JNS SPINE

CLINICAL ARTICLE

J Neurosurg Spine 34:498–505, 2021

Primary treatment of atlantoaxial rotatory fixation in children: a multicenter, retrospective series of 125 cases

Takahiro Shimazaki, MD, Kei Yamada, MD, PhD, Kimiaki Sato, MD, PhD, Kotaro Jimbo, MD, PhD, Hidetomo Nakamura, MD, PhD, Masafumi Goto, MD, Tsunemasa Matsubara, MD, PhD, Kenji Mizokami, MD, Shoji Iwahashi, MD, Takeharu Sasaki, MD, and Naoto Shiba, MD, PhD

Department of Orthopaedic Surgery, Kurume University School of Medicine, Kurume, Japan

OBJECTIVE The primary treatment for atlantoaxial rotatory fixation (AARF) remains controversial. The aim of this study was to investigate the primary treatment for AARF and create an algorithm for primary treatment.

METHODS The authors analyzed the data of 125 pediatric patients at four medical institutions from April 1989 to December 2018. The patients were reported to have neck pain, torticollis, and restricted neck range of motion and were diagnosed according to the Fielding classification as type I or II. As a primary treatment, 88 patients received neck collar fixation, and 28 of these patients did not show symptom relief and required Glisson traction. Thirty-seven patients were primarily treated with Glisson traction. In total, 65 patients, including neck collar treatment failure patients, underwent Glisson traction in hospitals.

RESULTS The success rate of treatment was significantly higher in the Glisson traction group (97.3%) than in the neck collar fixation group (68.2%) ($p = 0.0001$, Wilcoxon test). In the neck collar effective group, Fielding type I was more predominant ($p = 0.0002$, Wilcoxon test) and the duration from onset to the first visit was shorter ($p = 0.02$, Wilcoxon test) than that in the neck collar ineffective group. Using multivariate logistic regression analysis with the above items, the authors generalized from the estimated formula: logit [p(success group by neck collar fixation group)|duration from onset to the first visit (x_1), Fielding type (x_2)] = 0.4(intercept) − $0.15x_1 + 1.06x_2$, where x_1 is the number of days and $x_2 = 1$ (for Fielding type I) or −1 (for Fielding type II). In cases for which the score is a positive value, the neck collar should be chosen. Conversely, in cases for which the score is a negative value, Glisson traction should be the first choice.

CONCLUSIONS According to this formula, in patients with Fielding type I AARF, neck collar fixation should be allowed only if the duration from onset is ≤ 10 days. In patients with Fielding type II, because the score would be a negative value, Glisson traction should be performed as the primary treatment.

https://thejns.org/doi/abs/10.3171/2020.7.SPINE20183

KEYWORDS atlantoaxial rotatory fixation; Fielding type; neck collar fixation; Glisson traction; cervical

Neck conditions such as neck pain and torticollis are common in children. Although atlantoaxial rotatory fixation (AARF) is comparatively rare, it often recurs.[1,2] AARF is often refractory, chronic, and recurrent due to delayed diagnosis and ineffective conservative treatment.[3,4] Although conservative treatment occasionally improves AARF,[3–8] surgery is required in many cases.[9–11] Appropriate treatment yields good outcomes. However, treatment guidelines for AARF are not clearly established; there is debate regarding treatment with external fixation using neck collars or with Glisson traction.[5,12,13] Several case reports and small case series[9,10,12,14,15] have focused on AARF; however, to the best of our knowledge, no study has reviewed more than 100 patients with this disorder. The present study aimed to investigate a large number of AARF cases to construct an algorithm that could be used to determine the best treatment for each patient.

Methods

We investigated 169 patients diagnosed with AARF by orthopedists during their initial visit to their consulting doctor at one of four different medical facilities affiliated with our hospital—Kurume University Hospital, St.

ABBREVIATIONS AARF = atlantoaxial rotatory fixation; ROM = range of motion.
SUBMITTED February 18, 2020. **ACCEPTED** July 1, 2020.
INCLUDE WHEN CITING Published online December 4, 2020; DOI: 10.3171/2020.7.SPINE20183.

Mary's Hospital, Chikugo City Hospital, and Kawasaki Hospital—between April 1989 and December 2018.

The inclusion criteria were as follows: 1) the presence of neck pain, torticollis, and/or limited range of motion (ROM) of the cervical spine, 2) laterality in the distance between the lateral mass and the facet for dens on simple radiographs, 3) availability of patient CT images, and 4) implementation of follow-up until completion of treatment. The following cases were excluded: 1) patients for whom a cervical spine CT scan could not be obtained due to lack of patient cooperation or for whom CT images were not available, 2) cases not treated with neck collar fixation or Glisson traction during initial treatment, 3) Fielding classification[16] type III or IV, 4) cases with onset after school age, and 5) dropout cases. The definition of Fielding classification, according to Fielding and Hawkins,[16] is as follows: type I is a rotatory fixation with no anterior displacement of the atlas (displacement ≤ 3 mm) and with the odontoid acting as the pivot, whereas type II is a rotatory fixation with an anterior displacement of 3 to 5 mm and one lateral articular process acting as the pivot.

The decision to choose type I or II was made not only by orthopedists at the initial visit but also by more than two spine surgeons and based on the radiogram interpretation reports by radiologists at each hospital. In cases where the spine specialists had a disagreement on type I or II, a decision was made based on the radiogram interpretation reports from radiologists in each hospital. Among the 169 patients initially studied, 44 were excluded (Fig. 1).

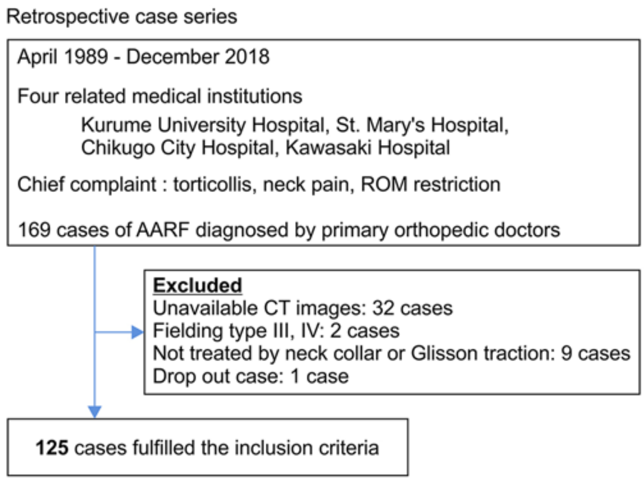

FIG. 1. Study design. Figure is available in color online only.

Study Parameters

We investigated the following parameters: age at diagnosis, sex, Fielding type[16] I or II, cause of onset, duration from onset to the first visit, duration of follow-up, primary treatment (neck collar fixation and/or Glisson traction), and overall success rate of the primary treatment. Patients were classified into four groups based on the cause of onset: the unknown group (no clear cause), the major trauma group (traffic accidents, falls, or sporting incidents), the minor trauma group (activities of daily living, such as turning around or undressing), and the postinflammation group (otorhinolaryngological inflammation such as parotitis or tonsillitis). The patients in the postinflammation group had improved inflammation findings at the initial visit and did not need additional antibiotic therapy. Patients were treated with external fixation using a neck collar or with Glisson traction in the hospital at the discretion of the first consulting doctor. If the attending physician determined that there was no improvement in pain or restriction of neck motion during the treatment with a neck collar, the patient was admitted and treatment was switched to Glisson traction. The Glisson traction method was performed with the patient in the supine position on a bed and undergoing traction with 1.5-kg weights. When there was no improvement with Glisson traction, manipulative reduction was performed under general anesthesia as described in the report by Ishii et al.[6] If the halo ring was used, the head was gently manipulated by holding the halo ring, and the 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. Then, halo vest fixation was performed. The duration of collar fixation, duration of Glisson traction, timing of conversion from collar fixation to Glisson traction, and duration of external fixation after Glisson traction were decided by the attending physician. The criteria for determining whether the primary treatment was a success or a failure were as follows: treatment was deemed successful in patients whose condition was cured with a neck collar and/or Glisson traction, and treatment was deemed a failure in patients whose condition did not improve with a neck collar and Glisson traction and who required transition to a halo vest.

To compare the treatment outcomes, we divided the patients into two groups: neck collar fixation and Glisson traction. Patient age, sex, Fielding classification,[16] cause of pain onset, duration from onset to the first visit, and the duration of each treatment were investigated for each fixation group.

Next, patients in the collar fixation group were classified into the neck collar effective group, in whom the condition was cured with a neck collar, and the neck collar ineffective group, in whom the condition was not cured with a neck collar and the patients were transitioned to Glisson traction. Factors associated with poor outcomes were extracted using statistical analysis. We investigated and analyzed each item using univariate analysis. Then, we performed a multivariate logistic regression analysis using items with significant differences as explanatory variables to help determine the primary treatment.

Ethical Considerations

We anonymized patient data and used them for research with parental consent or disclosure obtained via a website (http://www.med.kurume-u.ac.jp/med/joint/rinri/ushiromukikenkyu/18292.pdf, http://www.st-mary-med.or.jp/resources/file/pdf/2019061908_54_16.pdf). This study was implemented in accordance with the Declaration of Helsinki and was conducted with the approval of the Kurume University ethical review board (no. 18292) and the St. Mary's Hospital institutional ethical review board (no. 19-0506).

TABLE 1. Demographic data

Variable	Value
Age at diagnosis, yrs	
Mean ± SD	6.1 ± 2.5
Median (range)	6 (1–13)
Sex, no.	
Male	63
Female	62
Fielding classification	
Type I	101 (81%)
Type II	24 (19%)
Cause	
Unknown	54 (43%)
Major trauma	32 (26%)
Minor trauma	21 (17%)
Inflammation	18 (14%)
Duration from onset to the 1st visit, days	
Mean ± SD	4.2 ± 10.3
Median (range)	1 (0–60)
Duration of follow-up, days	
Mean ± SD	37 ± 58.6
Median (range)	10 (1–302)

Values are presented as number of patients (%) unless otherwise indicated.

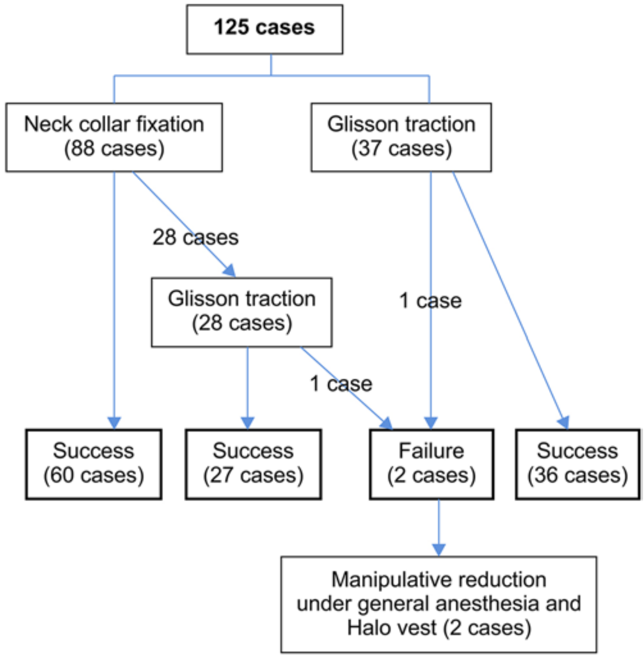

FIG. 2. The details of the primary treatment. Figure is available in color online only.

Statistical Analysis

We used the JMP Pro 13.0.0 software (SAS Institute Inc.) for the statistical analysis. Statistical analysis was performed in cooperation with experts, including a professor at the Biostatics Center at Kurume University. The Wilcoxon test, chi-square test, and Kruskal-Wallis test were used to compare the neck collar fixation and Glisson traction groups, and the effective and ineffective neck collar treatment groups. In addition, when comparing the effective and ineffective neck collar fixation groups, we analyzed items with significant differences using multivariate logistic regression analysis. A p value < 0.05 was deemed to be a significant difference for all tests.

Results

Among the cases studied, 125 fulfilled the inclusion criteria initially (Fig. 1). The demographic data are provided in Table 1. The mean age was 6.1 ± 2.5 years (range 1–13 years, median 6 years), and the study population included 63 boys and 62 girls. There were 101 (81%) and 24 (19%) cases of Fielding classification type I and II, respectively. There were 54 (43%), 32 (26%), 21 (17%), and 18 (14%) cases in the unknown, major trauma, minor trauma, and postinflammation groups, respectively. The mean duration between onset and the first visit was 4.2 ± 10.3 days (range 0–60 days, median 1 day). The mean follow-up duration was 37 ± 58.6 days (range 1–302 days, median 10 days) (Fig. 2).

Among the 125 cases, neck collar fixation and Glisson traction were used in 88 and 37 cases, respectively. Of the 88 cases in the neck collar fixation group, pain, torticollis, and ROM restriction were reduced in 60 patients. The other 28 patients did not show improvement and underwent Glisson traction, and 27 of these patients showed improvement. In the primary Glisson traction group, 36 of the 37 cases showed improvements. Two patients who did not improve after the primary treatment underwent manipulative reduction under general anesthesia and halo vest fixation. One had a reduction in all symptoms, and the other reported slight ROM restriction. The success rate of primary treatment was 98% (123 of 125 cases).

The median treatment duration was 8 days (range 1–56 days, mean 12.1 ± 10.6 days) in the neck collar fixation group and 6 days (range 1–41 days, mean 8.9 ± 9.1 days) in the Glisson traction group. The median duration of neck collar fixation after reduction with Glisson traction was 10 days (range 0–77 days, mean 13.7 ± 15.4 days). There were no significant differences in sex, cause, and duration from symptom onset to the first visit. In contrast, the age was significantly higher in the Glisson traction group than in the neck collar fixation group ($p = 0.037$). The success rate was significantly higher in the Glisson traction group (97.3%) than in the neck collar fixation group (68.2%) ($p = 0.0001$) (Table 2).

In the neck collar fixation group (Table 3), there were no significant differences in age, sex, or cause ($p = 0.25$, $p = 0.36$, $p = 0.29$, respectively) between the effective and ineffective groups. When patients were assessed based on the Fielding classification,[16] type I was significantly more prevalent in the neck collar effective group and type II was significantly more prevalent in the neck collar ineffective group ($p = 0.0002$). The duration from onset to the first visit was significantly longer in the neck collar effective group ($p = 0.02$). Finally, from the aforementioned in-

TABLE 2. Comparison of primary treatments: neck collar fixation and Glisson traction

	Patient Group		
	Neck Collar Fixation	Glisson Traction	p Value
Age, yrs	5.8 (1–13)	6.6 (3–12)	0.037*
Sex, M/F	44/44	19/18	0.89
Fielding classification			0.06*
Type I	75	26	
Type II	13	11	
Cause			0.83
Unknown	40	14	
Minor trauma	14	7	
Major trauma	21	11	
Inflammation	13	5	
Duration from onset to 1st exam, days	2.8 (0–49)	7.6 (0–60)	0.26
Rate of successful cases	60/88 (68.2%)	36/37 (97.3%)	0.0001*

Values are presented as number of patients (%) or median (range) unless otherwise indicated.
* Statistically significant.

formation, we performed multivariate logistic regression analysis using two significant items, the Fielding classification type and the duration from onset to the first visit. These results are shown in Table 4. The estimates of Fielding classification, SE, and OR were 1.06, 0.37, and 8.25 (95% CI 1.9–35) ($p = 0.0042$), respectively. The estimated duration from the onset to the first visit, SE, and OR were −0.15, 0.069, and 0.86 (95% CI 0.75–0.99) ($p = 0.0034$), respectively. In addition, we created the following formula: logit [p(neck collar effective group)|duration from onset to the first visit(x_1), Fielding type (x_2)] = 0.4(intercept) − 0.15x_1 + 1.06x_2, where x_1 is the number of days and $x_2 = 1$ (if Fielding type I) or −1 (if Fielding type II).

TABLE 3. Comparison between the success and failure subgroups of neck collar fixation

	Subgroup		
	Success	Failure	p Value
Age, yrs, mean ± SD (range)	5.6 ± 2.6 (1–13)	6.1 ± 2.1 (3–11)	0.25
Sex, M/F	32/28	12/16	0.36
Fielding classification			0.0002*
Type I	57	18	
Type II	3	10	
Cause			0.29
Unknown	28	12	
Major trauma	15	6	
Minor trauma	11	3	
Inflammation	6	7	
Median duration from onset to 1st visit, days	0	2	0.02*

* Statistically significant.

TABLE 4. Multivariate logistic regression analyses

	Estimate	SE	OR (95% CI)	p Value
Fielding classification	1.06	0.37	8.25 (1.9–35)	0.0042
Duration from onset to 1st visit	−0.15	0.069	0.86 (1.9–35)	0.0034

Reference Case

The patient was a 12-year-old boy diagnosed with likely torticollis while being treated for parotitis by his family doctor. However, he received expectant management. He was referred to our hospital 60 days after onset of neck pain, slight torticollis, and ROM restriction (Fig. 3A). His AARF was classified as Fielding type I based on cervical spine CT (Fig. 3B and C); engagement of joint surface and osteophyte formation were not indicated at the atlantoaxial joint on 3D CT (Fig. 3D). We admitted this patient after the first examination and administered Glisson traction for 2 weeks, which reduced the neck pain. ROM restriction was not evident in the supine position (Fig. 3E) but was substantial in the sitting position (Fig. 3F). Because ROM restriction persisted after 5 weeks of Glisson traction, we assessed the motor function of the atlantoaxial joint with CT according to the report by Pang and Li.[17] We plotted the angles of C1, C2, and C1–C2 at the intermediary and the right and left rotational positions. He was assessed as being in the diagnostic gray zone of the Pang classification. As osseous ankylosis was absent, we concluded that the patient could be discharged with a cervicothoracic brace and should be monitored. After discharge, the AARF recurred. However, we elevated the lower jaw part of the brace, which ameliorated his symptoms.

Discussion

AARF responds to conservative treatment (including traction) in most cases, with few patients requiring surgery.[3–8] Nevertheless, many of the available studies on AARF are individual case reports and small case series.[9,10,12,14,15] As a result, no treatment algorithm has been established. To the best of our knowledge, our study is the first AARF case series with more than 100 cases.

First, we examined the overall success rate of the primary treatment to determine the appropriate conservative treatment. Conservative treatment was successful in 123 of 125 patients (98%), with almost all patients cured. This success rate for the current study is higher than the following previously reported rates: 84% (24 of 28) in the study by Beier et al.,[12] 87% (20 of 23) in the study by Phillips and Hensinger,[15] and 61% (14 of 23) in the study by Rahimi et al.[2] This suggests that our primary treatment policy was appropriate.

The success rate in the current study for the Glisson traction group (97.3%) was significantly higher than the success rate for the collar fixation group (68.2%). Traction therapy is used widely without distinction between acute

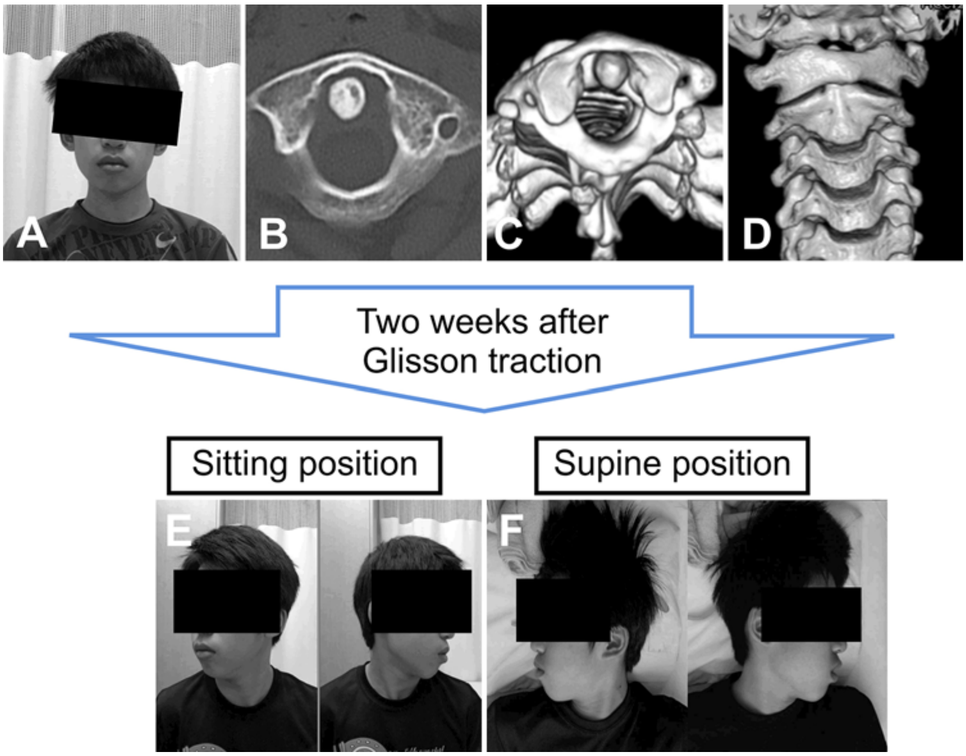

FIG. 3. Reference case: a 12-year-old boy presenting with inflammation. **A:** Residual slight torticollis. **B:** Height of the atlas on CT. **C:** 3D CT, Fielding type I. **D:** 3D CT showing no engagement of the joint surface or formation of osteophytes. **E:** In the sitting position, ROM restriction remained 2 weeks after Glisson traction. **F:** In the supine position, ROM restriction disappeared. Figure is available in color online only.

and chronic cases. Beier et al.[12] reported improvement in 3 of 7 AARF cases with traction as the initial treatment; the remaining 4 patients were transitioned from halter traction (range 1 day to 1 month) to halo traction, which yielded results. In this study, in the neck collar ineffective group, the duration from onset to the first visit was significantly longer than that in the neck collar effective group. Considering the pathology of AARF, from onset to the first hospital visit, there is soft tissue invaginating into the joint and contracture of soft tissue, including the joint capsule and surrounding muscles, as more time passes. The condition may become more intractable as the degree of rotational deviation increases.[1,18] Ishii et al. reported that chronic AARF caused deformation with osteophyte formation on the surface of the atlantoaxial joint.[19] Govender and Kumar reported that in the early stages, muscle spasm, swollen capsular, and synovial tissues prevented the reduction.[20] Lengthening the period from onset to the first visit results in the child remaining in the torticollis position, even if they are in pain, and the load of the head worsens the insertion status of the soft tissue, which is thought to worsen the condition. In the reference case presented in this report, there was no rotational limitation of the neck in the supine position at 2 weeks after starting Glisson traction; however, there remained a rotational limitation of the neck in the sitting position. In other words, the load of the head may cause rotation disorders in the atlantoaxial joint. There was no invagination of the atlantoaxial joint or osteophytes found on CT, which was a diagnostic gray zone pattern even when evaluated with the Pang classification, and no findings were suggestive of bony ankylosis. Therefore, the weight of the head may have worsened the state of the invagination of soft tissue. Missori et al. treated 14 cases of AARF after acute trauma with 8 to 24 hours of bed rest after fitting a cervical brace, followed by 14 days of wearing a neck collar, and reported that the condition was completely cured with no recurrence.[14] Mifsud et al. described a simple modification of halo traction that allows the children to move their heads while maintaining traction and reported that good outcomes were achieved by treating the condition with traction only, without limiting rotation.[7] That is, eliminating the factor of head load in the initial treatment may be important.

While Glisson traction is highly successful and useful, it has several associated problems. First, this treatment requires hospitalization. Many pediatric patients are younger than elementary school age. In Japan, young children admitted to the hospital must be accompanied by a family member, such as the mother, leading to an additional time burden. The economic burden of hospital fees also cannot be ignored. In this study, the approximate cost for Glisson traction with admission was approximately 100,000 Japanese yen (JPY), which is a significantly larger economic burden than the cost of 1 month of outpatient treatment for neck collar fixation, which is approximately 150 JPY. Another issue is obtaining the child's cooperation to continue with traction. In this study, the Glisson traction was adjusted depending on the extent of the child's torticollis so that only the head was slightly elevated, and the bed itself was kept horizontal; however, continuous traction full-time for 24 hours was difficult for all the pediatric patients. To counter this problem, various innovations could be considered, including switching to a seated position at mealtimes and providing the option of vertical traction, such as Good Samaritan traction. Another option could be continuous traction using sitting skull traction as reported by Chazono et al.[21] However, we treated cases with strong neck pain associated with torticollis, where vertical traction was difficult. Therefore, we did not use that method as it was not considered acceptable in children with strong neck pain, but it may be worth considering depending on the case.

To reduce the physical, mental, and economic burdens associated with Glisson traction, we investigated collar fixation treatment on an outpatient basis. In this study, the success rate of collar fixation treatment was 68.2%. Beier et al. reported that conservative treatment using collar fixation for acute-phase AARF had a success rate of 95% (21 of 22 cases).[12] Landi et al. also used conservative treatment with a collar for 9 patients, wherein the hyperintensity disappeared in MRI and all patients were cured.[22] Both reports had a higher treatment success rate than the present study, suggesting that collar fixation treatment is effective. The results of comparing the effectiveness of the neck collar showed that the effective group had significantly more cases of Fielding type I. The difference between Fielding types I and II, as reported by Landi et al., is that in type I, the STIR and T2 sequences showed a hyperintensity in the alar and capsular ligaments and in the posterior ligamentous system, with the integrity of the transverse ligament of the atlas (LTA), while in type II, the hyperintensity also involved the LTA.[22] These authors also indicated that patients with Fielding type II sustained a more unstable atlantoaxial joint, which caused a substantially greater distance between the dens and C1 anterior arch. The persistent anterior shift of C1 might provoke more severe muscle spasms and soft-tissue contracture. In other words, clear structural differences were seen between types I and II, and type I had less structural damage, suggesting that improvement of symptoms can be expected with collar therapy. Moreover, the effective group also had a significantly shorter time until the first consultation compared with the ineffective group. Phillips and Hensinger reported that the duration from onset to the first visit was related to the success of closed reduction and the length of hospitalization.[15] The reason for the effectiveness of the neck collar with a shorter duration from onset to the first visit may be that the pathology has not progressed during this time period, making it easier to achieve a better result. Given that there was no difference in the effectiveness of the neck collar based on the cause of onset in this study, we suggest that neck collar fixation should be chosen for treating Fielding classification type I cases at the early stage after onset, irrespective of the cause. Furthermore, the formula obtained by conducting multivariate logistic regression analysis using the Fielding type and the duration from onset to the first visit with significant differences as explanatory variables was as follows: score = 0.4 – 0.15 × the duration from onset to the first visit (days) + Fielding type (I, +1.06; II, –1.06). That is, if the score is > 0, then treatment success can be expected with a neck collar, while if the score is < 0, then it is most likely that treatment with a neck collar will be ineffective, and it would be better to use Glisson traction as the first treatment. In other words, based on this formula, neck collar fixation should be allowed in cases of Fielding type I when the duration from onset is about 10 days. Moreover, if the case is Fielding type II, then Glisson traction should be chosen primarily, irrespective of the duration from onset to the first visit. Figure 4 shows the algorithm obtained from the above results.

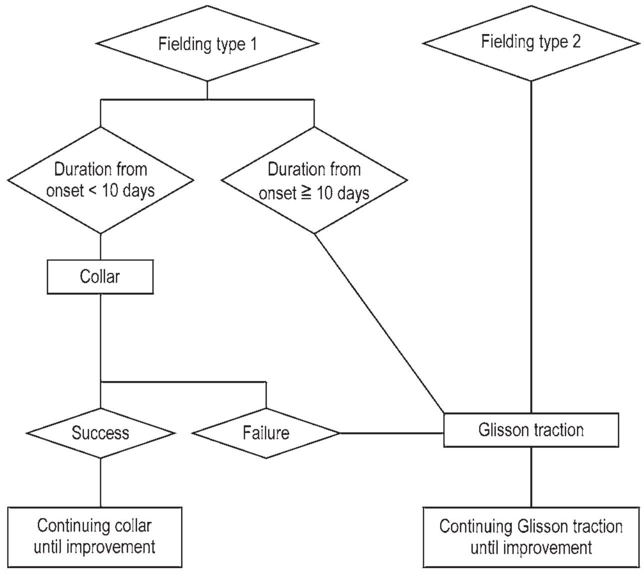

FIG. 4. Treatment algorithm of AARF.

Limitations

This study has several limitations. First, it was a retrospective study.

Second, all imaging evaluation was conducted with the Fielding classification.[16] The Fielding classification is a static evaluation, while the Pang classification[17] dynamically evaluates the movement of the atlantoaxial joint during the left and right rotation and in the neutral position, which might be more suitable for the pathological evaluation of limited ROM. Virtually none of the patients in this study were evaluated using the Pang classification, which is a limitation of the imaging evaluation. However, in this study, cooperation could not be obtained in 32 of 169 patients (19%) with AARF, suggesting that a CT scan could not be performed; therefore, conducting routine evaluations with the Pang classification is difficult and impractical. The radiation exposure with CT is a problem; thus, different imaging modalities are needed. Not all patients were evaluated using 3D CT. Several studies[9,14,20] have reported the usefulness of evaluating the damage to soft tissue, including ligaments, with MRI; however, MRI was not used for evaluation in this study. MRI for children often requires sedation, which makes MRI impractical for all pediatric patients. Landi et al. proposed confirming the absence of pathologic hyperintensity on MRI as a criterion for the removal of external fixation, but this was not confirmed in this study.[22] In addition, the determination of type I or II was made not only by orthopedists at the initial visit but also by more than two spinal surgeons and based on the radiograph interpretation reports from radiologists at each hospital. In cases where spinal specialists had a disagreement on whether it was type I or II, a decision was made based on the radiogram interpretation reports from radiologists in each hospital. However, because this study is a multicenter retrospective study involving many orthopedists, there was no rigorous criterion like that used in a prospective study.

Third, the details of the decision-making on whether to treat patients with neck collar fixation or Glisson traction could not be obtained. As this is a retrospective multiinstitutional study, we did not have any established decision-making process for treatment. The orthopedists decided on the treatment options at the initial visit for each patient. In addition, doctors involved in this study did not retain the information on treatment decision-making. However, because the overall success rate of treatment was as high as 98% (123 of 125 cases), we considered our treatment policy to be appropriate.

Fourth, we determined the improvement of symptoms with reference to the subjective symptoms of the child and examination findings, but not all patients underwent accurate objective evaluation with imaging tests, angle measurements, or other similar tests. Photographic imaging was taken as shown with the presented typical case, and the ROM was observed in some cases, but this imaging was not performed for all cases.

Fifth, it has been pointed out that the presence or absence of head support may contribute to improving the pathology; however, results were not evaluated based on different types of collars. Future studies investigating the treatment outcomes based on the type of collar selected for the initial treatment are required.

Finally, in this study it was not possible to show the permissible period of time for follow-up with the neck collar, namely, the time limit for transitioning from collar fixation to Glisson traction. Pang and Li reported using a soft cervical collar for 2 weeks for diagnostic gray zone cases, followed by halter traction for persistent symptoms.[23] Beier et al. transitioned from the cervical collar to halter traction after 3 weeks.[12] Future studies are required to examine the time limit for transitioning from collar fixation to Glisson traction.

Conclusions

We reported an AARF case series with a large number of patients in a multicenter study. Our results indicate that neck collar fixation with monitoring progress is an effective treatment for Fielding classification type I cases at an early stage after onset, irrespective of the cause.

Statistically, the score equals 0.4 − 0.15 × the duration from onset to the first visit (days) + Fielding type (I, +1.06; II, −1.06). Thus, this formula can be used as the method of decision-making for the primary treatment. In cases for which the score is a positive value, the neck collar should be chosen. Conversely, in cases for which the score is a negative value, Glisson traction should be the first choice.

Acknowledgments

We wish to thank Tatsuyuki Kakuma, Professor of the Biostatics Center, Kurume University, for useful advice on statistical analysis.

References

1. Kawabe N, Hirotani H, Tanaka O. Pathomechanism of atlantoaxial rotatory fixation in children. *J Pediatr Orthop*. 1989;9(5):569–574.
2. Rahimi SY, Stevens EA, Yeh DJ, et al. Treatment of atlantoaxial instability in pediatric patients. *Neurosurg Focus*. 2003;15(6):ECP1.
3. Chechik O, Wientroub S, Danino B, et al. Successful conservative treatment for neglected rotatory atlantoaxial dislocation. *J Pediatr Orthop*. 2013;33(4):389–392.
4. Tauchi R, Imagama S, Kanemura T, et al. The treatment of refractory atlanto-axial rotatory fixation using a halo vest: results of a case series involving seven children. *J Bone Joint Surg Br*. 2011;93(8):1084–1087.
5. Glotzbecker MP, Wasser AM, Hresko MT, et al. Efficacy of nonfusion treatment for subacute and chronic atlanto-axial rotatory fixation in children. *J Pediatr Orthop*. 2014;34(5):490–495.
6. Ishii K, Matsumoto M, Momoshima S, et al. Remodeling of C2 facet deformity prevents recurrent subluxation in patients with chronic atlantoaxial rotatory fixation: a novel strategy for treatment of chronic atlantoaxial rotatory fixation. *Spine (Phila Pa 1976)*. 2011;36(4):E256–E262.
7. Mifsud M, Abela M, Wilson NIL. The delayed presentation of atlantoaxial rotatory fixation in children: a review of the management. *Bone Joint J*. 2016;98-B(5):715–720.
8. Park SW, Cho KH, Shin YS, et al. Successful reduction for a pediatric chronic atlantoaxial rotatory fixation (Grisel syndrome) with long-term halter traction: case report. *Spine (Phila Pa 1976)*. 2005;30(15):E444–E449.
9. Crossman JE, David K, Hayward R, Crockard HA. Open reduction of pediatric atlantoaxial rotatory fixation: long-term outcome study with functional measurements. *J Neurosurg*. 2004;100(3)(Suppl Spine):235–240.
10. Crossman JE, Thompson D, Hayward RD, et al. Recurrent atlantoaxial rotatory fixation in children: a rare complication of a rare condition. Report of four cases. *J Neurosurg*. 2004;100(3)(Suppl Spine):307–311.
11. Rajasekaran S, Avadhani A, Parthasarathy S, Shetty AP. Novel technique of reduction of a chronic atlantoaxial rotatory fixation using a temporary transverse transatlantal rod. *Spine J*. 2010;10(10):900–904.
12. Beier AD, Vachhrajani S, Bayerl SH, et al. Rotatory subluxation: experience from the Hospital for Sick Children. *J Neurosurg Pediatr*. 2012;9(2):144–148.
13. Hicazi A, Acaroglu E, Alanay A, et al. Atlantoaxial rotatory fixation-subluxation revisited: a computed tomographic analysis of acute torticollis in pediatric patients. *Spine (Phila Pa 1976)*. 2002;27(24):2771–2775.
14. Missori P, Marruzzo D, Peschillo S, Domenicucci M. Clinical remarks on acute post-traumatic atlanto-axial rotatory subluxation in pediatric-aged patients. *World Neurosurg*. 2014;82(5):e645–e648.
15. Phillips WA, Hensinger RN. The management of rotatory atlanto-axial subluxation in children. *J Bone Joint Surg Am*. 1989;71(5):664–668.
16. Fielding JW, Hawkins RJ. Atlanto-axial rotatory fixation. (Fixed rotatory subluxation of the atlanto-axial joint). *J Bone Joint Surg Am*. 1977;59(1):37–44.
17. Pang D, Li V. Atlantoaxial rotatory fixation: part 2—new diagnostic paradigm and a new classification based on motion analysis using computed tomographic imaging. *Neurosurgery*. 2005;57(5):941–953.

18. Ono K, Yonenobu K, Fuji T, Okada K. Atlantoaxial rotatory fixation. Radiographic study of its mechanism. *Spine (Phila Pa 1976)*. 1985;10(7):602–608.
19. Ishii K, Chiba K, Maruiwa H, et al. Pathognomonic radiological signs for predicting prognosis in patients with chronic atlantoaxial rotatory fixation. *J Neurosurg Spine*. 2006;5(5):385–391.
20. Govender S, Kumar KPS. Staged reduction and stabilisation in chronic atlantoaxial rotatory fixation. *J Bone Joint Surg Br*. 2002;84(5):727–731.
21. Chazono M, Saito S, Liu K, Marumo K. Continuous skull traction followed by closed reduction in chronic pediatric atlantoaxial rotatory fixation. *Acta Neurochir (Wien)*. 2011;153(7):1443–1445.
22. Landi A, Pietrantonio A, Marotta N, et al. Atlantoaxial rotatory dislocation (AARD) in pediatric age: MRI study on conservative treatment with Philadelphia collar—experience of nine consecutive cases. *Eur Spine J*. 2012;21(suppl 1):S94–S99.
23. Pang D, Li V. Atlantoaxial rotatory fixation: part 3—a prospective study of the clinical manifestation, diagnosis, management, and outcome of children with alantoaxial rotatory fixation. *Neurosurgery*. 2005;57(5):954–972.

Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Shimazaki. Acquisition of data: Yamada, Shimazaki, Jimbo, Nakamura, Goto, Mizokami, Iwahashi, Sasaki. Analysis and interpretation of data: Shimazaki. Critically revising the article: Matsubara. Statistical analysis: Shimazaki. Study supervision: Yamada, Sato, Matsubara, Shiba.

Correspondence

Kei Yamada: Kurume University School of Medicine, Kurume, Japan. yamada_kei@kurume-u.ac.jp.