Atlantoaxial Fixation for Treatment of Chiari Formation and Syringomyelia with No Craniovertebral Bone Anomaly: Report of an Experience with 57 Cases

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Abstract *Aim:* In this paper we evaluate the role of atlanto-axial instability in the pathogenesis of Chiari formation type I and the role of atlantoaxial stabilization for treatment of this condition in cases with no obvious bone malformation in the region of the craniovertebral junction.

Materials, Methods and Results: During the period from January 2010 to July 2016, we identified 57 cases of Chiari formation where there was no bone malformation or evidence of craniovertebral junction instability that could be diagnosed on the basis of an abnormal increase in the atlantodental interval on dynamic imaging. Forty-eight of these patients had syringomyelia. The average duration of follow-up was 42 months. There were 30 males and 27 females in the series. The ages of the patients ranged from 4 to 57 years. The Japanese Orthopaedic Association (JOA), visual analogue scale (VAS) and Goel clinical grading systems were used to assess the patients' clinical status. Atlantoaxial instability was diagnosed on the basis of vertical mobility of the odontoid process on dynamic radiographs, facetal malalignment on imaging or direct bone handling during the surgical procedure. Surgical treatment was achieved using atlantoaxial fixation. Foramen magnum decompression or syrinx manipulation was not done. All patients had immediate postoperative and sustained clinical symptomatic recovery. A reduction in the size of the syrinx was observed in ten patients and regression of tonsillar herniation was observed in 12 of 23 cases in which postoperative magnetic resonance imaging (MRI) was possible.

Conclusion: Atlantoaxial instability is the prime factor in the genesis of Chiari formation even when there is no bone abnormality in the craniovertebral junction.

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Introduction

In 2014 we reported our experience with 65 cases of Chiari formation type I treated using atlantoaxial fixation [1]. It was identified that Chiari formation may not be the primary pathology; rather, it may be secondary to atlantoaxial dislocation. Essentially this meant that atlantoaxial stabilization may be the most appropriate treatment for Chiari formation and that foramen magnum decompression is unnecessary. In our earlier publication, there were 46 patients who had basilar invagination and 19 patients who had no bone anomaly at the craniovertebral junction [1]. This paper reports our current experience with 57 cases of Chiari formation with no craniovertebral junction bone anomaly or instability that could be diagnosed on the basis of existing and validated radiological parameters. Apart from our report discussing this pattern of treatment for Chiari formation, there is no other such report in the literature. Considering the potential significance of these observations, our current experience is presented here.

Materials and Methods

During the period from January 2010 to July 2016, 57 cases were identified in which there was Chiari formation in the absence of any bone anomaly in the craniovertebral junction. Five of these patients had previously undergone surgical foramen magnum decompression. This series excluded cases of Chiari formation associated with basilar invagination or bone anomalies and fusions of the region, and cases related to any gross structural brain or spinal cord malformation, hydrocephalus, tumour, infection or any connective tissue disorder.

Results

There were 30 males and 27 females in this series. The ages of the patients ranged from 4 to 57 years, the average being 34.5 years. Table 1 summarizes the clinical profile of the patients. All patients were evaluated on the basis of the Japanese Orthopaedic Association (JOA), [2] visual analogue scale (VAS) [3] and Goel clinical grading systems [1, 4] both before and after surgery. Pyramidal symptoms were present in 73% of patients. Kinaesthetic sensations were affected in 70.7% of patients and spinothalamic dysfunction was identified in 56% of patients (Table 1). Neck pain was a major presenting symptom in 29.26% of patients. Neuropathic pain in the shoulders and hands was present in 34.1% of patients. The duration of symptoms ranged from 1 month to 21 years (mean 48.48 months). All patients were investigated with dynamic computed tomography (CT) scanning (with the head in flexion, extension and neutral positions) and with magnetic resonance imaging (MRI) both before and after surgery. Vertical atlanto-axial dislocation was identified when the odontoid process moved vertically and rostrally on flexion of the head and returned to a normal position on head extension [5]. Such dislocation was identified in ten patients. Atlantoaxial facetal instability was diagnosed according to our recently described classification [6]. For this evaluation, radiological examinations were done with MRI or CT sagittal images passing through the atlantoaxial facets with the head in a neutral position. Type 1 atlantoaxial facetal instability was diagnosed when the facet of the atlas was dislocated anterior to the facet of the axis. Type 2 atlantoaxial facetal instability was diagnosed when the facet of the atlas was dislocated posterior to the facet of the axis. In type 3 atlantoaxial facetal instability, the facets of the atlas and axis were in alignment and atlantoaxial instability was diagnosed only during the operation by being observed during direct manual manipulation of the bones. Type 2 or 3 atlantoaxial instability was labelled as axial or central atlanto-axial instability, as alteration of the atlantodental interval was not a constant or primary feature (as is observed in type 1 atlan-toaxial instability). In 48 patients there was syringomyelia. In nine patients there was external syringomyelia [7], meaning that an excessive amount of cerebrospinal fluid (CSF) was present in the extramedullary space. In 20 cases, both internal and external syringes were present. In 15 cases there was exter-nal syringobulbia, suggesting there was an excessive amount of CSF surrounding the brainstem and cerebellum [7].

All patients underwent atlantoaxial fixation using the techniques described in 1994 and 2004 [8–10]. The aim of surgery in all cases was atlantoaxial stabilization leading to arthrodesis. A bone graft was harvested from the iliac crest, packed into the atlantoaxial joint cavity and placed in the midline, over the (appropriately prepared) host bone of the arch of the atlas and lamina and the spinous process of the axis. Stainless steel plates with monoaxial non-locking-type screws were used in the initial part of the series. After June 2013, the plates and screws used for fixation were made of MRI-compatible titanium material. Foramen magnum decompression was not done in any case.

Clinical improvement was observed in the immediate post-operative phase in all patients. The progress of improvement was sustained at follow-up. The follow-up period ranged from 6 to 78 months, the average being 42 months. All postopera-tive assessments were performed using clinical and radiologi-cal information obtained at least 6 months after surgical treatment. Dynamic plain radiography and CT scanning were used to confirm postoperative arthrodesis. In particular, fusion of the facets and the posterior elements of the atlantoaxial bone were observed. Despite the presence of artefacts related to the use of stainless steel and titanium metal implants, the CT image quality was satisfactory for demonstration of align-ment and bone fusion. Fusion was considered to be successful when the implant demonstrated maintenance of fixation on dynamic radiography and bone fusion was observed in the fac-ets. As titanium implants were used only after June 2013, it was impossible to use MRI to evaluate the long-term status of syringomyelia and Chiari formation. However, definite reduc-tions in the dimensions of the syrinx and reductions in tonsillar herniation were observed within a period ranging from 2 days to 12 months after surgery in 12 of 23 cases in which postop-erative MRI was available for evaluation (Figs. 1 and 2). No patient's clinical symptoms worsened after initial recovery. Tables 2, 3 and 4 depict the extent of clinical recovery at a minimum follow-up of 6 months. Five patients with preopera-tive symptoms related to lower cranial nerve weakness had symptomatic improvement following surgery. Although voice volume and quality were not assessed or evaluated during the preoperative examination, at least three patients reported improvements in their voice volume and quality at follow-up. No patient suffered delayed neurological worsening sufficient to warrant foramen magnum decompressive surgery or needed any kind of reoperation. No patient required re-exploration for failure of implant fixation.

Table 1 Presenting complaints

Presenting complaints	Number of patients
Pyramidal involvement	40
Kinesthetic involvement	41
Spinothalamic involvement	32
Neck pain	17

Discussion

Chiari formation and syringomyelia are frequently associated with bone anomalies at the craniovertebral junction [11, 12]. The issue up for discussion is whether Chiari formation

Fig. 1 Images of a 45-year-old male patient. (**a**) T2-weighted magnetic resonance imaging (MRI) showing Chiari formation and syringomyelia. Note the atrophic cerebellum. (**b**) Mid-sagittal computed tomography (CT) scan showing no evidence of any alteration in the atlantodental interval. (**c**) Sagittal CT scan with the cut passing through the facets. There is type 2 atlantoaxial facetal instability. (**d**) Postoperative mid-sagittal CT scan. (**e**) Postoperative CT scan showing the implant and fixation. (**f**) Postoperative MRI showing a reduction in the size of the syrinx and superior regression of the tonsils

Fig. 1 (continued)

differs in its pathogenesis, and requires different surgical treatment, according to the presence or absence of associated bone anomalies at the craniovertebral junction [13, 14]. We hypothesized that Chiari formation is always secondary to atlantoaxial instability irrespective of whether bone anomalies related to basilar invagination are associated with it [1]. The current report re-emphasizes this concept.

Hans Chiari first identified Chiari formation as a 'pathological' entity in 1887 [15]. Since then there has been considerable speculation regarding its pathogenesis and treatment [16–18]. Foramen magnum decompression has been the most widely accepted method of treatment [8]. When Chiari formation is associated with syringomyelia, tonsillar resection, arachnoid dissection, plugging of the obex, introduction of a shunt into the central spinal canal through the region of the obex, syringoperitoneal/subarachnoid shunts and a host of other treatment forms have been identified and successfully deployed [19–23]. Foramen magnum decompression surgery has ranged from bone decompression to dural decompression, introduction of pericranial grafts and fascia lata grafts for dural reconstruction [19, 20, 22–24], introduction of reverse foramen magnum bone grafts [25] and metal implants, and several such alternatives. In our earlier report, it was commented that there are as many different variations of the surgical procedure for foramen magnum decompression as there are surgeons performing this procedure [1, 23]. Every surgeon is convinced that the methods of foramen magnum decom-

pression used by him or her are optimal and produce the best results.

In 1998 it was identified that when Chiari formation was associated with basilar invagination, the posterior fossa volume was relatively small; accordingly, foramen magnum decompression was the recommended mode of treatment [21]. For the first time in the literature, it was suggested that bone decompression was sufficient and that there was no need to perform dural decompression for Chiari formation, even when there was associated syringomyelia [21]. Some authors have identified that Chiari formation associated with basilar invagination is pathogenetically different from the scenario in which there is no craniovertebral region bone anomaly [26]. Our observations in 2014 suggested that Chiari formation—with or without bone abnormalities at the craniovertebral junction—is secondary to atlantoaxial instability; accordingly, we treated this entity with atlantoaxial stabilization alone [1].

The conventional mode of identification of atlantoaxial instability has been alteration of the atlantodental interval on dynamic imaging. It was identified that atlantoaxial instability can be vertical [5], horizontal–anteroposterior or lateral [27], and central or axial in nature [6]. Identification of such forms of atlantoaxial instability has added a new dimension to our understanding of the subject. On evaluation of the dynamic images, we identified ten cases that had vertical mobile and reducible atlantoaxial dislocation [5]. In all
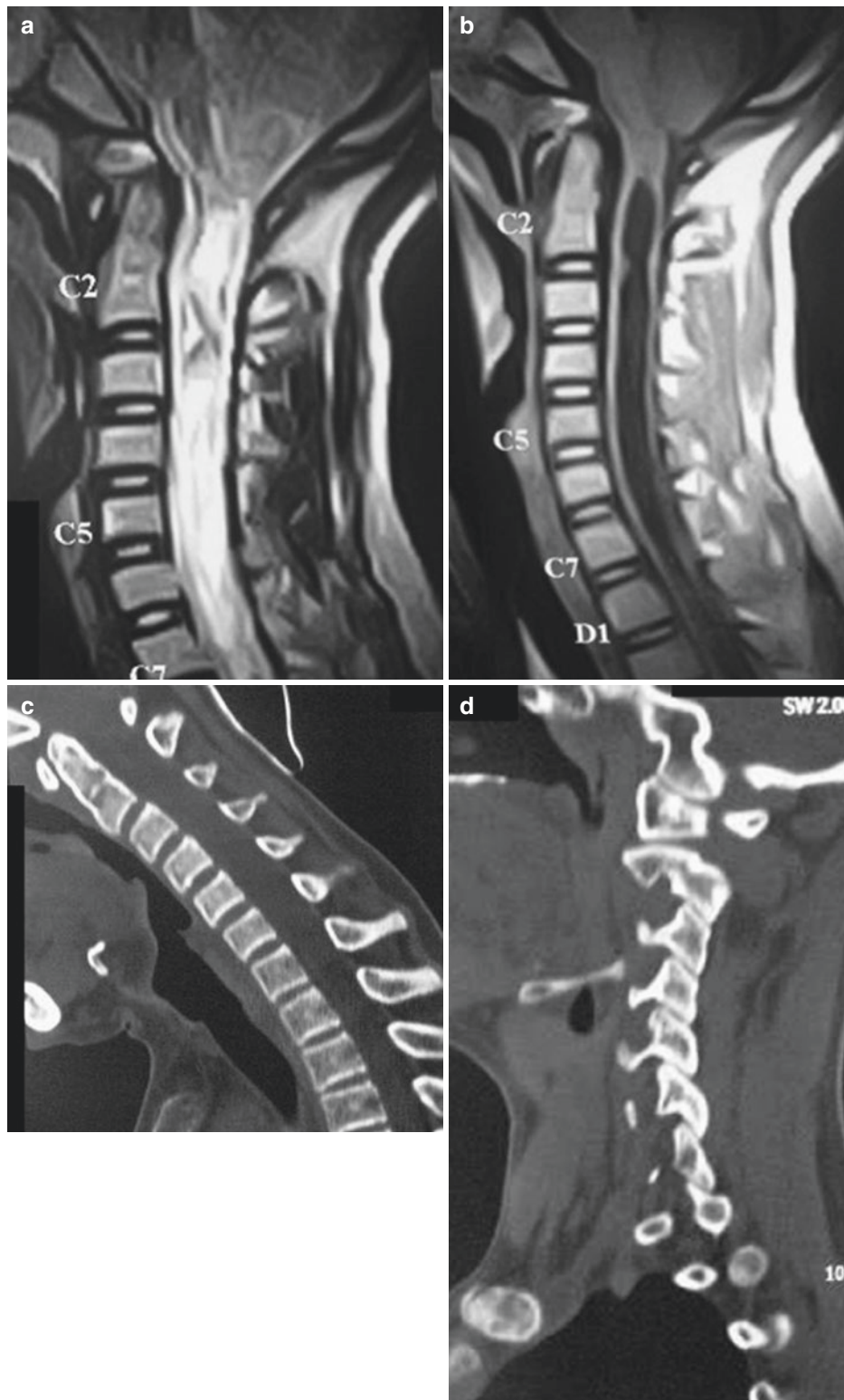
Fig. 2 Images of a 17-year-old male patient. (**a**) T2-weighted magnetic resonance imaging (MRI) showing Chiari formation and syringomyelia. (**b**) T1-weighted MRI showing Chiari formation and the syrinx. (**c**) Mid-sagittal computed tomography (CT) scan with the head in flexion, showing no evidence of any alteration in the atlantodental interval. (**d**) Sagittal CT scan with the cut passing through the facets. There is type 2 atlantoaxial facetal instability. (**e**) Postoperative mid-sagittal CT scan. (**f**) Postoperative CT scan showing the implant and fixation. (**g**) Immediate postoperative (after 18 h of surgery) T1-weighted MRI showing a reduction in the size of the syrinx. (**h**) Immediate postoperative T2-weighted MRI showing a reduction in the size of the syrinx. (**i**) Delayed postoperative T1-weighted MRI (12 months after surgery) showing a reduction in the size of the syrinx and superior regression of the tonsils. (**j**) Delayed postoperative T2-weighted MRI showing a reduction in the size of the syrinx and superior regression of the tonsils
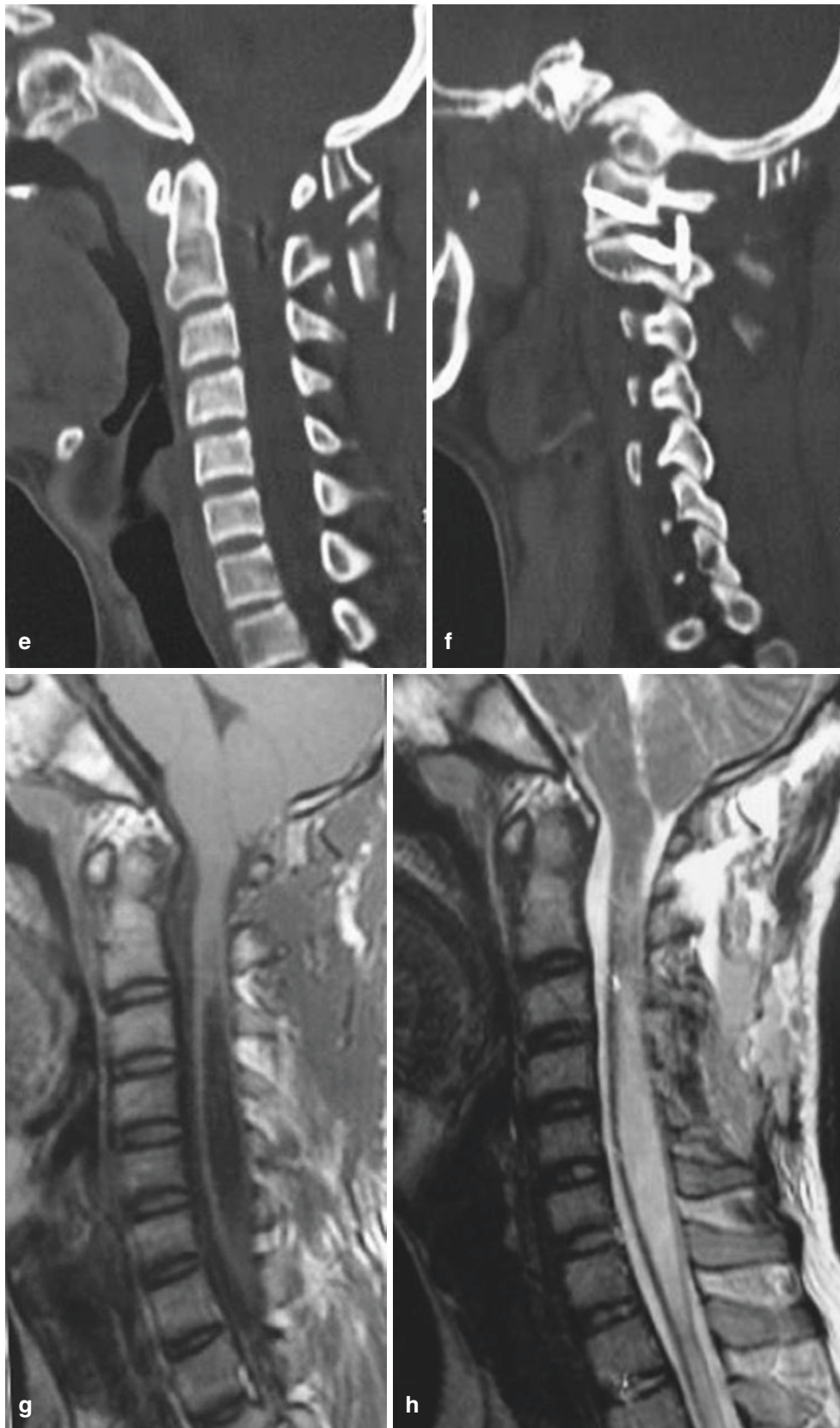
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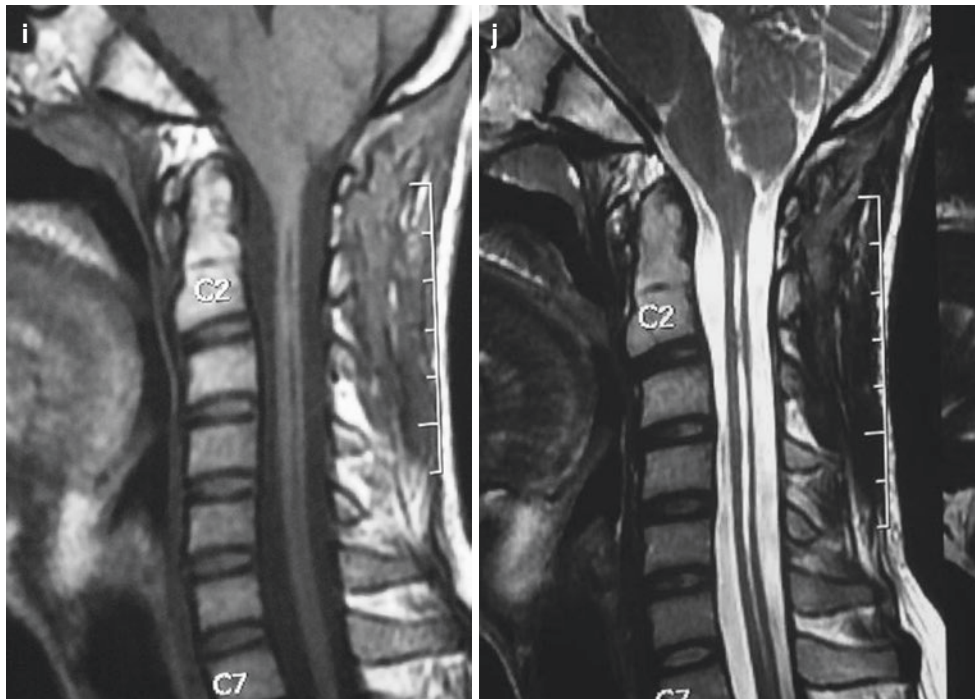
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Table 2 Table showing the pre-operative and post-operative clinical assessment as per JOA scoring system

JOA score	Pre-operative (No. of patients)	Post-operative (No. of patients)
<7	9	2
8–12	12	6
13–15	21	19
16–17	15	30

Table 3 Table showing pre-operative and post-operative clinical assessment as per VAS scoring system

VAS score	Pre-operative	Post-operative
Neck pain	3–7	0–1

Table 4 Distribution as per clinical grading system

Grade	Description	Number of patients (pre-operative)	Number of patients (post-operative)	L = lower cranial nerve deficits
Grade 1	Independent and normally functioning	17	30	
Grade 2	Walks on own but needs support/help to carry out routine household activities	19	22	1
Grade 3	Walks with minimal support and requires help to carry out household activities	12	3	1
Grade 4	Walks with heavy support and unable to carry out household activities	7	2	2
Grade 5	Unable to walk and dependent for all activities	2	–	1

cases, the vertical dislocation was subtle but definite. Identification of such vertical dislocation seems to be an important additional parameter suggesting that there is instability of the region and incompetence of the atlantoaxial joints. Analysis of facetal malalignment on sagittal imaging with the head in a neutral position provides additional information about the stability of the atlantoaxial joint. Although some millimetres of facetal malalignment can be considered to be within the spectrum of normal physiological limits, the association of this malalignment with other abnormalities in the region indicates the presence of instability. Apart from basilar invagination and Chiari formation, central or axial

atlantoaxial instability has been associated with chronic or long-standing spinal diseases such as cervical spondylosis, ossified posterior longitudinal ligament and Hirayama disease [28–31].

Earlier it was speculated that a short neck/torticollis, Klippel–Feil abnormalities, assimilation of the atlas, platybasia and several other musculoskeletal abnormalities were unrelated to embryonic dysgenesis but were secondary and naturally protective events in situations of long-standing or chronic atlantoaxial instability [32]. Altantoaxial stabilization can initiate immediate postoperative reversal of musculoskeletal changes. It was therefore decided that the term 'craniovertebral junction alterations' is preferable to the term 'craniovertebral junction anomalies' [1, 4]. As the various issues that are involved became more clarified, it was realised that just as musculoskeletal alterations are external manifestations, Chiari formation and syringomyelia can be internal and neural manifestations of atlantoaxial instability. Chiari formation is a protective 'airbag' that is placed in position in situations with potential or manifest atlantoaxial instability. It provides a natural cushion-like support for neural structures [33]. Along similar lines, it was speculated that syringomyelia is a protective self-neural alteration that ultimately works in the interests of neural structures and of the human body. Accordingly, it was decided that the term 'Chiari formation' is preferable to the term 'Chiari malformation', considering its functional role and the nature of its pathogenesis [34]. Subtle and long-standing atlantoaxial instability and minimal or no direct neural compression by the odontoid process allow both internal and external reparative processes to gradually develop, progress and mature.

In the patients in our series, anteroposterior measurements of the cervical spinal canal dimensions at the mid-C6 vertebral body level suggested that the spinal canal had increased in girth by 10%. Forty-eight patients had syringomyelia. In nine patients there was an external syrinx [7]. In 20 patients there were both internal and external syringes. Measurement of the total spinal cord girth revealed that the neural girth was reduced by 60% although the spinal canal dimension had increased by 10% (Table 5). The extra space thus created was filled with CSF, irrespective of whether it was present inside, out-

side, or both inside and outside the spinal cord. Evaluation of the posterior fossa content revealed that an excessive amount of CSF was present around the brainstem and cerebellum—an entity identified earlier as external syringobulbia [7]. The fourth ventricle was normal in its shape but larger in its anteroposterior dimensions. These spinal cord and cerebellar features suggested there was no evidence that the posterior fossa contents were tightly or compactly placed. The foramen magnum canal dimension was increased, suggesting that the herniated cerebellar tonsil progressively increased the foramen magnum size to facilitate its positioning.

The primary aims of surgery in all cases were to achieve firm stabilization of the atlantoaxial joint and segmental arthrodesis. It was observed that cases in which direct atlantoaxial fixation was performed in the presence of Chiari formation and syringomyelia had marked venous sinusoidal pooling in the lateral mass gutters, making dissection of the region difficult and fraught with the risk of excessive blood loss. The exact cause of venous pooling in the region was unclear, but chronic and long-standing alterations in the pressure dynamics of the region probably resulted in venous congestion. Moreover, the presence of a lax dura, an atrophic cord and an increase in potential extradural space in the region of the craniovertebral junction increased the scope of venous pooling in the region.

Our observations that 100% of patients recovered from their symptoms to a considerable extent following the surgical procedure of atlantoaxial stabilization alone and that these improvements in neurological symptoms were evident in the immediate postoperative period suggest that atlantoaxial instability plays a role in the pathogenesis of Chiari formation and syringomyelia. Moreover, these improvements in neurological symptoms were observed in patients who had previously undergone failed foramen magnum decompression surgery. Direct observation of the joint status was possible during surgery when the lateral mass plate and screw fixation technique was applied [8–10]. Instability of the atlantoaxial joint was invariably identified during surgery. The primary aims of surgery in all cases were to achieve firm stabilization of the atlantoaxial joint and segmental arthrodesis. During the follow-up period, all patients had firm arthrodesis of the region and there were no instances of implant failure or infection. No foramen decompression was done in any case, although such treatment is currently considered to be the gold standard in the treatment of this entity. From this experience, it is clear that foramen magnum decompression surgery may be unnecessary or even counterproductive. Although the follow-up period in our series was relatively short, our positive results indicate that treatment for Chiari formation should be directed toward atlantoaxial stabilization. The fact that no patient's clinical symptoms worsened after initial improvement during the period of

Table 5 Radiological parameters

Radiological parameter	Present cohort	Normal
Cervical spinal canal dimension at mid-C6 vertebral body level	11–20 mm (mean 15 mm)	11–14 mm (mean 12.78 mm)
Cervical cord girth at mid-C6 vertebral body level	2–7.3 mm (mean 4.4 mm)	6.9–9.4 mm (mean 10.9 mm)

follow-up provides further evidence of the validity of this concept.

This study reiterates our hypothesis that Chiari formation may be nature's own protective mechanism, providing a protective soft cushion, or airbag, to guard the vital spinal cord against potential or manifest compression by the odontoid process and pinching between bones [33]. It appears that Chiari formation is unrelated to reduced posterior cranial fossa volume and is not attributable to a primary or relative increase in cerebellar mass. The presence of syringomyelia, external syringomyelia and external syringobulbia suggest that the neural structures float within an excessive amount of CSF in the spinal canal and posterior fossa. As discussed earlier, the temporary neurological improvement that has been uniformly observed by several authors following foramen magnum decompression could be akin to the effects of deflating a full airbag, and, in the long term, such a form of surgery could be counterproductive. Syringomyelia may be a response of the body that balances the pressures within the neural structures in the interests of the patient [35, 36]. The presence of Chiari formation and syringomyelia suggests chronicity of the process and subtlety of atlantoaxial dislocation. The observations that syringomyelia was reduced and that the tonsils regressed from their herniated position in 52% of the patients in whom MRI was possible—without any form or bone manipulation or dural opening—validate this concept. To consolidate these observations, longer follow-up and multi-institutional experience are mandatory.

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Competing Interests The authors declare that they have no competing interests.

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