous processes of the cervical spine. **b** Left–right difference in C2 in the open position. **c** C2 atlantodental interval=6.75 mm, indicating that it is open and large. **d** Full-spine X-ray of the patient showing

completed growth. **e** Frontal view showing that the right side of C1 has slipped anteriorly. **f** Cephalad view showing rotation at C1 and C2. **g** Left lateral view. **h** Right lateral view showing mild degeneration of the joint at C1/2 (blue arrow)

Fig. 2 **a–d** Contrast-enhanced computed tomography after halo vest, showing that the patient has been repositioned. ✕ 2.56 mm, indicating a high-riding vertebral artery. **e** and **f** The rotation of C1 and C2 has improved. **g, h** Postoperative plain computed tomography. **g** Immediate postoperative. **h** Seven months postoperatively. **i** Twelve months postoperatively



cases that do not improve after 1 week to 2 weeks of orthotic treatment or when symptoms are severe [1, 4]. Surgical treatment is rarely performed and is indicated only in cases that are resistant to conservative treatment and with recurrence of symptoms [5]. Surgical treatment can be divided into anterior, posterior, and combined anterior–posterior methods. The purpose of the anterior method is to restore the rotational position to oral access [6, 7]. The anterior method involves oral entry to palpate the anterior arch of C1 to assist with traction and rotation [8].

This case is characterized by the following:

- The patient was a 17-year-old girl whose growth phase has ended, and she had adult bone morphology (Fig. 1d).
- The patient was refractory to conservative treatment.
- Although the dislocation was easily reduced with halo fixation traction under general anesthesia, the dislocation easily recurred without traction.

Based on the above characteristics, we believed that posterior fixation was necessary, and we were able to achieve adequate restoration and fixation through surgical treatment. Abnormal morphology of the atlantoaxial joint may

be the cause of the present AARF. Despite reports that morphologic abnormalities of the atlantoaxial joint may cause AARF if there is a history of Klippel-Feil syndrome [9], there has been no report of morphologic abnormalities of the atlantoaxial joint being the cause of AARF without a medical history; thus, it is unlikely to be the cause of AARF in this case. Few reports have shown an association between AARF and COVID-19. We are the first to report this case of AARF after COVID-19 exposure in a young-adult female whose growth phase has ended. Although there is a report of soft tissue inflammation around the cervical spine as a cause of AARF [10], it is unlikely that soft tissue inflammation is caused by COVID-19. Furthermore, there is no reported association between COVID-19 and Grisel syndrome. Because the patient had mild inflammatory findings on the blood tests during her first medical examination, there was a small possibility of Grisel syndrome; however, Grisel syndrome in adults is rare, and conservative treatment has been reported to be unsuccessful [11]. Katsuyama et al. summarized previous reports of AARF in adults and developed a new treatment flowchart. They reported that early diagnosis, i.e., within 1 month, is necessary to achieve improvement with conservative treatment. However, adults with AARF

Fielding types 2, 3, and 4 are resistant to conservative treatment, and surgical treatment is necessary [12]. Kitamura et al. [13] reported that AARF with atlantoaxial dysplasia in childhood can be remodeled by halo fixation for 3 months. However, in our case, the growth phase has ended, and the possibility of remodeling the C1/2 joints was deemed to be low; thus, the patient underwent surgical treatment with halo traction and fixation for approximately 1 month. As the postoperative reconstructed CT images obtained at 3, 7, and 12 months did not show any remodeling of the C1/2 anterior facet joint and as the growth phase of the patient has already stopped, we recommended fixation surgery early in this case.

Conclusion

This is the first report on a case of nontraumatic AARF that may have been triggered by COVID-19 in a healthy 17-year-old young girl. The association between COVID-19 and Grisel syndrome remains unclear, but we believe there may be a relationship. We believe that early surgery is preferable in AARFs with completed growth phase because the C1/2 joints are less likely to remodel when the growth period is already completed.

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Data availability Not applicable.

Declarations

Conflict of interest The authors declare no conflict of interest.

Ethical approval This study was conducted after the protocol was approved by the Institutional Review Board of Kindai University Hospital (Control Cohort Study, no. 2020–25).

Consent to participate Informed consent was obtained from the patient and her parents.

Consent to publish The patient and her parents were informed that the case would be submitted for publication, and they provided consent.

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