



Original Research Article

Traumatic spondylolisthesis of the axis. Analysis of treatment and outcome in 40 cases

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The current treatment of traumatic spondylolisthesis of the axis (TSA) is widely variable and management options include conservative options and surgical procedures. Unfortunately, the literature has several heterogeneous classification schemes, stability criteria and treatment algorithms thus making it difficult to derive clinical practice guidelines. This study seeks to identify predictors of successful outcome with conservative management. This retrospective study presents a consecutive series of 40 patients with TSA. Statistical analysis was performed to correlate clinical and radiographic measures with nonunion, residual neck pain and range of motion at final follow-up. Statistical analysis reveals Francis grade V having a significant correlation with non-union. Logistic regression analysis indicates that age ($p=0.0107$), initial translation ($p=0.0189$) and chronic tobacco use ($p=0.0104$) are significant risk factors for non-union in patients primarily treated with conservative management. Chronic tobacco use is the strongest predictor of non-union with an odds ratio of 65.99 (95% CI 2.39-21123.13). Initial translation was associated with an odds ratio of 1.62 (95% CI =1.08-3.15) and age had an odds ratio (per year) of 1.13 (95% CI = 1.02-1.39). The majority of TSA managed with conservative treatment achieved a satisfactory outcome, while early surgical fusion could be considered when risk factors exists.

Key words: axis fracture, traumatic spondylolisthesis, classification, treatment

INTRODUCTION

Traumatic spondylolisthesis of the axis (TSA) is defined as bilateral fractures involving the pars interarticularis of the axis (C2 vertebra) and is frequently associated with concurrent fractures of the lamina, articular facets, pedicles or posterior elements. These fractures are the second most common fracture of axis following odontoid fractures. Traumatic spondylolisthesis of the axis occurs with or without translation and/or angulation of the C2 vertebra on C3. These fractures have also been referred to as "hangman's fractures" although they were believed to have a different mechanism from fractures associated with judicial hanging. These injuries are commonly caused by motor vehicle accidents and falls; patients seldom present with

neurological injuries.

The current treatment of TSA is widely variable and management options include conservative options such as traction, nonrigid and rigid immobilization as well as surgical procedures through anterior or posterior approaches. Unfortunately, the literature has several heterogeneous classification schemes, stability criteria, and treatment algorithms, making it difficult to derive clinical practice guidelines.

This retrospective study presents a consecutive series of 40 patients with TSA. The purpose of this study is to identify the predictors of successful outcome with conservative management. Three generally accepted classification

schemes are compared as predictors of fracture: healing, residual neck pain and cervical mobility.

MATERIALS AND METHODS

During the period between January 2009 and December 2015, 40 patients with traumatic fractures of the C2 pars interarticularis were admitted to hospital. Patients' charts and radiographic studies were retrospectively reviewed and relevant clinical and imaging data were recorded. All patients underwent radiographic studies including three-view cervical spine plain radiographs (anterior-posterior, lateral and open-mouth) and high resolution computed tomography (CT) with sagittal and coronal reformatted images performed at the time of their initial admission. Additionally, magnetic resonance imaging (MRI) and angiography (MRA) were performed and available in 83% (33/40) of patients to assess cervical canal compression, spinal cord signal change, and vertebral artery integrity.

Inclusion criteria consisted of age (>18 years), a traumatic mechanism of injury and imaging studies demonstrating bilateral C2 par interarticularis fractures. Data points consisted of: patient demographics, medical comorbidities, contributing conditions that might affect bony union, initial fracture translation and angulation, the morphological type of displacement, presence of inferior facet fractures, primary treatment method, extent of arthrodesis, follow-up translation and angulation.

Measurements of fracture translation and angulation were assessed on the initial and latest follow-up lateral plain radiographs. Fracture translation was measured as the distance between two lines drawn parallel to the posterior margins of the C2 and C3 bodies, as described by Bono et al. (2007). Similarly, the degree of angulation was measured as described by Bono et al (2007) as the angle between the two inferior endplates of the C2 and C3 vertebrae (Levine and Edwards, 1985). Based on the initial translation (IT) and angulation (IA) measurements, in addition to the type of displacement, each fracture was classified according to three systems: Effendi et al. (1981), Francis et al (1981) and Levine-Edwards (1985). The classification proposed by Effendi et al. (1981), which was further modified by Levine and Edwards (1985) categorizes fractures according to the severity of translation, angulation and facet dislocation without specifying measurements for displacement. The Francis classification differs in that it grades disc disruption (Grade V) as C2 presenting more than half the sagittal width of the body of the C3 vertebra, or if angulation producing anterior or posterior borders of the damaged disc space is greater than the height of the centre of the normal disc below as other four grades (I-IV) are grouped referring to the criteria (translation of 3.5 mm and angulation of 11°). Fracture lines were confirmed with the reformatted coronal and sagittal plane images obtained on CT scan.

The fracture union rates were evaluated radiologically using the CT scan and/or dynamic films. Follow-up records were reviewed for neurological examination, cervical range

of motion (ROM) and clinical conditions such as cervical and musculoskeletal pain. A solid union was defined as absence of motion on dynamic flexion and extension films or the presence of continuous trabecular bone through the previous fracture site on reformatted CT images.

Clinical and radiographic measures were compared between the subset of conservatively treated patients who developed successful union (group union) and the subset who did not (group failure). Statistical analysis was performed to correlate these variables with residual neck pain and range of motion at final follow-up. Multiple logistic analysis was also used to assess relative risk factors for nonunion, residual pain and restriction of range of motion.

The variables included in this analysis were age, fracture classification (Effendi, Francis, and Levine and Edwards), initial translation of fracture (IT), initial angulation of fracture (IA), inferior facet fracture (IFF) and primary treatment method. Medical comorbidities (renal disease, diabetes and thyroid disorders) and contributing conditions (alcohol consumption, tobacco use) that might influence fracture healing were also included in the assessment. Age was converted to categorical data by stratifying patients into 65 years of age or younger and geriatric. Chronic tobacco users were defined as tobacco consumption while cessation of smoking for less than 5 years as ex-smokers.

Statistical analysis was performed using JMP (version 7.0.2, SAS Institute, Cary, NC, USA). In univariate analysis, comparison of quantitative data was carried out with Student's *t* test or Wilcoxon-Mann Whitney test, and categorical data with Fisher's analysis. Logistic regression and odds ratio were used in multivariate analysis. This study was approved by the Medical Investigational Review Board of Thomas Jefferson University Hospital and Wuxi People's Hospital.

RESULTS

Forty patients met the study inclusion criteria and were treated for traumatic C2 par fractures during the six-year period. The study group consisted of 18 female and 22 male patients with an average age of 54.7 years (range 18- 87 years). The mean follow-up period was eight months (range 3-23 months). The mechanism of injury consisted of motor vehicle collisions in 18 cases (45%), fall in 18 cases (45%), diving accidents in two patients (5%) and direct facial or head trauma in two patients (5%).

Thirteen of the TSA patients (32.5%) had significant concurrent injuries, 12 (30%) extremity fractures, six (15%) head injuries, eight (20%) thoracic injuries, two (5%) abdominal injuries, and two (5%) pelvic injuries. Concomitant spinal fractures were present in 11(28%) patients, including contiguous upper cervical spinal injuries in two patients.

Four patients had associated spinal cord injuries (ASIA score D) on admission. The neurological examinations improved to normal (ASIA E) during the follow-up period in all patients.

The mean duration of acute hospital stay was nine days (range 1-61 days). There were 37 cases primarily treated with non-rigid or rigid cervical orthosis, including 20 cases with halo vest immobilization, 15 cases with Philadelphia collar and two cases with soft collar. Among these, one patient underwent C5-7 posterior cervical fusion due to concomitant C6 fracture-dislocation but the TSA was treated with halo vest immobilization. During the three months follow-up, bony union occurred in 33/37 patients (89%) primarily treated conservatively. Of the four patients who failed to heal with three months of conservative management, three were treated with halo vest and one with hard collar. Two patients were then treated with secondary internal fixation and the other two were treated with prolonged halo vest (24 weeks) or hard collar (28 weeks) immobilization.

Primary surgical management was performed in three patients: (1) one anterior C2-3 fusion for severe anterolisthesis of the C2 vertebral body in relation to C3, (2) one posterior C1-3 fusion in a patient with concomitant C1-2 rotatory dislocation, and (3) one anterior-posterior C2-4 fusion for concurrent C3-4 instability. A rigid collar was worn constantly for 6-8 weeks postoperatively in all patients treated surgically. Bony union was obtained in all (3/3) primarily surgically treated cases.

Four patients (10%) experienced persistent pain which was unrelieved with rest and the use of nonsteroid analgesics (NSAIDs). All four had been primarily treated with halo vest immobilization. In 12 cases (30%) there was limitation in the final cervical range of motion, which was defined as restriction of more than 30% in any one of three planes (axial rotation, flexion-extension and lateral bending).

Complications associated with treatment were present in both conservative and surgical treatment arms. Three halo orthosis complications were noted including pin-loosening (2) and pin site infection (1). The patient treated with posterior cervical fusion required a revision surgery due to screw disengagement from the rod after a fall while in the halo vest. The patient treated with an anterior cervical fusion developed transient dysphagia that fully recovered two weeks later. Finally, a bone graft migrated in one patient but did not affect healing.

Univariate analysis revealed Francis grade V as having a significant correlation with fracture non-union. Three of seven (43%) patients classified with Francis grade V fractures failed unions with primary conservative treatment, while only one of thirty (3.33%) patients classified with Francis grades I-IV had a failed union ($p=0.016$). With primary conservative treatment, three (chronic tobacco users) of 10 (30%) patients' fractures failed to heal, compared with one of 27 patients (3.7%) who was not a tobacco user ($p=0.052$). All other factors did not show statistically significant differences.

Logistic regression and odds ratio analysis indicated that age ($p=0.0107$), initial translation ($p=0.0189$) and chronic tobacco use ($p=0.0104$) were all significant risk factors for non-union in patients primarily treated with conservative management. The odds ratio analysis revealed that CTU was

the strongest predictor of non-union in this model with an odds ratio of 65.99 (95% CI 2.39-21123.13). IT was associated with an odds ratio of 1.62 (95% CI =1.08-3.15) and age had an odds ratio (per year) of 1.13 (95% CI = 1.02-1.39) (Table 1).

Residual neck pain occurred significantly more often in patients with fracture lines extending into the inferior facet joint than in patients without IFF ($p=0.0324$) (Figure 1). Patients with the limitation of cervical range of motion showed significantly more IA than patients without ($p=0.0135$). Neither logistic or univariate regression analysis found further risk factors for pain and restriction of ROM.

DISCUSSION

Fracture-dislocation of the axis was described in 1866 by Houghton in the setting of judicial hangings and Wood-Jones (1908) studied this population more extensively with an emphasis on fracture mechanism and morphology. In 1954, Grogono published the first radiographs of a similar injury sustained in a motor vehicle accident, and Schneider et al. (1965) developed the commonly used term, "hangman's fracture" to describe this class of injuries. Since then, several reports about the classification and treatment of TSA have been described (Cosan et al., 2001; Effendi et al., 1981; Francis et al., 1981; Govender and Charles, 1987; Greene et al., 1997; Levine and Edwards, 1985; Marton et al., 2000; Muller et al., 2000; Robertson et al., 2006; Samaha et al., 2000; Starr and Eismont, 1993; Tan and Balachandran, 1992; Vaccaro et al., 2002; Vieweg et al., 2000). These fractures are differentiated from the classical "hangman's fractures" by their mechanism of injury. TSAs are commonly caused by motor vehicle accidents and falls and are rarely associated with neurological injury as the fractures widen the vertebral canal. In this current series, the majority of cases were as a consequence of motor vehicle accidents and falls (90%) and only four patients (10%) had a minor neurological injury (ASIA score D).

The overall prognosis of this fracture subtype is excellent and it is apparent that the majority of these fractures can be treated conservatively. Early surgical treatment may be necessary in certain situations, such as a traumatic disk herniation with spinal cord compression (Levine and Edwards, 1985), concurrent adjacent segment fractures and/or instability and new occurrence of vascular deficits (Tuite et al., 1992). Immediate surgical treatment is infrequent and in most cases, unions are achieved after approximately 12 weeks of immobilization (Borne et al., 1984; Coric et al., 1996; Cosan et al., 2001; Effendi et al., 1981; Francis et al., 1981; Govender and Charles, 1987; Marton et al., 2000; Pepin and Hawkins, 1981; Robertson et al., 2006; Tan and Balachandran, 1992). However, conservative treatment with immobilization may not provide satisfactory fracture healing and a non-union may result in persistent pain and instability. Delayed surgical treatment may be warranted if initial conservative

Table 1. Risk factors analysis of failure in primarily conservatively treated patients

Risk factors	Union	Failure	p value*	Multivariate logistic regression
Age	33	4	0.276	0.0107*
Preexisting diseases and contributory conditions				
Alcohol consumption	4	1	0.456	0.0104*
Chronic tobacco using	7	3	0.052	
Renal diseases	1	0	1.000	
Diabetes	2	0	1.000	
Thyroid disorders	1	1	0.207	
Classification schemes				
Effendi's			0.720	
Francis's (grade V vs. I-IV)			0.016*	
Levine and Edwards's			0.475	
Radiological records				
IT	2.3±2.5	5.7±5.5	0.180†	0.0189*
IA	5.7±5.7	8.5±6.3	0.353	
IFF	8	2	0.291	
Treatments				
Halo vest	17	3	0.698	
Hard collars	14	1		
Soft collars	2	0		

†Wilcoxon-Mann Whitney test; *Significance level is <0.05; IT indicates initial translation; IA, initial angulation; IFF, inferior facets fractures



Figure 1: Lateral plain x-ray and reformatted computed tomography show that the fracture involves inferior facet

management fails to promote a successful union. In recent years, immediate surgical fusion was advocated in cases with unstable TSA (Xu et al., 2010; Ying et al., 2008).

The various classification systems attempt to stratify these injuries optimally to facilitate the development of treatment algorithms. Numerous TSA classification schemes have been proposed. Effendi et al. (1981) described a classification scheme based on the fracture displacement on plain lateral radiographs, while Francis et al. (1981) defined five grades for stability based on White and Panjabi's

biomechanical criteria. Roy-Camille (1984) further emphasized a system utilizing stability criteria. Levine and Edwards (1985) modified the Effendi classification system and offered a continuum of four fracture types. This system is frequently utilized and attempts to combine radiographic appearance with the mechanism of injury to facilitate the development of management algorithms.

Francis et al. (1981) reported 123 patients primarily treated conservatively. In his series, non-union occurred in six patients: three were Francis grade II, one was grade IV

and two were grade V. He therefore noted that marked angulation was a risk factor of nonunion. Hadley et al. (1989) in a retrospective review of axis fracture treatment noted that 5% (2/39) of patients with hangman's fractures treated with halo orthosis did not heal. Tuite et al. (1992) treated five patients surgically after failure of traction and halo vest immobilization and noted that all five patients had Effendi type II fractures. In Greene et al.'s (1997) retrospective review, 10% (7/72) of patients with conservatively managed hangman's fractures failed to develop a solid union. Classification according to Effendi's criteria revealed that six were type II, and one was type III. According to the Francis classification system, types I, II and V were identified in one patient each while types III and IV were identified in two patients each. 33 and 36% of the patients characterized as Effendi Types II-III or Francis Grade III-V, respectively, required early surgical treatment. Muller et al. (2000) described four Effendi type II spondylolisthesis patients needing secondary internal fixation due to persistent instability and reported that type II spondylolisthesis fractures might be highly associated with nonunion, further suggesting that internal fixation is recommended.

In this study, the three most commonly utilized classification schemes were compared. The incidence of non-union did not differ between types categorized according to Effendi and Levine-Edwards. However, the Francis Grade V fractures had the highest failure rates and their incidence was greater than the sum of all the four other types combined. Francis classification can be differentiated from the other two classification schemes in that the Effendi and Levine-Edwards systems emphasize mechanism. The Francis classification system specifically identifies disk disruption as Grade V lending importance to discoligamentous disruption as a marker of instability. The findings of the present study support this emphasis.

Clinically, it is necessary to distinguish between stable and unstable fractures, however, there is controversy on the criteria for stability. Although existing classification systems for TSA attempt to define surrogates for stability, none of these has yet been absolute. Most of the classifications do not provide detailed radiographic criteria to define stability although discoligamentous integrity is clearly one component. One commonly held belief is that, without other lesions, bilateral pars interarticularis fractures are stable; on the other hand, if there are concomitant discoligamentous injuries then instability occurs (Mestdagh et al., 1984; Roy-Camille et al., 1984), which in Francis classification system is type V.

Magnetic resonance imaging (MRI) may also be a valuable instrument in clinical decision-making as it affords supplemental information about the spinal canal and discoligamentous architecture and integrity (Vieweg et al., 2000). This information should be used judiciously with other available studies. Even though MRI might have the ability to visualize disruption of intervertebral disks, anterior longitudinal ligament (ALL) and posterior longitudinal ligament (PLL), the reliability of these findings

in predicting instability is not known, particularly when superimposed on acute soft tissue injury or degenerative changes (Flanders et al., 1992; Hall et al., 1993).

Through cadaveric studies (White, 1978) clinical instability of the cervical spine has been correlated with greater than 3.5 mm translation between two adjacent vertebra and/or greater than 11° of angulation. Borne et al. (1984) considered displacement less than 2 mm as measured by interpedicular diastasis of C2 as stable, whereas displacement greater than 2 mm indicated spondylolisthesis and suggested that the fracture was unstable. Coric et al. (1996) reported that the best indicator of ligamentous disruption was translation greater than 6 mm on the lateral radiograph. Muller et al. (2000) considered initial translation greater than 4 mm or angulation greater than 11° to be unstable. Moon et al. (2001) characterized a fracture as unstable if there was translation between C2 and C3 with an unusual widening or rotation between the vertebral body and arch. According to our data analysis, patients failed to obtain a solid union with primary conservative treatment if they had greater initial translation, but an absolute cutoff point could not be identified.

Further analysis of the initial clinical and radiographic factors also indicated that increased patient age and chronic tobacco use were significant risk factors for fracture non-union.

Nonrigid immobilization was generally regarded as the modality of choice for the treatment of minimally displaced or nondisplaced fractures (Longo et al., 2010), however there are some reports supporting its use as the primary treatment for fractures even with displacement. Coric et al. (1996) suggested that the majority of Hangman's fracture may be treated by nonrigid immobilization. In their series, 6 of 64 patients had translation up to 6 mm and all were successfully treated with non-rigid immobilization alone. Grady et al. (1986) used Philadelphia collar instead of halo orthosis to treat 27 C2 and C3 fractures, even for one case with greater than 4 mm spondylolisthesis; all patients developed a successful fusion. In this series, hard and soft collars were as effective as halo orthosis for facilitating fracture healing. Iizuka et al. (2016) treated 8 elderly cases with coronally oriented vertical fracture on their C2 vertebral body with Philadelphia collar and all got fusion. One advantage of nonrigid immobilization is that it is more comfortable and carries a lower cost. Furthermore, this modality avoids the morbidity of halo vest which has been reported in the range of 12 to 36% - including skin breakdown, pin site infection, osteomyelitis, septicemia, cerebrospinal fluid leak, and subdural abscess (Chan et al., 1983; Cooper et al., 1979; Garfin et al., 1986). However, collar fit, patient reliability and age should be considered when nonrigid immobilization is being employed.

In addition to the goal of obtaining a healed fracture, the optimal treatment should target early mobility, rehabilitation and return to painless function. The development of a pseudarthrosis can result in cervical pain not only with movement but also with rest (Bouloosa et al., 2004). Residual neck pain in cervical injury patients is a significant cause of

reduced daily activity and failure to return to work. Watanabe et al. (2005) used CT to assess the fracture line of TSAs and noted that fracture involvement of the inferior facet was a potential risk factor for residual neck pain. In their study, final follow-up angulation was significantly higher in patients with residual neck pain while translation was not. Angulation was therefore thought to be a predictor for residual neck pain. In the present series, the conservatively treated patients with fractures of the inferior facets also had the greatest risk for residual neck pain. ($p=0.0324$).

The upper cervical spine (occipital-C3) is the most mobile region of the spine; therefore, range of motion is an important component of patient outcome. Monterumici and Sinigaglia (2007) treated six hangman's fracture patients with halo vest immobilization and noted no difference in ROM compared with a control group, but young patients had a better ROM than elderly. Muller et al. (2000) documented severe restriction of motion in one patient who was classified as Effendi type I and treated conservatively with an orthosis, and in three patients who were classified as type II without mentioning the specific treatment. All of their other patients had full range of motion on follow-up evaluation. In the current study, age, fracture classification and other radiographic factors did not correlate with final cervical range of motion.

Surgical procedures for hangman's fractures include anterior fusion, posterior fusion, combined approach fusion and Judet's procedure (transpedicular osteosynthesis). Anterior discectomy and fusion are reported to prevent neck pain (Palit et al., 1999) and maintain rotatory motion (Moon et al., 2001), while posterior approaches can provide more reliable fixation but are associated with prolonged axial pain (Ying et al., 2008). The Judet procedure maintains the majority of the motion but is only indicated when the fractures are not accompanied by discoligamentous injury (Taller et al., 2000). Full reduction and realignment are rarely achieved by acute conservative treatment; even if they are initially achieved, most will lose this reduction after immobilization in external devices. In this study, however, residual deformity did not appear to affect the overall outcome. Moreover, there is an inherent risk with any invasive surgical treatment. In the five surgically treated patients- three primary and two secondary- complications or instrument revision occurred (60%).

There have been few previous reports examining factors that influence fusion in TSA patients treated conservatively. The current study attempts to identify radiographic and clinical factors that influence fusion so that surgical intervention may be reserved for circumstances with poor conservative outcomes. Additionally, three commonly used classification schemes are comparatively examined as predictors of fusion with conservative management. Certain outcomes and relevant factors are also discussed. The limitations of this study should be acknowledged and include the bias associated with retrospective studies, the absence of locked facet dislocation cases and the absence of clinical

outcome evaluation in the chart review.

Conclusion

In summary, this series indicates that, the majority of TSA managed with conservative treatment achieved a satisfactory outcome including residual deformity, clinical symptoms and functional outcomes. However, C2-3 disk disruption defined as Francis Grade V, oldness, chronic tobacco use and marked initial translation reduced the probability of a solid union with primary conservative treatment. Additionally, fracture lines that extend into the inferior facets are a risk factor for persistent residual neck pain.

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Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this manuscript.

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