under general anesthesia at previous hospitals before visiting our institute, and temporal reduction was obtained only in one patient (recurrent unstable AARF case: case 2), and no reduction was obtained after the traction for averaged period of 2.7 weeks in the other six patients (irreducible AARF cases). The clinical data at the initial visit were summarized in Table 1.

IMAGING AND RADIOGRAPHIC PARAMETERS

All patients underwent plain radiography of the cervical spine. They also received 3D CT reconstructions and dynamic CT, under sedation if necessary, as proposed by Pang and Li.[30] AARF was classified according to Fielding's classification,[2] a typing system established by Pang and Li,[30] and our grading system reported previously.[6] Dynamic CT was examined at three different positions, consisting of the neutral position and the maximum rotated positions to both sides. The angles made by occiput and the vertical 0° (the midline) and the separation angle between C1 and C2 were calculated at each head position. Magnetic resonance images were also obtained to assess the degree of spinal cord compression and the presence of an inflammatory lesion.

CLOSED MANIPULATION AND EXTERNAL IMMOBILIZATION

The patients were positioned supine on an operating table under lateral fluoroscopy. To prevent the spinal cord damage, either endoscopically guided endotracheal or awake tracheal intubation was performed. Under the general anesthesia, a halo ring was placed using six skull pins. The head was gently manipulated by holding the halo ring, and reduction was attempted by applying slight axial traction first, followed by translating the head posteriorly with only minimal rotation until a slight click indicating reduction was felt. This typically extends the inferior cervical spine and holds the superior cervical spine in a neutral or slightly extended and nearly reduced normal position (Video 1, supplemental digital content, online only, available at: http://links.lww.com/BRS/A496). After confirmation of the reduction by fluoroscopy (Figure 1, supplemental digital content, online only, available at: http://links.lww.com/BRS/A497), the head was slowly rotated to check if a rotatory movement of C1–C2 joints was regained. The rotation of C1–C2 could be confirmed by the fluoroscopic image. If the confirmation fails, the bumps of each C1 and C2 facet joint which appears during the neck rotation is easily felt through the mouth by a finger (Video 2, supplemental digital content, online only, available at: http://links.lww.com/BRS/A498). The bumps created from C1 to C2 facet joint generally reveal a see–saw relationship during the neck rotation, suggesting that C1 could cross over C2 at each rotational movement. Finally, the halo ring was connected to a vest with neck in slight extension.

Follow-up plain radiographs were taken every 1 to 2 weeks, and CT images were performed every month after closed reduction and halo vest fixation until the remodeling was confirmed. After confirmation of C2 remodeling on CT images, the halo vest was removed and the patients were encouraged to start a gentle neck motion exercise in all directions for approximately 10 minutes three times a day. Clinical courses and radiographic findings were retrospectively reviewed.

RESULTS

Pretreatment Radiographic Finding

Plain radiographs and CT images at the initial visit revealed atlantoaxial subluxation with a mean atlantodental interval of 6.3 (3.5–13) mm. According to Fielding's classification, type I subluxation was observed in one case, type II in three cases, and type III in three cases (Table 1). Three-dimensional-CT represented a tilt of C1 in the coronal plane and C2 facet deformity in the side of dislocation in all cases. Using our criteria reported previously, the subjects were classified into grade II in four and grade III in three cases. The degrees of the C2 facet deformities were slight in four cases, moderate in one case, and severe in two cases. On axial CT images, an average angle between the occiput and the midline, was 10.8° (6.7°–15°) toward the contradislocated side. On dynamic CT images, a maximum mean angle of neck rotation was 17.1° (−3°–25°) from the midline in the dislocated side, which was significantly limited compared to 45.4° (37°–54°) in the contralateral side (Figures 1A–D). No mobility of the C1–C2 joints was observed in five cases, which fell into type I of Pang's classification. *Pang's classification type I* is defined as an unaltered ("locked") C1–C2-coupled configurations regardless of corrective counterrotation (Figures 1E and 1F). However, no bony union between C1 and C2 was observed on 3D CT in

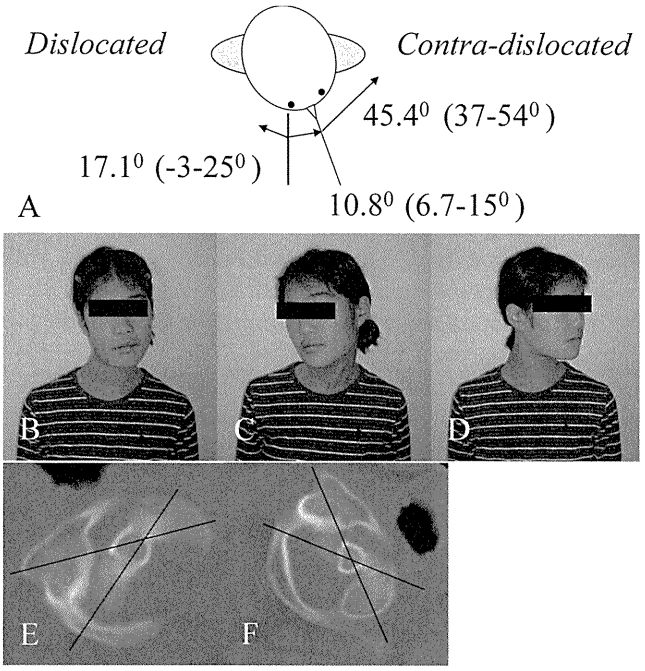

Figure 1. **A**, Averaged angles of neck rotation on pretreatment dynamic CT images in all cases. Torticollis (so-called cock-robin posture) **(B)** limitation of neck rotation **(C, D)** at the initial visit. The pretreatment dynamic CT images under awake condition forcing the maximum rotation to both sides reveal that the separation angle between C1 and C2 are fixed **(E, F)**.

these five cases of our series. Remaining two cases were classified as Pang's type II, which has the reduction of the rotation angle between C1 and C2 with forced correction, but C1 could not be made to cross C2. No active inflammation around the bilateral C1–C2 joints was detected on magnetic resonance images in any case.

CLOSED MANIPULATION

Because no patients showed C1–C2 bony unions, the closed reduction under the general anesthesia was attempted to be reduced in all seven cases. After the halo ring was installed, the head was gently held with natural torticollis position, and the reduction of subluxation is easily achieved immediately after the patient being under anesthesia in five of all cases (71.4%). In other two cases (28.6%), the reduction was easily obtained by slight axial traction and translating the head posteriorly with only minimal rotation, through a halo ring. The confirmation of reduction by fluoroscopic image was not clearly obtained in four patients, whereas the bumps created from C1 to C2 facet joint with a see–saw movement during the neck rotation was easily felt to confirm the C1–C2 rotational movement in these all patients. After the reduction of subluxation, a halo vest was installed. On CT images and clinical examinations immediately after the reduction, neither bone fracture nor spinal cord injury was observed in all patients.

FOLLOW-UP RADIOGRAPHIC FINDINGS: REMODELING OF THE C2 FACET DEFORMITY

All patients were carefully monitored using both plain radiographs and CT images after the application of the halo vest. Interestingly, a sign of remodeling of the C2 facet deformity appeared on 3D CT images at approximately 1 month after halo fixation in all seven cases. When the adequate remodeling of C2 facet deformity was recognized on 3D CT (Figure 2), the halo ring was dismantled and replaced by a soft cervical collar at the average of 2.8 months after the application of the halo vest. Thereafter, the gentle exercise of neck motion in all directions was started. The cervical ROM recovered within 2 weeks after removing the halo in five cases (Figures 2E–G). The mean occiput angle from the midline measured with a goniometer improved from 10.8° (6.7°–15°) to 0° (−3°–4°). The cervical ROM was macroscopically 87° (80°–90°) at the dislocated and 91° (85°–95°) at the contradislocated side from the midline. Although in other two irreducible AARF cases (cases 3 and 5) with severe C2 facet deformity, closed manipulation was obtained successfully, C1–C2 fibrous contracture developed after halo vest fixation resulting in the limitation of neck ROM. However, neither recurrence of symptoms nor subluxation was observed at the final follow-up in any case.

On CT images at the final follow-up, the averaged atlantodental interval improved from 6.3 (3.5–13) mm to 2.0 (1.5–2.3) mm on axial CT images. The C2 facet deformity remodeled completely in five patients and partially in two patients (cases 3 and 5). On dynamic CT images, the mean angle of maximum neck rotation from the midline was 75.9° (50°–90°) to the dislocated side and 82.4° (65°–92°) to the contralateral side (Table 2). Furthermore, the separation angle between C1 and C2 improved significantly after the treatment (Figures 2H, 2I; Table 2).

DISCUSSION

The pathophysiology of chronic irreducible and recurrent unstable AARF remains unclear despite numerous previous studies regarding AARF. In recent years, algorithms advocated either by Phillips and Hensinger[3] or by Pang and Li[13] have generally been used as a treatment protocol for chronic AARFs. According to their algorithms, either halter or skull caliper tractions are recommended as an initial treatment. If the reduction can be achieved, halo external fixation should be the next step of treatment. If the subluxation is irreducible or recurrent, surgical procedures including posterior C1–C2 fixation with or without an anterior release have been recommended in both algorithms. Generally, the recurrence has often been observed even after the closed reduction in patients with a chronic AARF.[2–4,6,7,9,10] Pang and Li[13] also reported a high recurrence rate in especially Pang's type I and II chronic AARFs. Therefore, a majority of surgeons favor surgical treatments for chronic AARF to prevent recurrence.[2,6,7,9,13] On the contrary, atlantoaxial arthrodesis is associated with several problems including serious complications such as vertebral artery injury,[26,27] pseudarthrosis, and loss of ROM at the atlantoaxial joint.[28,29] These downsides of surgical treatments make conservative treatments more feasible option. Previously, several investigators reported a chronic AARF case treated by a simple traction and a bracing.[29,31] Burkus and Deponte[29] revealed that successful closed reduction was obtained using cervical traction and bracing in a chronic AARF case. Recently, Park *et al*[31] reported that a chronic AARF patient was successfully treated with 6-week long-term halter

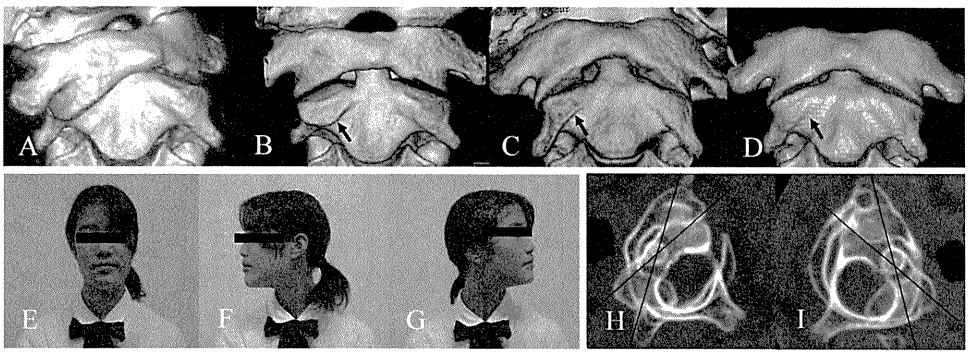

Figure 2. Three-dimensional-CT images before (A) and after the closed manipulation (B, immediately; C, 3 months; D, 1 year after closed reduction) in case 1. The remodeling of the C2 facet deformity is observed (arrow in B–D). The normal posture and neck ROM are obtained 1 year after closed reduction (E–G, macroscopically images; H, I, dynamic CT images).

TABLE 2. Summary of Clinical Data at the Final Follow-up

Case	Follow-up Period (mo)	Closed Reduction	Remodeling of C2 Facet Deformity	Period of Halo Fixation (mo)	ADI (mm)	Recurrence	Max Rotation Angle (°)		C1–C2 Angle (°)		
							Rt	Lt	Rt	Midline	Lt
1	28	+	+	3.1	2.3	−	85	92	−35	3	33
2	23	+	+	2.3	1.9	−	90	90	−27	4	42
3	21	+	+	3.8	2.2	−	55	65	−0.4	7	5
4	18	+	+	2.1	1.5	−	86	88	−25	−1	41
5	17	+	+	2.9	2.2	−	50	65	4.4	16	12
6	11	+	+	2.1	2.3	−	80	90	−33	−6	31
7	6	+	+	3.0	1.5	−	85	89	−35	2	34

ADI indicates atlantodental interval;; Lt, left; Max, maximum; Rt, right; +, slight; −, none.

traction followed by 4-month bracing and 2-month collar fixation. However, the lack of an appropriate algorithm and the uncertainty of the outcomes of conservative therapy remain to be clarified.

In the present study, we have reported a novel conservative treatment strategy for chronic AARF. By careful closed manipulation followed by halo fixation, the remodeling of C2 facet deformity was successfully achieved preventing recurrence of subluxation in seven consecutive patients with chronic irreducible and recurrent unstable AARFs. Prevention of recurrence and good cervical ROM was obtained in all seven cases at the final follow-up. To our knowledge, this is the first report describing that the remodeling of the C2 facet deformity can be a useful radiographic parameter to determine the timing for the removal of a halo device. Moreover, the prevention of recurrent subluxation by the remodeling of C2 facet deformity supports our previous data that C2 facet deformity is a risk factor of the recurrence.[6] In our protocol, we use closed manipulation instead of halter or skull caliper tractions for reduction. Phillips and Hensinger,[3] and Pang and Li,[13] respectively, recommend a bed rest with halter or skull caliper tractions as an initial treatment in the chronic AARF. However, a patient usually suffers from severe stress during these tractions. And the successful reduction is not always achieved despite prolonged traction, resulting in greater stress. In contrast, the closed manipulation under the general anesthesia is very simple and prompt. According to our experiences from seven consecutive cases, the reduction has been easily obtained by manipulation with or without slight axial head traction under general anesthesia. Moreover, Pang and Li[13] mention that highest relapse rate was observed in their chronic type I and II AARF cases even if the reduction was achieved during the halter or skull caliper tractions. From this standpoint, the closed manipulation would also be advantageous, because the halo vest can be applied under the fluoroscopy as soon as the reduction is achieved, preventing the recurrence. We suggested that the remodeling of facet deformity is a definite basis in our protocol. Even if the reduction can be obtained by closed reduction in chronic AARF cases, no one knows exactly when to remove external immobilization after reduction. Even though Pang and Li[13] recommend external immobilization with halo vest for a length of 3 months after reduction, this 3-month period is based on their clinical experiences. In this study, we have found that the remodeling of the C2 facet deformity prevents the recurrence and that this remodeling sign can be a useful radiographic parameter to determine the timing for the removal of a halo device. In deed, our three patients (cases 2, 4, and 6) obtained a good clinical outcome without recurrence even after approximately 2 months external immobilization, because these three patients revealed the sufficient remodeling of the facet deformity on 3D CT at 2 months after the application of the halo vest.

In this series, gradual remodeling of the C2 facet deformity appeared on 3D CT at approximately 1 month after reduction in all seven cases. In five cases, complete remodeling was seen at 2 to 3.1 month on 3D CT and the normal cervical ROM was obtained within 2 weeks after the removal of the halo vest (*e.g.*, case 1; Figure 2). In other two cases with severe C2 facet deformity resulting from trauma (cases 3 and 5; Figure 3), a partial remodeling was obtained at approximately 3 months (Figure 3). These two cases had a limitation of cervical ROM at the final follow-up, possibly because of fibrotic contracture of the C1–C2 joints. It is considered that the direct damages of the articular surface of the C1–C2 joint due to trauma may cause a severe C2 facet deformity resulting in a limited neck ROM. Our treatment strategy may have a limitation for such patients with severe C2 facet deformity resulting from trauma. On the contrary, the contraindication to this procedure is a patient with bony union between C1 and C2 joints that can be elucidated on 3D CT and dynamic CT images. The manipulation of C1–C2 subluxation may have high risks of bone fracture and spinal cord injury in such cases. It is most likely, however, that the movement of C1–C2 joints is prevented by neck pain and spasm of the sternocleidomastoid muscle[3,6] during dynamic CT examination in most cases. Therefore, we recommend administration of sleep agents during the dynamic CT examination to

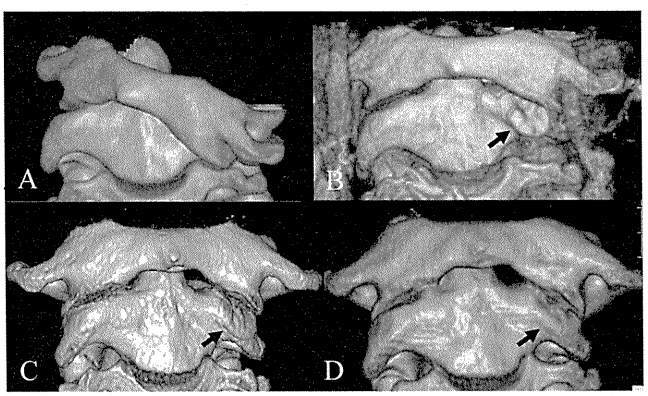

Figure 3. Three-dimensional-CT images before (A) and after the closed manipulation (B, immediately; C, 2 months; D, 6 months after closed reduction) in case 5 with a severe C2 facet deformity. The incomplete remodeling of C2 facet deformity is seen (arrow in B–D).

assess the exact status of C1–C2 joints. In fact, no movement of C1–C2 joints was observed on 3D CT images under awake condition in five of seven patients in our series, despite no patients had bony or fibrous union between C1 and C2 joints under sleeping condition.

It is well-known that the pediatric bones have a remodeling potential. Toyama *et al*[32] have reported that realignment of the postoperative cervical kyphosis by vertebral remodeling in the children indicating that the vertebrae have a remodeling potential similar to that of long bones of the extremities. However, there are no previous reports describing the remodeling of the facet joint in AARF patients. The axis has five primary and two secondary ossification centers. The superior C2 facet is derived from lateral masses (neural arches), which are two of five primary ossification centers. The neural arches at both sides generally fuse by 3 years of age, whereas two secondary ossification centers fuse with base of the dens between 10 and 13 years and with the vertebral body between 15 and 25 years.[33,34] It is suggested that the axis may have a higher remodeling potential even in adolescence, compared with other vertebral bones. The oldest patient in our series was an 11-year-old girl in whom the complete C2 facet remodeling was obtained 3 months after closed reduction. Considering that AARF involve children with a wide range of age, this novel treatment strategy is applicable to most children with chronic AARFs.

CONCLUSION

Chronic AARFs without C1–C2 bony union were managed successfully by careful closed manipulation followed by halo fixation. The remodeling of the C2 facet deformity on 3D CT is a useful radiographic parameter to decide the appropriate period of halo fixation in this new treatment strategy that can obviate the need for surgical intervention.

➢ Key Points

- ❑ A new treatment strategy for chronic AARF was proposed.
- ❑ The C2 facet deformity observed in seven chronic AARF patients remodeled spontaneously after closed manipulation followed by halo fixation.
- ❑ The remodeling of C2 facet deformity prevented the recurrence of AARF maintaining neck ROM intact in most cases.
- ❑ Closed manipulation followed by the halo vest fixation techniques can be a good alternative to surgery.
- ❑ The remodeling sign on CT scans can be a useful radiographic parameter to determine the period of halo fixation.

Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF versions of this article on the journal's Web site (www.spinejournal.com).

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