Acta Orthopaedica et Traumatologica Turcica 53 (2019) 402–407

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

Clinical and radiological outcomes of conservative treatment for unilateral sagittal split fractures of C1 lateral mass

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A R T I C L E I N F O

Article history:
Received 7 January 2019
Received in revised form
25 May 2019
Accepted 22 August 2019
Available online 11 September 2019

Keywords:
Unilateral sagittal split fracture
C1 lateral mass
Conservative treatment
Transverse atlantal ligament
Outcomes

A B S T R A C T

Objective: The aim of this study was to assess the effect of transverse atlantal ligament (TAL) integrity on clinical and radiological outcomes in patients with unilateral sagittal split fracture (USSF) of the C1 lateral mass (LM).
Methods: Twenty-six consecutive patients (16 men and 10 women; mean age: 52 years (range: 32–69)) with C1 LM USSF were included in this study. Sixteen were TAL injury group (nine of type I injuries and seven of type II injuries according to Dickman's classification) and ten were TAL intact group. All cases were conservatively treated with a rigid brace for TAL intact or by halo-vest stabilization for TAL injury for three months. The mean follow-up was 16 months (range, 12–47 months). The results were compared with radiological assessment of fracture healing, LM displacement and Neck visual analog scale.
Results: At the last follow-up, for TAL intact group, total LM displacement (LMD), unilateral LMD of fracture side, atlanto-dental interval, basion-dental interval, clivus canal angle, and atlanto-occipital joint axis angle were maintained compared to initial presentation. However, for TAL injury group, all radiological parameters were worsened. The worsening of radiological parameters was more severe in type I injury than type II injury except for total LMD and unilateral LMD. Neck visual analog scale significantly decreased and patient's satisfaction was higher in TAL intact group compared to TAL injury group.
Conclusion: Conservative treatment for USSF of C1 LM with TAL injury failed to achieve healing of the fracture, which resulted in lateral displacement of C1 LM. This caused coronal and sagittal malalignment of occipitocervical junction, resulting in unsatisfactory clinical outcomes. Our results suggest that early surgical stabilization should be considered in USSF of C1 LM with TAL injury, especially type I injury. However, conservative treatment may be sufficient for a USSF of the C1 LM with TAL intact.
Level of Evidence: Level III, Therapeutic Study.

Introduction

Recently, the unilateral sagittal split fracture (USSF) of the C1 lateral mass (LM) was recognized as a rare variant of C1 atlas fracture.[1–4] The integrity of the transverse atlantal ligament (TAL) is a critical factor for determining the stability of C1 atlas fracture. According to Bransford et al's study, even without TAL injury, a USSF of the C1 LM seems to be unstable and most of the nonsurgically-treated cases resulted in late cock-robin deformity, requiring a reconstructive surgical procedure of the occipitocervical junction [3]. They also reported satisfactory surgical treatment for a USSF of

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Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

https://doi.org/10.1016/j.aott.2019.08.006
1017-995X/

the C1 LM and recommended early surgery to avoid major surgery that seriously limits movement of the occipitocervical junction.[4] However, due to the rarity of a C1 LM USSF, previous studies are case reports or small number of case series. Definite standard treatment guidelines have not yet been established. Moreover, impact of the TAL injury on treatment outcomes remains controversial in a C1 LM USSF.[5–9] Therefore, we performed the current study to investigate these two issues and to suggest appropriate treatment guidelines for a C1 LM USSF.

Materials and methods

Twenty-six consecutive cases of C1 LM USSF were included from five trauma centers at tertiary university hospitals for retrospective analysis. The fractures associated with other high cervical spine sites, such as C2 and the occiput, were excluded from the study. The mean age was 52 years (range, 32–69 years). Sixteen were male and ten were female. At the time of initial presentation, magnetic resonance imaging (MRI) and computerized tomography (CT) were checked for the evaluation of the injuries in all cases. All cases were conservatively treated with a rigid brace for TAL intact or by halo-vest stabilization for TAL injury for three months at five trauma centers between 1997 and 2017. The mean follow-up was sixteen months (range, 12–47 months).

For the current study, two radiologists evaluated and determined the presence and type of TAL injury on MRI and CT using Dickman's classification and divided into two groups: TAL injury and TAL intact.[10,11] If the results of two judgements were not identical, the third radiologist re-evaluated. As a result, sixteen were included in the TAL injury group and ten were in the TAL intact group. Among the sixteen cases in the TAL injury group, nine were type I ligamentous injury and seven were type II bony avulsion injury.

Three spine surgeons measured radiologic parameters on 2-dimensional sagittal and coronal reconstructed CT scans and MRI of the initial presentation and at the last follow-up. Measured radiologic parameters were as follows: Total LM displacement (LMD) of both sides, unilateral LMD of fracture side, atlanto-dental interval (ADI), basion-dental interval (BDI), clivus canal angle (CCA), and atlanto-occipital joint axis angle (AOJAA) (Fig. 1). Measurements of radiological parameters were done based on methods described in previously published papers.[10–12] The LMD was defined as the distance between the lateral margin of the C2 dens body and the lateral margin of the C1 lateral mass. The distance between the anterior arch of the atlas and the anterior cortex of the dens was defined as the ADI. The distance between the basion and the tip of the dens was defined as the BDI. The CCA is the angle formed by Wackenheim's line and a line constructed along the posterior margin of the dens and axis body. The AOJAA is formed at the intersection of tangents drawn parallel to the atlanto-occipital joints. The averages of three measurements were used as final results. The radiologic outcomes were evaluated by comparing initial presentation and last follow-up in two groups. Clinical outcomes were evaluated by the neck visual analog scale (VAS) and Odom's criteria (Table 1).

Statistical analyses were done by independent sample T-test, paired T- and Chi-square tests. A p value less than 0.05 was considered statistically significant. This multicenter retrospective study was approved by the Institutional Review Board of the corresponding author's university hospital and informed consent was waived.

Results

Demographic data are summarized in Table 2. Age, body mass index (BMI), and follow-up period were not statistically different between three groups by Independent sample T-test. In addition, smoking status was not statistically different between three groups by Chi-square test. The radiological outcomes of conservative treatment for a USSF of the C1 LM are summarized in Table 3, depending on the presence of a TAL injury. At the last follow-up, for TAL intact group, total LMD (1.2 mm vs 1.2 mm, $p = 0.973$), unilateral LMD of fracture side (1.0 mm vs 1.1 mm, $p = 0.828$), ADI (1.5 mm vs 1.3 mm, $p = 0.162$), BDI (4.2 mm vs 3.7 mm, $p = 0.079$), CCA (154.8° vs 151.5°, $p = 0.105$), and AOJAA (105.9° vs 105.3°, $p = 0.800$) were maintained well compared to the initial presentation. However, for TAL injury group, all radiological parameters, such as total LMD (5.9 mm vs 6.7 mm, $p < 0.05$), unilateral LMD of fracture side (4.3 mm vs 4.7 mm, $p < 0.001$), ADI (2.0 mm vs 3.0 mm, $p < 0.001$), BDI (4.4 mm vs 2.6 mm, $p < 0.001$), CCA (155.6° vs 145.2°, $p < 0.001$), and AOJAA (107.8° vs 98.3°, $p < 0.001$), worsened compared to the initial presentation.

For evaluation of the effect of the TAL injury on a USSF of C1 LM, we stratified and compared the data of the TAL injury group into two subgroups, including a type I ligamentous injury versus a type II bony avulsion injury (Table 4). The radiological outcomes of conservative treatment for a USSF of the C1 LM with type I TAL injury and type II TAL injury are summarized in Table 3. At the last follow-up, for type I TAL injury group, all radiological parameters including total LMD (8.0 mm vs 5.1 mm, $p < 0.05$), unilateral LMD of fracture side (6.2 mm vs 4.4 mm, $p < 0.01$), ADI (3.4 mm vs 2.5 mm, $p < 0.05$), CCA (142.2° vs 149.0°, $p < 0.05$), and AOJAA (94.1 vs 103.8°, $p < 0.05$) worsened compared to the initial presentation. However, for type II TAL injury group, ADI (2.0 mm vs 2.5 mm, $p < 0.01$), BDI (4.0 mm vs 2.5 mm, $p < 0.01$), CCA (155.9° vs 149.0°, $p < 0.01$), and AOJAA (110.1° vs 103.8°, $p < 0.001$) worsened compared to initial presentation but total LMD (5.6 mm vs 5.1 mm, $p = 0.139$) and unilateral LMD of fracture side (4.0 mm vs 4.4 mm, $p = 0.444$) were not significantly worsened.

In terms of clinical outcomes, the neck VAS significantly decreased in the TAL intact group (4.7 points vs 2.1 points, $p < 0.001$) but not in the TAL injury group (6.8 points vs 4.7 points, $p = 0.435$). According to Odom's criteria, satisfactory outcomes were higher in the TAL intact group than in the TAL injury group (80% vs. 37.5%; $P < 0.05$) (Table 5).

Discussion

In the current study, we demonstrated that at the last follow-up, for TAL intact group, total LMD) unilateral LMD of fracture side, ADI, BDI, CCA, and AOJAA were maintained compared to initial presentation. However, for TAL injury group, all radiological parameters were worsened. The worsening of radiological parameters was more severe in type I injury than type II injury except for total LMD and unilateral LMD of fracture side. Neck VAS significantly decreased and patient's satisfaction was higher in TAL intact group compared to TAL injury group. Based on current results, we suggest that early surgical stabilization should be considered as choice of treatment for USSF of C1 LM with TAL injury, especially type I injury. However, conservative treatment may be sufficient for a USSF of the C1 LM with TAL intact.

Since the C1 burst fracture was first described as Jefferson's fracture in 1927, several fracture classifications have been introduced to describe and classify various fracture patterns of C1 burst fractures.[13–16] But, there have been no classifications that accurately reflect and categorize the various aspects of C1 burst fractures. The presence of a TAL injury is known to be the important factor in determining the stability of C1 burst fracture and deciding upon the appropriate treatment strategy.[17,18] If there is no TAL injury, it is regarded as a stable fracture and conservative treatment is performed. But, a fracture with a TAL injury is regarded as an

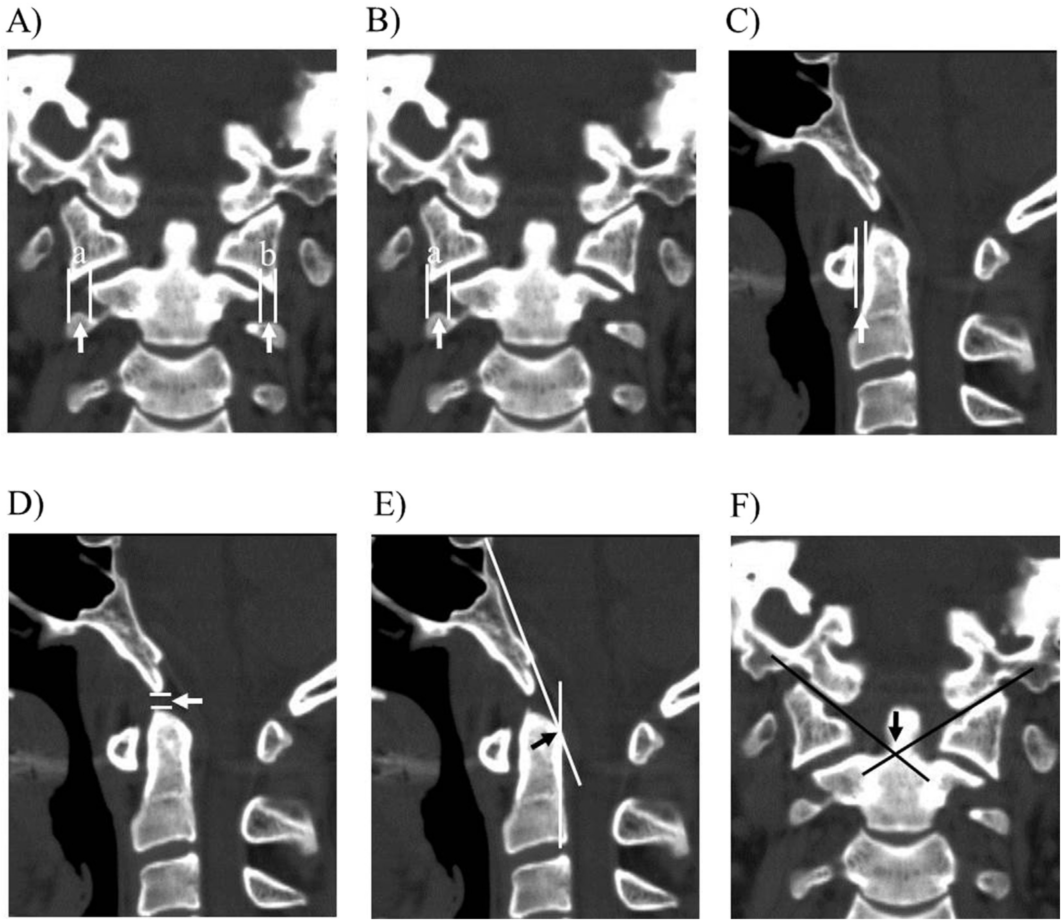

Fig. 1. Measurements of radiological parameters. **A)** Total lateral mass displacement **B)** Fracture side lateral mass displacement **C)** Atlanto-dental interval **D)** Basion-dental interval) **E)** Clivus canal angle **F)** Atlanto-occipital joint axis angle. Measurements of radiological parameters were performed based on methods described by previously published papers [11–13].

Table 1
Odom's criteria.

Grading	Definition
Excellent	All preoperative symptoms and abnormal findings improved
Good	Minimal persistence of preoperative symptoms. Abnormal findings improved
Fair	Definite relief of some preoperative symptoms. Other symptoms slightly improved
Poor	Symptoms and signs unchanged or worse

unstable fracture and surgical treatment is performed. According to Dickman's study, there are two subtypes of TAL injury.[17,18] The type I TAL injury is an inter-substance ligament injury and a type II TAL injury is a bony avulsion injury. The type II TAL injury is more likely to heal more effectively with conservative treatment than the type I TAL injury. The results of our study were consistent with the above evidence presented in previous studies of C1 burst fractures.[19–23] Based on these results, we therefore suggest that early surgical

Table 2
Demographic data of unilateral sagittal split fractures with TAL intact, type I TAL injury, and type II TAL injury groups.

	TAL Intact (N = 10)	Type I TAL Injury (N = 9)	Type II TAL Injury (N = 7)
Age (Years)*	51.7 ± 9.2	51.8 ± 10.3	52.6 ± 12.8
Sex (Male: Female)	5: 5	6: 3	5: 2
BMI**	23.7 ± 3.1	24.8 ± 2.6	23.9 ± 2.0
Smoking (Yes: No)##	3: 7	2: 7	2: 5
Injury mechanism			
Fall down	7	4	7
Traffic accident	2	3	1
Diving	1	0	0
Neurologic status			
ASIA grade	E: 10	E: 9	E: 7
Frankel grade	E: 10	E: 9	E: 7
Follow-up (months)#	16.6 ± 3.7	16.8 ± 11.5	15.4 ± 3.4

TAL = Transverse Atlantal Ligament; BMI = Body Mass Index; ASIA = American Spinal Injury Association.
Age*, BMI**, and follow-up period# were not statistically different between three groups by Independent sample T-test. Smoking status## was not statistically different between three groups by Chi-square test.

Table 3
Comparison of radiological treatment outcomes of unilateral sagittal split fractures of C1 lateral mass with TAL intact versus TAL injury.

	TAL Intact (N = 10)			TAL Injury (N = 16)		
	Initial	Last follow-up	P value	Initial	Last follow-up	P value
Total LMD (mm)	1.2 ± 2.0	1.2 ± 1.1	P = 0.973	5.9 ± 2.0	6.7 ± 2.8	P < 0.05
Unilateral LMD (mm)	1.0 ± 1.1	1.0 ± 1.1	P = 0.828	4.3 ± 1.2	4.7 ± 1.8	P < 0.001
ADI (mm)	1.5 ± 0.4	1.3 ± 0.4	P = 0.162	2.0 ± 0.9	3.0 ± 1.7	P < 0.001
BDI (mm)	4.2 ± 1.4	3.7 ± 1.4	P = 0.079	4.4 ± 1.8	2.6 ± 1.7	P < 0.001
CCA (degree)	154.9 ± 9.4	151.5 ± 6.3	P = 0.105	155.6 ± 7.1	145.2 ± 8.2	P < 0.001
AOJAA (degree)	105.9 ± 14.0	105.3 ± 14.1	P = 0.824	107.8 ± 8.7	98.3 ± 10.5	P < 0.001

LMD = Lateral mass displacement; Fx = Fracture; ADI = Atlanto-dental interval; BDI = Basion-dental interval; CCA = Clivus canal angle; AOJAA = Atlanto-occipital joint axis angle; P-value was calculated by paired t-test.

Table 4
Comparison of radiological treatment outcomes of unilateral sagittal split fractures of C1 lateral mass with type I TAL injury versus type II TAL injury.

	Type I TAL Injury (N = 9)			Type II TAL Injury (N = 7)		
	Initial	Last follow-up	P-value	Initial	Last follow-up	P-value
Total LMD (mm)	6.2 ± 2.0	8.0 ± 2.7	P < 0.05	5.6 ± 2.1	5.1 ± 2.0	P = 0.139
Unilateral LMD (mm)	4.6 ± 1.1	6.2 ± 1.2	P < 0.001	4.0 ± 1.5	4.4 ± 1.1	P = 0.444
ADI (mm)	2.0 ± 0.9	3.4 ± 0.9	P < 0.001	2.0 ± 0.9	2.5 ± 1.8	P < 0.01
BDI (mm)	4.8 ± 1.9	2.3 ± 1.6	P < 0.001	4.0 ± 1.6	2.5 ± 1.8	P < 0.01
CCA (degree)	155.4 ± 6.1	142.2 ± 8.3	P < 0.001	155.9 ± 8.7	149.0 ± 6.6	P < 0.01
AOJAA (degree)	105.8 ± 7.8	94.1 ± 8.8	P < 0.001	110.1 ± 9.6	103.8 ± 10.4	P < 0.001

TAL = Transverse atlantal ligament; LMD = Lateral mass displacement; Fx = Fracture; ADI = Atlanto-dental interval; BDI = Basion-dental interval; CCA = Clivus canal angle; AOJAA = Atlanto-occipital joint axis angle; P-value was calculated by paired t-test.

treatment should be considered as fit first choice treatment for a USSF of the C1 LM, especially type I TAL injury. However, conservative treatment may be sufficient for a USSF of the C1 LM with TAL intact.

Fracture of the C1 LM has been defined in various fracture classification systems. However, a USSF of the C1 LM does not match any of the fracture classification systems and is considered to be a variant of the C1 atlas fracture.[13–16] Recently, Bransford et al reported that a USSF of the C1 LM is considered to be unstable because of the lateral displacement of the fractured LM, despite TAL integrity.[3] The LMD results in an incongruence of the occipito-cervical junction including C0-C1 and C1-C2 joints, causing late cock-robin deformity. And all three cases were treated by reconstructive surgical procedures finally resulting in a large loss of the occipito-cervical junction. After this study, they published another study to report satisfactory surgical treatment for a USSF of the C1 LM.[4] The results of Bransford et al's two papers are inconsistent with those of our current study.

Table 5
Comparison of clinical treatment outcomes of unilateral sagittal split fractures of C1 lateral mass with TAL intact versus TAL injury.

	TAL Intact (N = 10)	TAL Injury (N = 16)
A) Neck visual analog scale (VAS)		
Initial	4.7 ± 1.3	6.8 ± 1.2
Last follow-up	2.1 ± 0.3	4.7 ± 2.7
P value	P < 0.001	P = 0.435
B) Odom's criteria		
Excellent	3	1
Good	5	5
Fair	2	8
Poor	0	2

P value is determined by Paired T-test.
*P value is less than 0.05 by Chi-square test.

As we can see in Fig. 2, the fracture site of the USSF of the C1 LM with TAL intact was well healed and/or maintained by conservative treatment. Therefore, the alignment of the occipito-cervical junction, especially C0-C1 and C1-C2 joints, was well maintained while lateral displacement of C1 LM did not occur. This resulted in satisfactory clinical outcomes. In our cases of a USSF of the C1 LM with type II TAL injury, conservative treatment resulted in deterioration of some radiologic parameters during the initial healing period of fracture sites but eventually healed (Fig. 3). On the contrary, in our cases of a USSF of the C1 LM with type I TAL injury, the ligament injuries were not healed with rigid cervical brace or halo-vest stabilization so as to cause deterioration of lateral displacement of the C1 LM (Fig. 4). So, radiological and clinical outcomes were poor.

Just as Bransford et al described, a limitation of their study was a small number of case series, containing only three patients.[3,4] In addition, they did not have follow-up CT imaging. However, we understand that the drawbacks of their two studies are inevitable because a USSF of the C1 LM is so rare. Therefore, to overcome this shortcoming, we conducted a retrospective multicenter study to collect more cases from tertiary trauma centers at university hospitals. To our knowledge, our study is the largest to investigate many cases of a USSF of the C1 LM, more than any others in published papers. Another strength of our study is that we had follow-up CT imaging in all cases. Despite the fundamental limitation of a retrospective multicenter study, these strengths of our study suggest that our results may be sufficiently appealing to other spine surgeons and practitioners as well. The follow-up period of our study was not prolonged (average of 16 months). There was no case in which the operation was not performed in cases of a USSF of the C1 LM with TAL injury with unsatisfactory outcomes in conservative treatment. Therefore, a longer follow-up is necessary to answer the question of whether surgical treatment is needed as reported in other published papers such as the Bransford et al study.

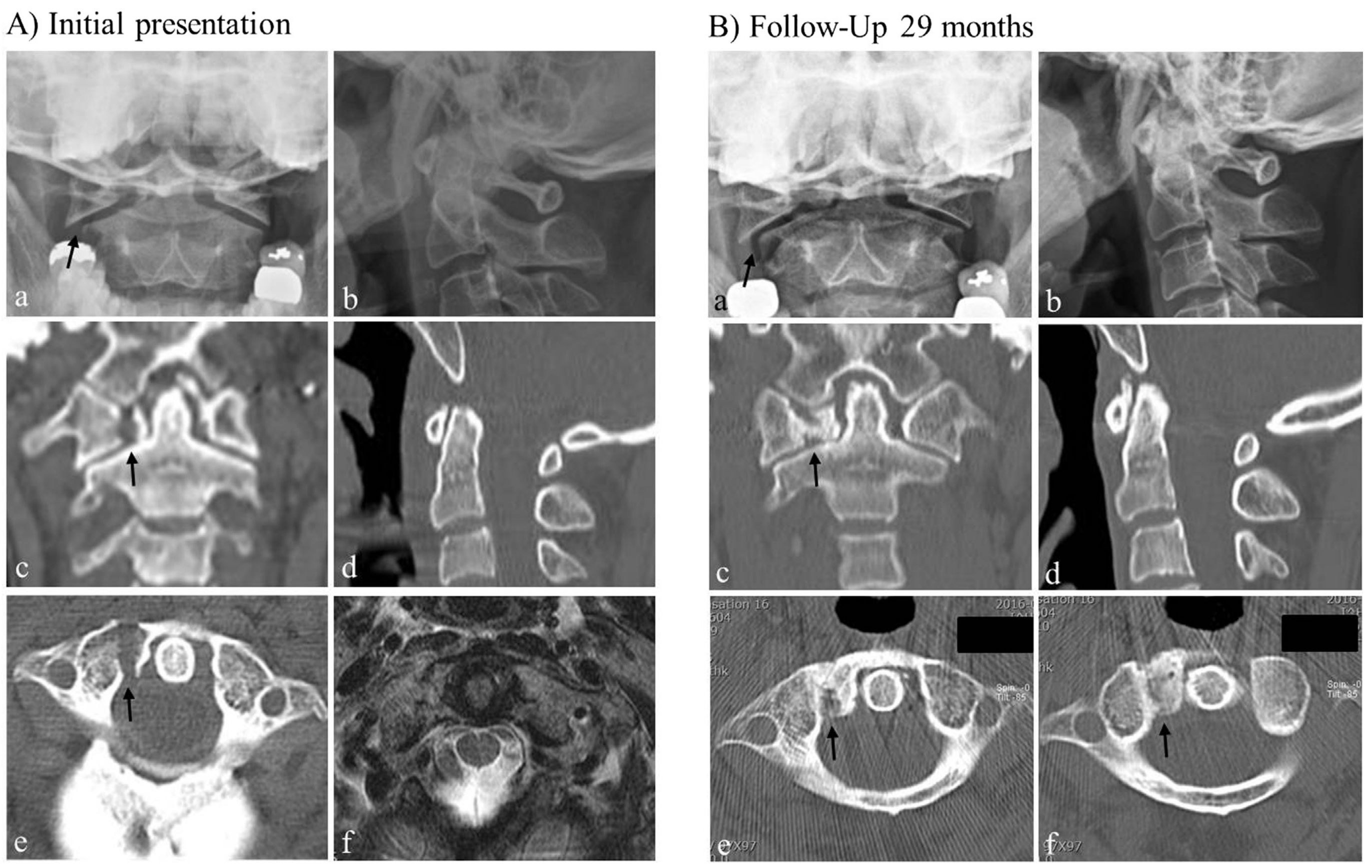

Fig. 2. **A)** Initial presentation of unilateral sagittal split fracture with intact transverse atlantal ligament. Plain radiographs **(a and b)**, CT scans **(c, d and e)**, and MRI **(f)** showing unilateral sagittal split fracture of C1 lateral mass and intact transverse atlantal ligament. **B)** At follow-up 29 months after conservative treatment. Plain radiographs **(a and b)**, CT scans **(c, d, e, and f)** showing progression of healing of the unilateral sagittal split fracture of C1 lateral mass.

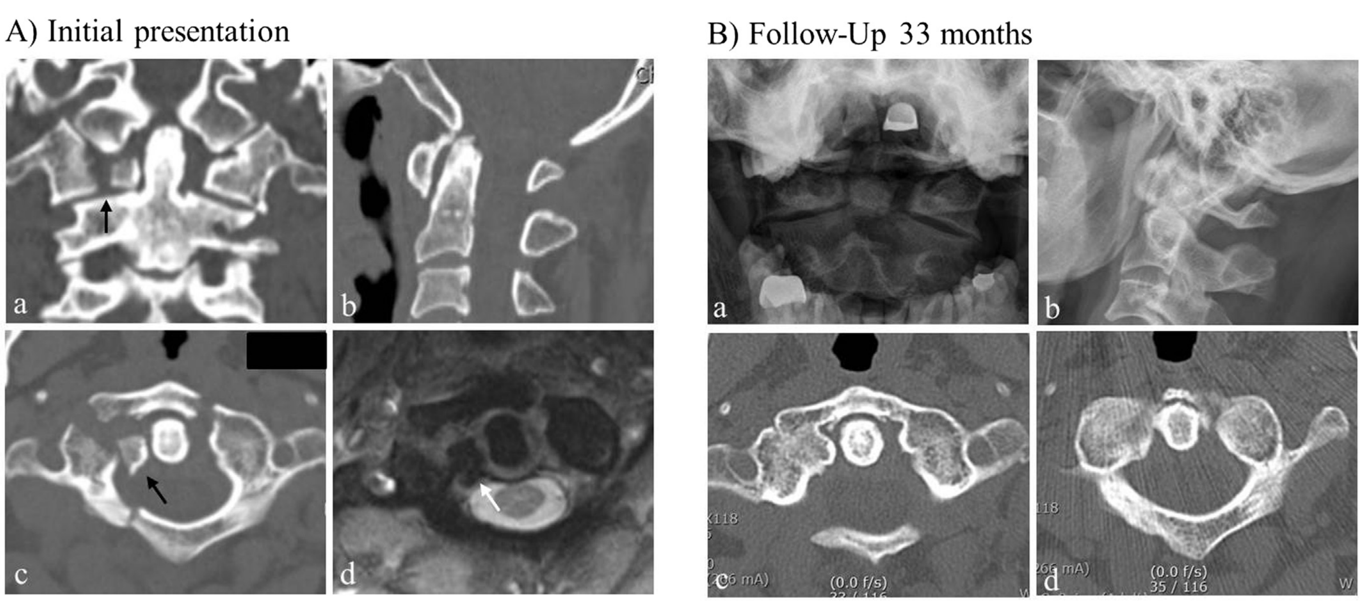

Fig. 3. **A)** Initial presentation of the unilateral sagittal split fracture with transverse atlantal ligament injury type II. CT scans **(a, b, and c)** and MRI **(d)** showing the unilateral sagittal split fracture of the C1 lateral mass with transverse atlantal ligament injury type II. **B)** At follow-up 33 months after conservative treatment. Plain radiographs **(a and b)** and CT scans **(c and d)** showing well healing status of the unilateral sagittal split fracture of the C1 lateral mass.

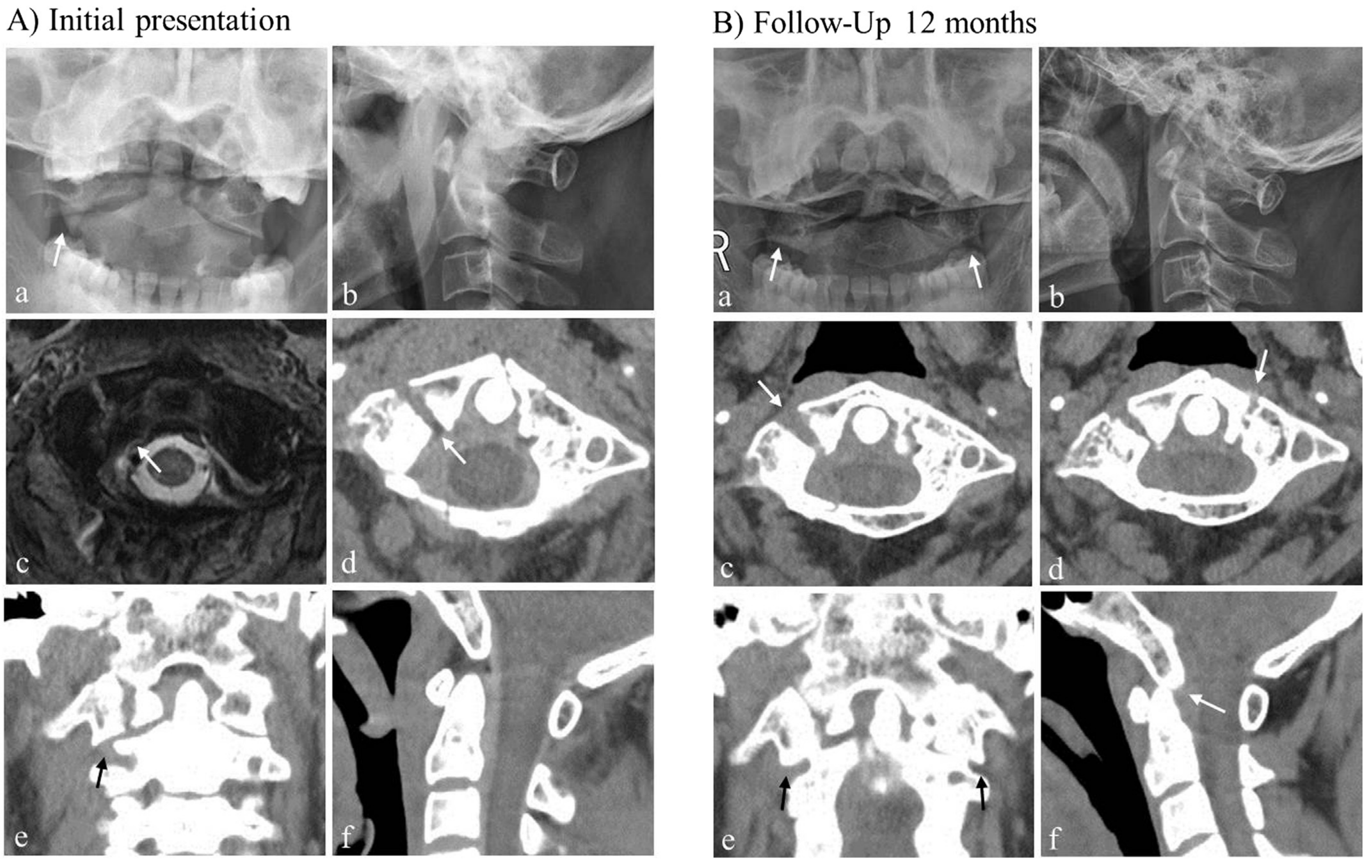

Fig. 4. **A**) Initial presentation of the unilateral sagittal split fracture with transverse atlantal ligament injury type I. Plain radiographs (**a and b**), MRI (**c**) and CT scans (**d, e, and f**) showing the unilateral sagittal split fracture of the C1 lateral mass with transverse atlantal ligament injury type I and associated anterior and posterior arch fractures. **B**) At follow-up 12 months after conservative treatment. Plain radiographs (**a and b**), CT scans (**c, d, e, and f**) showing nonunion of the unilateral sagittal split fracture of the C1 lateral mass and malalignment to craniovertebral junction.

In conclusion, our results suggest that early surgical stabilization should be considered as the first choice treatment for a USSF of the C1 LM with type I TAL injury. However, conservative treatment may be sufficient for a USSF of the C1 LM with TAL intact.

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