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Incidence and risk factors of anterior arch fracture of the atlas following C1 laminectomy without fusion

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Study Design.A retrospective multi-center study

Objective. To identify the incidence of anterior arch fracture (AAF) of the atlas following C1 (first cervical vertebra) laminectomy without fusion, and its risk factors.

Summary of Background Data.C1 laminectomy without fusion is awidely accepted surgicalprocedureperformed to decompress the spinal cord that is compromised at the C1/C2 level, but without instability. Several case series have reported spontaneous AAF following this procedure. However, theincidenceof post-laminectomy AAF and its risk factors have not been studied.

Methods. This retrospective study included patients who underwent C1 laminectomy without fusion in any ofthe four participating institutions between April 2002 and March 2016. The incidence of AAF following C1 laminectomy was determined, and the included patients were grouped into those who developed AAF (AAF group) and those who did not (non-AAF group). Patient demographics and radiographic parameters including subaxial cervical balance on X-ray (C2-7 sagittal vertical axis, C2-7 lordosis, C2-7 coronal cobb angle, and T1-slope), and morphology of the atlas on computed tomography (CT) scan were compared between the AAF and non-AAF groups.

Results. Seventy patients who underwent C1 laminectomy without fusion were included in the study. The incidence of AAF was 14.2% (10/70). Multivariate analysis revealed that a large inferior facet angle (IFA, defined as the coronal inclination angle of the C1/2 facet as measured on CT) and the presence of subaxial ankylosis (bony ankylosis below C2 on CT) were independent risk factors for AAF. There were no significant differences in the subaxial cervical balance as measured on X-ray between the AAF and non-AAF groups ($p > 0.05$).

Conclusions. The incidence of AAF after C1 laminectomy without fusion is not uncommon. Preoperative assessment using CT may identify patients at high risk of AAF.

Key Words

Anterior arch fracture, atlas, C1 laminectomy, fusion, spontaneous, ankylosis, facet angle, cervical balance, C2, retroodontoid pseudotumor

Level of Evidence:4

Introduction

Laminectomy of the first vertebra (C1) without fusion (CLWF) is asurgical interventionperformed for indications that include Arnold-Chiari malformation (combinedwith suboccipital craniotomy), intracanal tumor at the level of C1, and retroodontoidpseudotumor without instability.[1-3] It is a widely accepted minimally-invasive surgical procedure to decompress the spinal cord compromised at the level of C1. This is especially true in the Japanese population wherein ossification ofthe posterior longitudinal ligament (OPLL)is a common cause of spinal cord compromise.[4] Until recently, the complications with this popular procedure were considered rare, although there is no data supporting the same.O'Shaughnessy et al first reported two cases of anterior arch fracture (AAF) of the atlas following CLWF for Arnold-Chiari malformation.[5] Interestingly, AAF which normally occurs secondary tohigh-energy trauma and is classified as a type of Jefferson fracture, occurred spontaneously in these two cases. The authors hypothesized that the iatrogenic interruption of the posterior arch integrityof C1 contributesto an increased risk of spontaneous AAF. A recent case series that analyzed the fractures from a biomechanical standpoint by using finite element analyses described the concentration of stress distributionin the anterior arch of C1 following CLWF, thus explaining the predilection for spontaneous AAF.[6] Although these small case series highlight the need to recognize the possibility of AAF, there are no published reports describing its incidence and risk factors. This information, if available, may support surgeons in appropriate case selection for CLWF and reduce the incidence of complications.

Thus, the aim of the present study was to determine the incidence and risk factors of AAF after CLWF.

Materials and Methods

Patient population and incidence of anterior arch fracture

This study was approved by the institutional ethics board. A retrospective review of the medical records of patients who underwent CLWF in any of the four participating institutions between April 2002 and March 2016 was performed. A total of 73patients underwent CLWF during this period and satisfied the following inclusion criteria: (1) absence of congenital nonunion of the anterior arch and (2) follow-up period greater than eight months. The diagnosis of AAF was made based on computed tomography (CT) scans that were usually performed when the patients experienced new-onset neck pain following CLWF. Patients who did not develop new-onset neck pain were included in the non-AAF group based on CT scan findings if available or cervical X-ray findings if a routine CT scan was not performed.

Surgical procedure

The surgical procedure (of C1 laminectomy) performed by the different surgeons of the four participating institutions was similar with the exception of the decision on whether to dissect the muscles insertinginto the C2 spinous process: following a longitudinal skinincision, the nuchal ligament was divided in the middle to identify the C2 spinous process. While some surgeons divided the C2 spinous processlongitudinally by using a bone saw, preserving the muscles insertinginto the C2 spinous process (rectus capitis posterior major andobliquus capitis inferior muscle), and reconstructed the spinous process following the resection of the C1 posteriorarch, other surgeons dissected the muscles, either totally or partially. The C1 posterior arch resection included a segment approximately 20 mm in size followingthe dissection of the muscles inserting into itssurface (rectus capitis posterior minor muscle). Decompression of the narrow levels (laminectomy or laminoplasty) was also performed in cases of co-existing subaxial canal narrowing.

Patient Data

The baseline demographics including, age, sex, preoperative diagnosis, body mass index (BMI), and comorbidities were recorded. The following radiographic parameters that may have affected the kinematics of the cervical spine were evaluatedbased on preoperative and postoperative images (postoperative data were collected from the AAF group at the time of detecting the fracture, and in the non-AAF group, at their last follow-up visit): C2-7 lordosis (CL, the cobb angle between the lines parallel to the inferior endplate of C2 and C7 on lateral view), C2-7 sagittal vertical axis (SVA, the distance between a plumb line from thecenter of C2 and the posterosuperior corner of the C7 vertebral body), T1-slope (TS, the angle between the superior endplate of T1 and ahorizontal reference line), C2-7 coronal cobb angle (the cobb angle between the lines parallel to the inferior endplates of C2 and C7 on anterior-posterior view), and C1/2 instability (considered positive if the atlanto-dens interval was > 4 mm on dynamic X-ray). These parameters were measured on X-ray images according to a previous study (Fig. 1A).[7] The parameters evaluated on CT scans included the anterior arch length (the distance between the inside edges of the lateral mass on axial view), laminectomy length (the distance between the edges of the excised posterior arch), superior and inferior facet angles of C1 (SFA/IFA, the angle subtended by the tangential line of the superior/inferior articular process and the parallel line of the inferior endplate of C2 on coronal view: an average value calculated from 3 consecutive coronal slices), facet tropism (tropism was defined as positive if [the right SFA/IFA – left SFA/IFA] was > 7° as described in a previous study[8]), and the presence of subaxialankylosis (ankylosis was defined as positive if any bony bridge formation was observed between C2 and C3 on sagittal or coronal view)

(Fig. 1B). The radiographic and baseline demographic parameters were compared between the AAF and non-AAF groups.

Statistical analyses

Data are presented as mean ± standard deviation unless specified otherwise. Chi-square test and Student t test were used for categorical variables and continuous variables, respectively. To identify risk factors for AAF, an initial univariate analysis was performed. Multivariate logistic regression analysis was performed by using the variables with $p < 0.10$on univariate analysis. Areceiver operating characteristic (ROC) curve was created to determine the cutoff values of IFA as a predictor of AAF after CLWF. The accuracy of diagnosis wasevaluated by calculating the area under the curve (AUC).All the radiographic parameters were measured by two board-certified orthopedic surgeons. Inter-observer reliability was calculated according tointraclasscorrelation coefficients (ICCs) and classified as poor (0–0.39), moderate (0.4–0.74), or excellent(0.75–1).JMP version 11 (SAS, Cary, NC) was used for all analyses. The statistical significance was set at $p < 0.05$.

Results

Of the 73 patients who underwent CLWF in the study period, three patients were excluded owing to congenital nonunion of the anterior arch or a short follow-up period (< 8 months). A total of 70 patients were included for analyses.

Incidence of anterior arch fracture

The incidence of spontaneous AAF following CLWF was 14.2% (10/70) over a mean follow-up period of 8.1±8.0 months. Nine out of these 10 patients were diagnosed as having AAF on CT scans performed for complaints of sudden-onset neck pain, while one was incidentally diagnosed on a routine follow-up CT scan. Salvage fusion surgery was required only in two patients who had intractable neck pain. All other patients reported improvement in symptoms following conservative treatment with neck collar, although there was nonunion of the fracture site. The clinical details of the AAF cases are presented in Table 1.

Baseline demographics

As shown in Table 2, the AAF and non-AAF groups were similar in their baseline demographics except for statistically significant differences in their sex ($p = 0.01$) and follow-up period ($p = 0.03$). Preoperative diagnoses included ossification of posterior longitudinal ligament (OPLL), atlantoaxial subluxation (AAS), retroodontoidpseudotumor, degenerative cervical spondylotic myelopathy (CSM), and spinal cord tumor. Rheumatoid arthritis (RA), athetoid cerebral palsy (CP), hemodialysis, and prostatic carcinoma were identified as comorbidities with the potential to affect bone quality or cervical kinematics; however, these characteristics were similar between the two groups. Similarly, there was no intergroup difference in the presence of simultaneoussubaxial decompression during the surgical procedures.

Radiographic parameters

The inter-observer agreement was classified as excellent (ICC=0.78). Twenty-five out of 60 non-AAF patients were excluded from radiographical analyses owing to the lack of adequate CT data.Table 3 shows the comparative analysis of the radiographic parameters. The subaxial balance on X-ray (CL, C2-7SVA, C2-7 coronal cobb, and TS) was not significantly different between the two groups, in both the preoperative and postoperative periods. Among the CT variables, IFA, facet tropism of IFA, and the presence of subaxialankylosis were associated with AAF on univariate analysis $p < 0.10$. On multivariate logistic regressionanalyses, IFA (Odds ratio [OR] per unit (°), 2.5;$p <0.01$) and the presence of subaxialankylosis (OR, 26.3;p = 0.02) were identified as significant risk factors for AAF. The ROC curve for IFA as a predictor for AAF had an AUC of 0.931 (Fig. 2), indicating high accuracy. A cutoff value of IFA of 23.0°demonstrated a sensitivity of 90% and a specificity of 86.2%. Sex was not included for multivariate analysis as its clinical relevance was considered to be low.

Representative cases

An86-year-old male with retroodontoidpseudotumordeveloped AAF after CLWF. The preoperative CT scan showed presence of subaxialankylosis (C2-4) (Fig. 3a) and IFA (20.5°) was less than the cutoff value (Fig. 3b). He complained of sudden-onset neck pain 6 months following his surgery and was found to have an AAF on CT scan(Fig. 3c). A58-year-old male with CSM demonstrated the absence of subaxialankylosis(Fig. 4a), but the IFA (35.8°) was large (Fig. 4b). He presented with severe neck pain 15 months following CLWF and an AAF was confirmed on CT scan (Fig. 4c).

Discussion

The present study is the largest case series of spontaneous AAF following CLWF, and the first identifying its incidence and radiographic risk factors. O'Shaughnessy et al, in 2004, first reported two cases of AAF after C1 laminectomy for Chiari malformation type 1. Subsequently, five case series have been published during the past five years, indicating increasing interest in the pathology of this condition among spine surgeons (Table 4).[6,9-12]

C1 laminectomy has been in practice for years, for a variety of indications including Chiari type 1, RST, CSM, and OPLL, with minimal concerns regarding its complications. Therefore, the incidence of 14.2% as found in our study may be higher than expected. This high incidence warrants the need for preoperative assessment of risk for AAF among patients being considered for CLWF.

A recent finite element analysis attempted to explain the possible mechanism of AAF following C1 laminectomy.[6] In the report, the authors demonstrated an abnormal concentration of von Mises stress in the anterior arch in the presence of a defective posterior arch, which could result in spontaneous AAF. However, since all the patients who undergo C1 laminectomy do not develop AAF, other possible risk factors are likely to contribute to the risk of AAF.

The present study focused on radiographic parameters, especially the difference in the morphology of the C1 facets. In a cadaveric study of Jefferson fractures, Gebauer et al described that the axial compressive forces on the head are transmitted through the horizontally inclined facets of the atlas in a lateral direction.[13] This transmission contributes to burst fracture of the atlas.Therefore, it is likely that a greater coronal inclination of the facet may result in a greater quantum of laterally directed force, increasing the possibility of Jefferson fracture. Based on this theory, we hypothesized that in the presence of a posterior arch defect, both the SFA and the IFA may contribute to the risk of AAF after CLWF. However, our data identified IFA alone as a risk factor. It ispossible that the AAF associated with a posterior defect is a stress fracture caused by repetitive low-level stress and is unlike the Jefferson fracture, which is a burst fracture resulting from high-energy trauma. A large IFA, especially >23.0° as indicated by the ROC curve, would result in greater flexural stress on the anterior arch than the normal IFA. This is becausea larger IFA may result ingreater displacement of the inferior articular process of the atlas, and thus, a greater chance of a stress fracture of the atlas (Fig. 5). It is important to recognize that this hypothesis is based on the fact that the C1/2 facet may allow the joint to slip tangentially, while the atlanto-occipital joint, which has less mobility due to it being a condylar joint, may resist the same. Similarly, the biomechanical theory may also explain the incidence of AAF associated with congenital cleft of posterior arch.[14-16]Allam et al suggested that a congenital posterior arch defect due to failure of fusion of the posterior synchondrosis may result in a subclinical AAF in a small percentage of the population, subsequently mimicking the bipartite atlas.[11]We observed that in almost all the cases of AAF in our study, bone union of the fracture site was obtained only with salvage surgical treatment. Therefore, unless compared with the preoperative CT data, the nonunion site of AAF may be misinterpreted as a congenital defect.

The role of subaxialankylosis, another risk factor identified in our study,in contributing to the risk of AAF, may be explained by the theory of adjacent segment disease. It is well known that arthrodesis results in increased stress in the motion segments adjacent to the fusion level.This leads to overloading of the adjacent segments, especially the intervertebral disc.[17,18]Since the C1/2 joint lacks an intervertebral disc, the increased stress may be directly transmitted to the anterior arch through the C1/2 inferior facet in conditions wherein the posterior arch is defective, thus contributing to the risk of AAF.The sagittal and coronal balances of the subaxial cervical spine were not associated with an increased risk of AAF. It is possible that the incidence of AAF is related to the focal kinematics of the cervical region and not to factors affecting the cervical balance.

Male sex may be a possible risk factor for AAF as demonstrated on univariate analysis, although we excluded it in the logistic regression model as we considered its clinical relevance to be relatively low. Fujimori et al demonstrated that the prevalence of ossification of the anterior longitudinal ligament (OALL) and OPLL were higher in men than in women, indicating that men may have a higher tendency for subaxialankylosis than women.[19]

The fact that 8/10 of the patients who developed AAF were successfully treated with bracing alone indicates that the cost-benefit outcome,of preoperative CT assessment for all patients undergoing CLWF,appears minimal. Nevertheless, it is unlikely for fusion of the AAF to occur in the absence of salvage fusion surgery, as seen in our case series. Although conservative treatment allows neck pain to subside, the incongruity of the C1/2 facet secondary to non-union may lead to secondary osteoarthritis over time. Mandatory pre-operative CT assessment of all patients, and the avoidance of CLWF in cases found to be at risk of AAF may be the best approach to prevent the incidence of this complication.

This retrospective study has certain limitations. First, the short period of follow-up of patients in the control group may have resulted in missed cases of AAF, as this fracture may occur between 1-28 months following CLWF. In addition, among the 25 patients without adequate CT data in the non-AAF group, there is the possibility of subclinical AAF. However, evidence suggests that almost all patients with AAF will complain of new-onset severe neck pain.Second, bone fragility and bone mineral density (BMD) were not evaluated. Gehauer et al suggested that the etiology of atlas fractures may be related to osteoporosis[13]; in the setting of increased IFA or subaxialankylosis, patients withosteoporosis could be at an increased risk of AAF. Third, the cross-sectional area of the anterior arch could not be evaluated owing to the lack of precise CT data. Finally, the technical differences in the procedures performed for different patients (whether or not the muscles inserting to C2 spinous process were preserved) may have affected the C1/2 kinematics. In addition, the duration and type of neck braces following surgery were not uniform among the participating institutions.Since this was a retrospective study, these details could not be compared. A prospective study that accounts for these limitations is required to confirm our findings.

Conclusions

The incidence of AAF following C1 laminectomy is relatively higher than expected, and warrants careful preoperative assessment. CT findings of large IFA and the presence of subaxialankylosis are predictive of a high risk of AAF.

References

1. Sindou M, Chavez-Machuca J, Hashish H. Cranio-cervical decompression for Chiari type I-malformation, adding extreme lateral foramen magnum opening and expansile duroplasty with arachnoid preservation. Technique and long-term functional results in 44 consecutive adult cases -- comparison with literature data. *Acta neurochirurgica* 2002;144:1005-19.

2. Takemoto M, Neo M, Fujibayashi S, et al. Clinical and Radiographic Outcomes of C-1 Laminectomy Without Fusion in Patients with Cervical Myelopathy that is Associated with a Retro-odontoid Pseudotumor. *Journal of spinal disorders & techniques* 2013.

3. Kakutani K, Doita M, Yoshikawa M, et al. C1 laminectomy for retro-odontoid pseudotumor without atlantoaxial subluxation: review of seven consecutive cases. *European spine journal : official publication of the European Spine Society, the European Spinal Deformity Society, and the European Section of the Cervical Spine Research Society* 2013;22:1119-26.

4. Lee SE, Chung CK, Jahng TA, et al. Long-term outcome of laminectomy for cervical ossification of the posterior longitudinal ligament. *Journal of neurosurgery. Spine* 2013;18:465-71.

5. O'Shaughnessy BA, Salehi SA, Ali S, et al. Anterior atlas fracture following suboccipital decompression for Chiari I malformation. Report of two cases. *Journal of neurosurgery. Spine* 2004;1:137-40.

6. Shimizu T, Otsuki B, Fujibayashi S, et al. Spontaneous anterior arch fracture of the atlas following C1 laminectomy without fusion: A report of three cases and finite element analysis. *Journal of orthopaedic science : official journal of the Japanese Orthopaedic Association* 2016;21:306-15.

7. Ames CP, Smith JS, Eastlack R, et al. Reliability assessment of a novel cervical spine deformity classification system. *Journal of neurosurgery. Spine* 2015;23:673-83.

8. Vanharanta H, Floyd T, Ohnmeiss DD, et al. The relationship of facet tropism to degenerative disc disease. *Spine* 1993;18:1000-5.

9. Hirano Y, Sugawara A, Mizuno J, et al. Spontaneous C1 anterior arch fracture as a postoperative complication of foramen magnum decompression for Chiari malformation type 1. *Surgical neurology international* 2011;2:138.

10. Baghdassarian A, Piatt JH, Jr., Giordano K. Fracture of the anterior arch of atlas after minor trauma of the immature spine postlaminectomy. *Pediatric emergency care* 2014;30:340-2.

11. Allam E, Zhou Y. Bipartite Atlas or Jefferson Fracture? A Case Series and Literature Review. *Spine* 2015;40:E661-4.

12. Song K-J, Ham D-H, Ko J-H, et al. Stress Fracture of the Anterior Atlas Arch Following C1 Posterior Arch Resection for Cervical Myelopathy with Retro-Odontoid Pseudotumor. *J Korean Orthop Assoc* 2015;50:407-11.

13. Gebauer M, Goetzen N, Barvencik F, et al. Biomechanical analysis of atlas fractures: a study on 40 human atlas specimens. *Spine* 2008;33:766-70.

14. Le Minor JM, Rosset P, Favard L, et al. Fracture of the anterior arch of the atlas associated with a congenital cleft of the posterior arch. Demonstration by CT. *Neuroradiology* 1988;30:444-6.

15. Corominas L, Masrouha KZ. Congenital absence of the posterior arch of the atlas associated with a fracture of the anterior arch. *The Journal of bone and joint surgery. British volume* 2010;92:1300-2.

16. Hudek R, Wanner G, Simmen HP, et al. C1 fracture in a patient with a congenital cleft in the posterior arch: report on a failed conservative treatment. *BMJ case reports* 2013;2013.

17. Fielding JW. NORMAL AND SELECTED ABNORMAL MOTION OF THE CERVICAL SPINE FROM THE SECOND CERVICAL VERTEBRA TO THE SEVENTH CERVICAL VERTEBRA BASED ON CINEROENTGENOGRAPHY. *The Journal of bone and joint surgery. American volume* 1964;46:1779-81.

18. Schwab JS, Diangelo DJ, Foley KT. Motion compensation associated with single-level cervical fusion: where does the lost motion go? *Spine* 2006;31:2439-48.

19. Fujimori T, Watabe T, Iwamoto Y, et al. Prevalence, Concomitance, and Distribution of Ossification of the Spinal Ligaments: Results of Whole Spine CT Scans in 1500 Japanese Patients. *Spine* 2016;41:1668-76.

Figure captions

Fig 1.Radiographic measurements. A. X-ray parameters: (a) C2-7 sagittal vertical axis (SVA), C2-7 lordosis (CL), and T1 slope (TS) are measured on lateral view. (b) C2-7 coronal cobb angle is measured on anteroposterior view. B: Computed tomography (CT) parameters. (a) Anterior arch length and laminectomy length on axial view. (b) Superior facet angle (SFA) and inferior facet angle (IFA) on coronal view. (c) Subaxialankylosis, defined as positive as bony fusion is observed between C2 and C3 vertebral body on sagittal or coronal view (white arrow head).

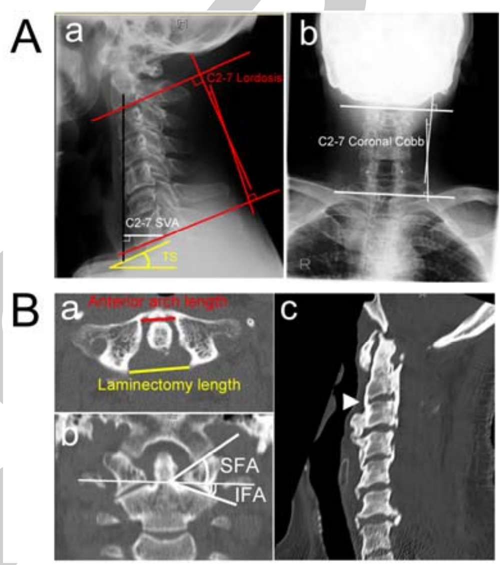

Fig 2.Receiver operating characteristic (ROC) curve of IFA as a predictor for AAF. Area under the curve

(AUC) is 0.931, indicating high accuracy. A cut-off value of IFA of 23.0° demonstrates sensitivity of

90% and specificity of 86.2%. AAF, anterior arch fracture; IFA, inferior facet angle

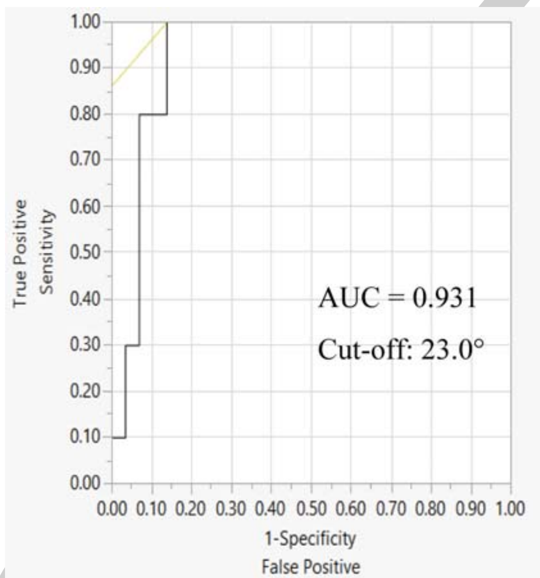

Fig 3.A case of an AAF with subaxialankylosis. (a) Bony fusion from C2 to C4 is observed on the sagittal view of the preoperative CT scan (white arrow head). (b) The coronal view of the preoperative CT shows normal SFA and IFA (35.9° and 22.8°, respectively). (c) The axial view of the postoperative CT at 6 months reveals an AAF. AAF, anterior arch fracture; CT, computed tomography; SFA, superior facet angle; IFA, inferior facet angle.

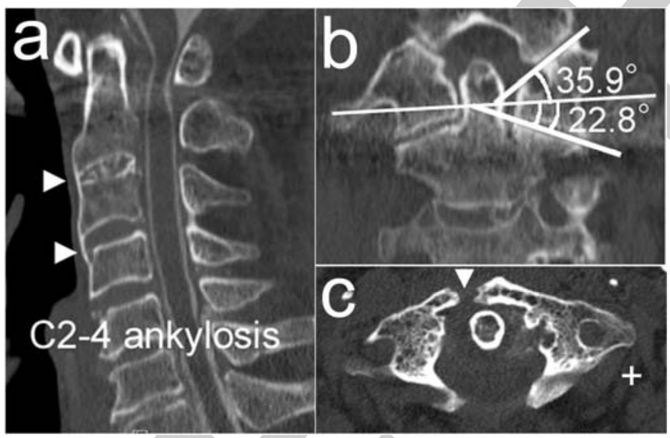

Fig 4.A case of an AAF with large IFA. (a) No subaxialankylosis is observed on the sagittal view of the preoperative CT scan. (b) The coronal view of the preoperative CT demonstrates a normal SFA and a large IFA (30.1° and 35.8°, respectively). (c) The axial view of the postoperative CT scan at 15 months shows an AAF.

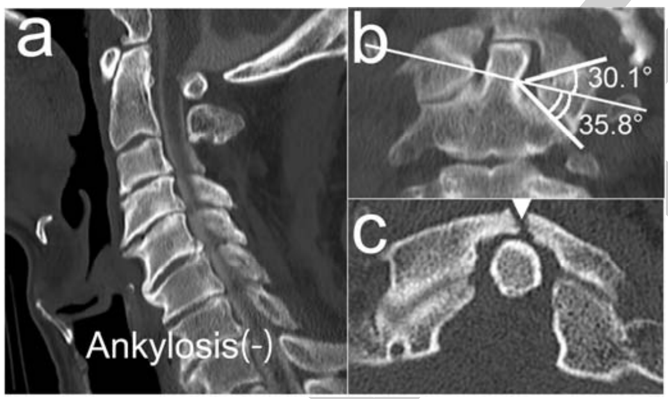

Fig 5.Schema of the potential mechanism of AAF following C1 laminectomy caused by large IFA. (Left) In case of normal IFA, the axial force yields horizontal tensile stress (black arrow) on C1. (Right) Contrary to this, flexural stress (black arrow head) is yielded by the slippage of inferior articular process of C1 due to a large IFA, leading to AAF.

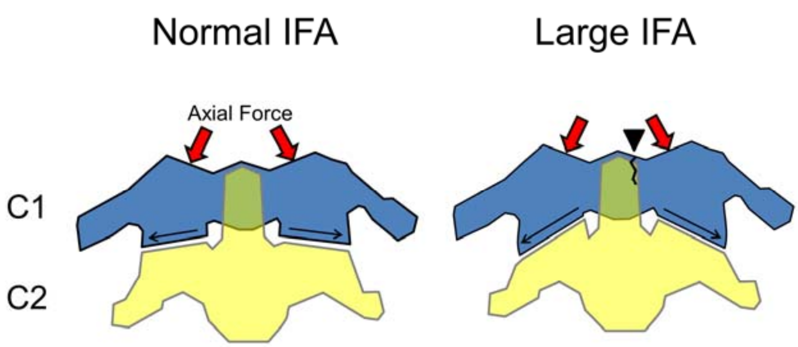

Table 1.Clinical summary of patients with anterior arch fractures

Case	Age (yrs)	Sex	Pre-C1 laminectomy diagnosis	Timing of fracture after C1 laminectomy (months)	Inciting event	Description of symptom (neck pain)	Salvage treatment/ Outcome
1	70	M	OPLL	3	No	Severe	Neck collar/Non Union, no pain
2	58	M	OPLL	15	No	Severe	Neck collar/Non Union, no pain
3	60	M	OPLL	4	No	Severe	Neck collar/Non Union, no pain
4	65	M	CSM	3	No	Severe	Neck collar/Non Union, no pain
5	62	M	OPLL	7	No	Severe	Neck collar/Non Union, no pain
6	66	M	RST	1	No	Mild	Neck collar/Non Union, no pain
7	55	M	CSM with athetoid CP	10	No	Persistent	O-C fusion / Union, no pain
8	62	M	AAS	4	No	Severe	O-Cfusion/Union, no pain
9	86	M	RST	6	No	Subsiding	Neck collar/Non Union, no pain
10	50	M	CSM	28	Rotated neck	Severe	Neck collar/Non Union, no pain

OPLL, ossification of posterior longitudinal ligament; CSM, cervical spondylotic myelopathy; RST, retroodontoidpseudotumor; AAS, atlanto-axial sublaxation; RA, rheumatoid arthritis; CP, cerebral palsy

Table 2. Patient demographics

	Fracture group (n=10)	Non - Fracture group (n = 60)	P
Age (yrs)	63.4 ± 9.7	68.6 ± 14.4	0.27
Sex (M : F)	10 : 0	35 : 25	**0.01**
BMI (kg/m^2)	24.9 ±3.1	23.4±3.5	0.23
Follow-up (months)	20.5 ± 13.7	47.2 ± 31.8	0.03
Diagnosis			
OPLL	5 (50%)	32 (53.3%)	0.84
CSM	2 (20%)	9 (15%)	0.16
RST	2 (20%)	7 (11.7%)	0.53
AAS	1 (10%)	6 (10%)	1.0
Spinal tumor	0 (0%)	6 (10%)	0.29
Comorbidity			
Hemodialysis	1 (10%)	1 (1.6%)	0.14
Prostatic Carcinoma	0 (0%)	2 (3.3%)	0.55
RA	0 (0%)	3 (5%)	0.40
Athetoid CP	1 (10%)	3 (5%)	0.65
Subaxial decompression (+)	6 (60%)	37 (61.6%)	0.92
BMI, body mass index; OPLL, ossification of posterior longitudinal ligament; CSM, cervical spondylotic myelopathy; RST, retroodontoidpseudotumor; AAS, atlanto-axial sublaxation; RA, rheumatoid arthritis; CP, cerebral palsy			

Table 3.Comparison of the radiographic parameters between the AAF and non-AAF groups.

	Fracture group (n = 10)	Non - Fracture group (n = 35)	P (univariate)	Odds Ratio / P (multivariate)
Pre-op X-ray parameters				
C2-C7 SVA	30.3 ± 4.8	32.0 ± 2.5	0.76	-
CL	9.9 ± 2.6	11.5 ± 1.4	0.59	-
C2-7 Coronal cobb	2.9 ± 1.5	4.0 ± 0.8	0.50	-
TS	27.5 ± 2.8	27.2 ± 1.5	0.92	-
TS – CL	17.5 ± 3.5	15.6 ± 1.8	0.63	-
Post-op X-ray parameters				
C2-C7 SVA	34.0 ± 5.7	35.9 ± 3.0	0.77	-
CL	6.6 ± 3.3	11.3 ± 1.7	0.21	-
C2-7 coronal cobb	2.6 ± 2.3	2.7 ± 1.2	0.95	-
TS	26.4 ± 2.8	27.9 ± 1.5	0.63	-
TS – CL	19.7 ± 4.0	16.6 ± 2.1	0.50	-
CT parameters				
C1 superior facet angle (SFA)	33.0 ± 1.3	32.0 ± 0.8	0.51	-
C1 inferior facet angle (IFA)	25.6 ± 0.9	20.0 ± 0.5	**< 0.01**	**2.5 / <0.01**
Facet tropism (+) (SFA)	1 (10%)	4 (11.4%)	0.78	-
Facet tropism (+) (IFA)	1 (10%)	0 (0%)	**0.07**	NA/ 0.99
Laminectomy length	15.8 ± 0.5	16.1 ± 0.3	0.61	-
C1 anterior arch	22.2 ± 1.2	23.3 ± 0.7	0.43	-
C1/C2 instability (+)	1 / 10 (10%)	6 / 35 (17.1%)	0.58	-
Subaxialankylosis (+)	4 / 10 (40%)	2 / 35 (5.7%)	**< 0.01**	**26.3 / 0.02**
CT, computed tomography; SVA, sagittal vertical axis; CL, C2-C7 lordosis; TS, T1 slope				

Table 4. Summary of previous publications describing anterior arch fracture

Author and Year	Number of cases	Age (yrs)	Sex	Pre-C1 laminectomy diagnosis	Timing of fracture after C1 laminectomy (months)	Inciting event	Description of symptom (neck pain)	Salvage treatment/ Outcome
O'Shaughnessy et al (2004)[5]	2	18	F	Chiari 1	5	Cough	Severe	C1-3 fusion after 3-month neck collar / Bone union, no pain
		49	F	Chiari 1	6	No	Persistent	BMP-2 infusion + Halo-vest after 3-monthneck collar / Bone union, no pain
Hirano et al (2011)[9]	1	63	M	Chiari 1	7	No	Progressive	C1-2fusion/Union, no pain
Baghdassarian et al (2014)[10]	1	4	F	Spinaltumor	4	Fall on buttocks	Worsening	Miami J orthosis/Union, no pain
Allam et al (2015)[11]	1	4	F	Spinal tumor	5	No	Stiffness	Unknown
Song et al (2015)[12]	1	70	F	RST	12	Unknown	Unknown	O-C fusion / Union, no pain
OPLL, ossification of posterior longitudinal ligament; CSM, cervical spondylotic myelopathy; RST, retroodontoidpseudotumor; AAS, atlanto-axial sublaxation; RA, rheumatoid arthritis; CP, cerebral palsy								