

Post-traumatic Collet–Sicard syndrome: personal observation and review of the pertinent literature with clinical, radiologic and anatomic considerations

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Abstract

Study design The lesion of the lower four cranial nerves, commonly called Collet–Sicard syndrome, can be caused by a blunt head and neck trauma. It may be associated to an isolated fracture of the occipital condyle or of the atlas.

Objective The aim of this report is to assess the modality of the trauma, the type of fracture, the anatomic characteristics, the treatment and clinical results of this syndrome.

Summary of background data We discuss 14 cases of fracture of occipital condyle and of atlas and 1 personal case.

Methods We analyzed 14 cases collected from the literature between 1925 and 2013, reported a further personal case and performed an anatomical study of the paracondylar, atlas and styloid process region. The anatomical dissection was performed to assess the anatomic relationships in the site of transit in which the nerves IX, X, XI and XII are injured.

Results A total of 14 cases of p-CSS were collected: 9 caused by a condyle fracture and 5 by an atlas fracture. The patients were 13 males and only 1 female, 10 of them had a blunt trauma due to the result of axial loading (force directed through the top of the head and through the spine) falling on the head. The nine cases with a condyle fracture were associated to the dislocation of part of it, while those with atlas fractures showed the fracture and/or disjunction of the

articular mass. The anatomical evaluations reveal that the lower four cranial nerves, at their emergency, pass through a close osteo-ligamentous space in relationship to the condyle. Below they run through a little wider channel between the articular mass of C1 and the styloid process. Two cases underwent surgical procedure. All the other cases were treated conservatively with immobilization of the cervical spine. During follow-up three cases with condylar fractures were found to be clinically unchanged and six showed modest improvements while one case with atlas fracture had a complete recovery and four improved significantly.

Conclusions The p-CSS is caused by force directed through the top of the head. We suppose that the nerve injuries are due to their laceration caused by a displacement of a condyle fragment or to their compression and stretching when they pass between the lateral mass of the atlas and the styloid process. These modalities of trauma explain the better clinical results in patients affected by C1 fractures. Conservative treatment is the option of choice. Surgical option, when choosed, is not considered to fix nerve damages.

Keywords Collet–Sicard syndrome · Atlas · Occipital condyle · Vertebral fracture · Jefferson fractures

Abbreviation

CSS Collet–Sicard syndrome

p-CSS Post-traumatic Collet–Sicard syndrome

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Introduction

The Collet–Sicard syndrome (CSS) is an unilateral paralysis of the lower cranial nerves (IX, X, XI and XII). It is named after Justin Frederic Collet [1] and Jean-Athanase

Table 1 Cases of p-CSS collected in the literature

Authors	Age/ sex	Type of trauma	Type (T) fracture: condyle (C) or atlas (A)	Clinical deficit	Treatment	Clinical results
Rebattu et al. [19]	43, M	Beaten	C	IX, X, XI, XII	None (autopsy)	Death for pulmonary infection
Bolender et al. [11]	23, M	Motor vehicle accident	C—avulsion and displacement	IX, X, XI, XII	Collar	Slight improvement
Hashimoto et al. [15]	71, M	Bicycle accident	C—avulsion and displacement	IX, X, XI, XII	Conservative	Unchanged
Wani MA et al. [21]	67, M	Fallen down the stairs	C—two linear fracture of the occipital bone	IX, X, XI, XII	Conservative	Unchanged
Bridgman and McNab [33]	32, M	Fallen down the stairs	C—avulsion fracture of the occipital condyle	X, XI, XII	Collar	Recovery XI
Sharma et al. [20]	35, M	Motor vehicle accident	C—medial displacement	IX, X, XI, XII	Surgery	Partial recovery XI, XII
Young et al. [22]	26, F	Motor vehicle accident	C—compression of the medulla	IX, X, XI, XII	Halo brace	Slight improvement
Legros et al. [18]	44, M	Motor vehicle accident	C—fracture of the clivus involving jugular foramen	IX, X, XI, XII	Collar	Slight improvement
Caroli et al. [12]	34, M	Motor vehicle crash	C—Anderson–Montesano T III	IX, X, XI, XII	Halo vest	Improved slightly
Erol et al. [14]	31, M	Motor vehicle accident	C—avulsion fracture of the occipital condyle	IX, X, XI, XII	Halo vest	Unchanged, except IX partial recovery
Zielinski et al. [23]	23, M	Rammed head into the wall	A—Displacement of the lateral mass and fracture of the posterior arch	IX, X, XII	Halo vest	Complete recovery
Connolly et al. [13]	56, M	Motor vehicle accident	A—Three-part “Jefferson fracture”	IX, X, XI, XII	Halo vest	Complete recovery X, XI, XII and partial IX
Hsu et al. [16]	18, M	Motorcycle accident	A—Jefferson T III Landells T II E	IX, X, (XI), XII	Collar and Traction	Complete recovery except partial IX
Kwon et al. [17]	46, M	Falling 2 m.	A—“Jefferson fracture” left arches, transverse and articular mass	IX, X, XI, XII	Surgery	Partial recovery XII
Our case	58, M	Fell from the horse	A—Jefferson T III Landells T II	IX, X, (XI), XII	Philadelphia	Partial recovery IX e XII, unchanged X, complete recovery XI

Sicard [2] who first described it independently. It is an uncommon syndrome caused by lesions of the skull base as primary or metastatic tumors [3–5], vascular lesions [6, 7], inflammatory processes [8], iatrogenic complications [9, 10] and trauma [11–23].

Closed head and neck injuries resulting in atlas or occipital condyle fractures in some cases may cause a post-traumatic Collet–Sicard syndrome (p-CSS). We report a clinical case of such syndrome presented in a patient with a C1 fracture. Moreover, we conducted a review of the literature analyzing the closed head trauma resulting in p-CSS. We studied the paracondylar, atlas and styloid region through a cadaver dissection to evaluate the relationships between vascular, osteo-ligamentous structures and the cranial nerves involved in this syndrome. In the light of the review, our case and the anatomical study performed, we also discuss the biomechanics that causes the lesion of the four cranial nerves involved in the p-CSS. Finally, we outline the possible therapeutic options and long-term results.

Methods

We conducted a literature review of the p-CSS, reported a further personal case and performed an anatomical study on cadavers.

Literature review

A computer-aided search of MEDLINE using pubmed was conducted from 1925 to 2013 to collect p-CSS cases in closed head and neck trauma. The research was performed using keywords as “Collet–Sicard syndrome”, “atlas

fracture”, “condyle fracture” and was limited to human studies.

Traumatic cervical lesions associated with a p-CSS reported and well described in the literature were 13, 9 of which were caused by a condyle fractures and 4 by atlas fractures. We reported also a personal observation of a p-CSS in a patient with C1 fracture. Table 1 summarizes all the cases collected. All reported cases were male except the case described by Young et al. [22] which involved a female with a condyle fracture. The overall mean age was 44.2 years (range 18–71), which for condyle fractures was 41.5 years (23–71) and for atlas was 40.2 years (range 18–58).

The type of fracture was identified according to the classifications reported in the literature [24–28]. Only two authors in the review [12, 16] as well as us explicitly referred to those classifications. In the remaining ten cases the fractures were not classified, so we assigned a type and reported it in Table 1 using the description provided from the manuscripts.

The classification system used in condyle fractures was the one proposed by Anderson and Montesano [24], updated by Tuli et al. [29]. This classification is based on clinical and radiological criteria and provides: type 1 non-displaced OCF (stable); type 2A displaced OCF with intact ligaments (stable); type 2B displaced OCF with radiographic evidence of craniocervical junction instability (unstable). For the atlas fracture we used the anatomical classification proposed by Jefferson [30] and updated by Landells and Van Peteghem [28] that provides: type I fractures involve the anterior or posterior arch alone (Jefferson types I and II); type II fracture involves fractures of both the anterior and posterior arches (Jefferson type III); type III fracture is a fracture of the lateral mass with or without a fracture of the arch (Jefferson type IV).

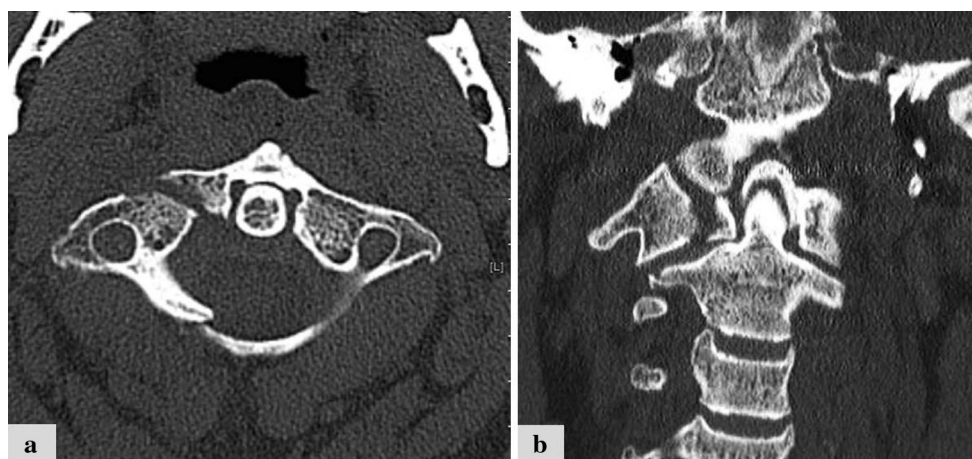


Fig. 1 CT scan performed after trauma shows in axial section (a) fracture of the right arches of the atlas. The coronal reconstruction (b) shows the fracture of the right articular mass that is laterally displaced

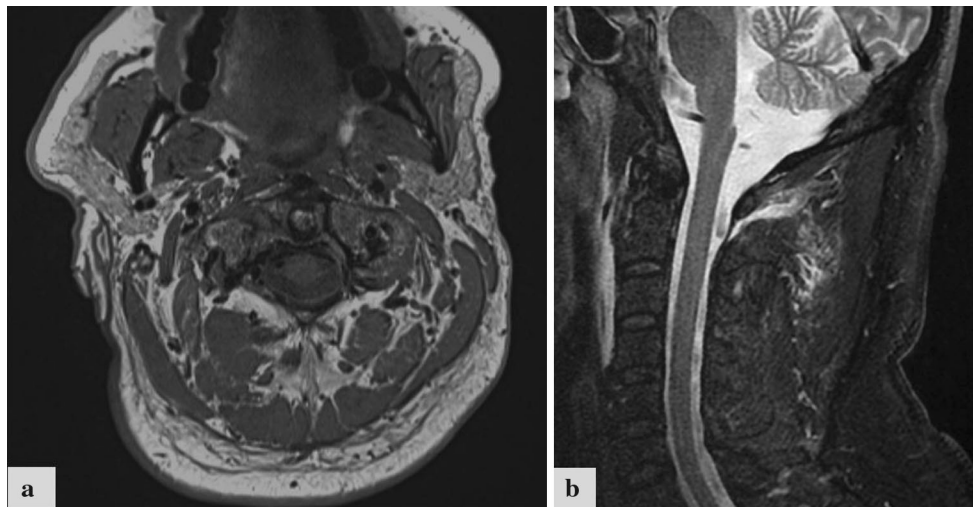


Fig. 2 MRI performed after trauma shows in T1-weighted axial images (a) the right articular mass fracture of the atlas with transverse ligament apparently intact; the sagittal STIR (=short inversion time

inversion recovery images) images (b) show suboccipital muscles and C1–C2 posterior ligamentous structures post-traumatic edema with normal representation of the spine

Neurological examination showed unilateral deficit of cranial nerves IX, X, XI and XII in nine cases of condylar fracture and in five cases of atlas fracture even if less severe, in particular as regards the XI cranial nerve in three cases it was only just mentioned.

In only two cases a surgical treatment was performed, in all the other cases the treatment of choice was conservative. A case of atlas fracture [17] underwent occipito-cervical fixation, and a case of condyle fracture [20] with marked medial displacement of the bony fragment underwent suboccipital approach to remove the fragment. Conservative treatments consisted in immobilization of the cervical spine by halo vest (five cases) or in any other brace.

One case died because of pneumonia [19]. The long-term results in the nine cases of condyle fracture showed an unchanged clinical status in three patients and a slight improvement in all the other cases, while in the five cases of atlas fracture, one had a complete recovery and four significantly improved and had not any residual disabling deficits.

Personal observation

A 58-year-old man was sent from another hospital to our attention, 3 days after the incident, because of a cervical trauma that occurred falling from a horse. A CT scan performed after the fall documented a fracture of the right lateral mass involving both the right anterior and posterior arches of the atlas (Fig. 1). The patient was discharged with the prescription to wear an immobilization collar, Philadelphia cervical collar.

At the first clinical exam the patient complained chief complaint of neck pain, hoarseness and difficult in swallowing and at the neurological examination showed tongue deviation to the right side, slurred speech, dysphonia, paradoxical dysphagia (i.e. swallowing of solids is fine but swallowing liquids elicit difficulty) and difficulty in elevation of the shoulders, mostly on the right side. Otolaryngology examination revealed absent gag reflex on the right side, the soft palate pulling to the left side, insensate oropharyngeal wall on the right side and the laryngoscopic examination showed (partial) right vocal cord palsy and pooling of secretions on the right side. In summary there was a IX, X, XII cranial nerve palsy of the right side and a doubtful XI cranial nerve palsy, thereby identifying a p-CSS.

The patient, then, performed a cervical spine MRI that documented the transverse ligament integrity and confirmed the right articular mass fracture of C1 extended to the point of insertion of the ligament (Fig. 2). We decided to continue the conservative treatment wearing the Philadelphia cervical collar and performing radiological seriated controls over time by CT, MRI and standard X-ray. At the first, CT scan control was performed 30 days after injury and it was observed that the space between the styloid process and the transverse process of the atlas C1 (Fig. 3a) that at the side of the fracture was lower of about 2 mm compared to the contralateral side. A 3D reconstruction of the region (Fig. 3b) let us see the relationship between fractured atlas and styloid process.

After 120 days the CT scan control showed the fusion of the fracture (Fig. 4) so that the patient has gradually reduced the use of the collar that was stopped after 30

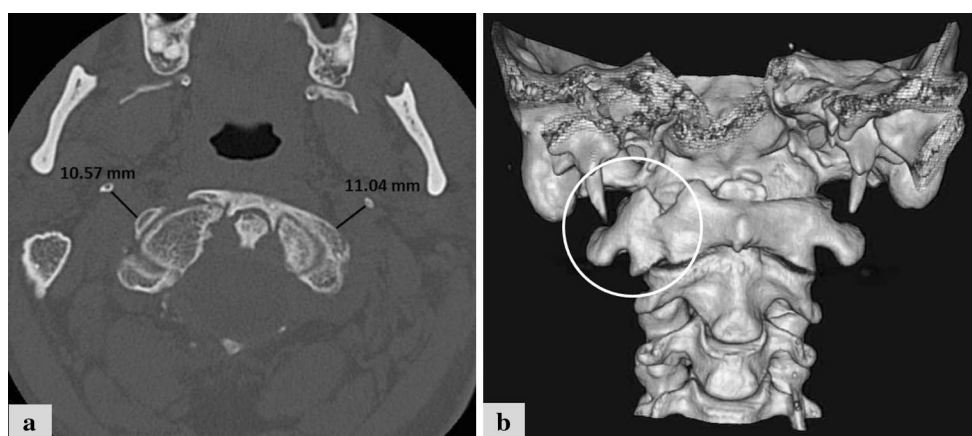


Fig. 3 Axial CT scan (a) performed after 30 days from trauma showing the distance between the styloid process and the atlas lateral mass with bilateral measurement. 3D CT reconstruction (b) with a

posterior view of the occipital-cervical junction that highlights the relationship between atlas and condyle processes. The *white circle* indicates the C1 fractured portion

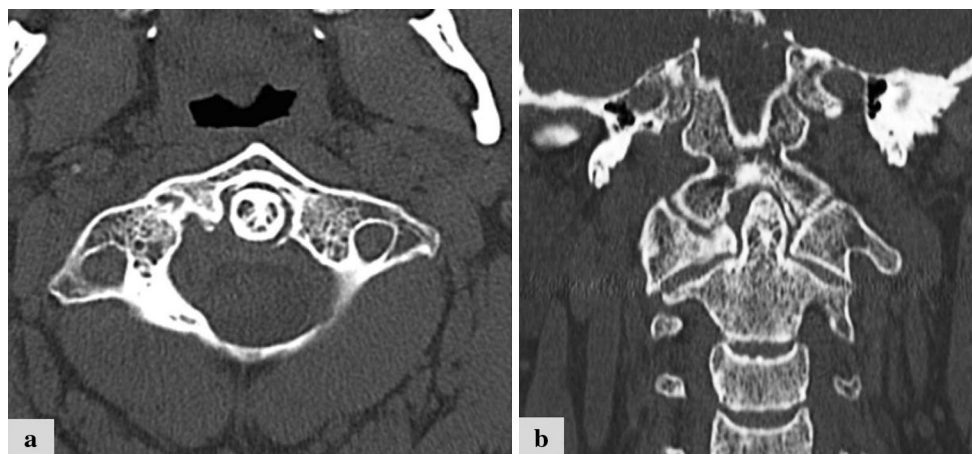


Fig. 4 Axial CT scan (a) performed after 120 days from trauma showing the complete fusion of the fractured arches. Coronal reconstruction images (b) showing the complete fusion of the

fractured articular mass. Moreover, coronal image shows a left side displacement of the odontoid process

more days. 10 months after the trauma his residual symptoms were a slight dysphonia with dysphagia and persisted evident, although reduced, the deviation of the tongue associated with right side hypotrophy. Taste alteration and reduced sensitivity of the palate persisted unchanged.

Anatomical observation

Anatomical specimens' preparation was obtained using two different exposures: anterolateral in the supine position with the head rotated 45° contralateral to the approach, a presternocleidomastoid incision was performed (Fig. 5) and the other one was a posterolateral approach in the 3/4 prone position a curvilinear incision was performed from the midline to the high lateral portion of the mastoid

(Fig. 6a, b). The first one, which shows an anterolateral approach to the paracondylar, atlas and styloid regions, illustrates the transit of IX, X and XI cranial nerves in the deeper position and therefore not visible. It is possible to see the course of these nerves at the level of the atlas which run through a narrow space between the C1 transverse process and the styloid process. It is evident, moreover, that these nerves are collected at their emergency in the skull base, in the paracondylar region. The second representation exposes the same region using a posterolateral approach, which highlights the narrowness of the transit area of the lower four cranial nerves, particularly their emergence from the skull base. The dissection shows the course of the twelfth intracondylar after condyle subtotal removal (Fig. 6b). It is possible to view the occiput and the C1 lamina, which are not evident in the previous preparation.

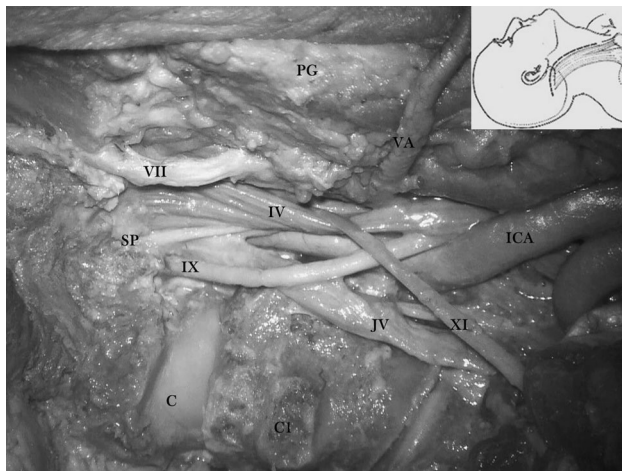


Fig. 5 Photographs obtained during cadaveric studies. On the *upper side* of the picture we put a schematic drawing of the skin incision (*dashed line*). It shows an anterolateral approach in supine position with the head rotated 45° contralateral, to expose the paracondylar–C1 region. For anatomical details see text. PG parotid gland, VA vertebral artery, ICA internal carotid artery, IV, VII, IX and XI cranial nerve, JV jugular vein, C condyle, Cl atlas, SP styloid process

These preparations offer the possibility to identify the means by which it would be possible to proceed to a stabilization of the condylar region.

Discussion

Frédéric Justin Collet (1870–1966), French pathologist and otoryngologist, in 1915 described a disorder he called “glossolaryngoscapulopharyngeal hemiplegia”, later

named “Collet’s syndrome”. This disorder is caused by a unilateral lesion of cranial nerves IX, X, XI, and XII, resulting in paralysis of the vocal cords, palate, trapezius muscle and sternocleidomastoid muscle. Homolateral nerve lesions may also results in hemianesthesia of the larynx, pharynx and soft palate. This condition is sometimes referred to as “Collet–Sicard syndrome”, named in conjunction with Jean-Athanase Sicard (1872–1929), French neurologist and radiologist, who provided a description of the disorder independent of Collet.

Rebattu et al. [19] in the 1925 first reported a case of lower cranial nerve palsy associated to condyle fracture. Zielinski et al. [23] in the 1982 first described the p-CSS associated to atlas fracture. After these two observations many other cases have been report as summarized in Table 1.

In the p-CSS all ages are represented even if younger groups are predominant (mean age 40 years) with no differences between condyle and atlas fractures. The mechanisms of trauma causing condyle or atlas fractures are attributable to activities that may cause the projection of the body upward with subsequent relapse on top of the head. In fact, in ten cases the trauma was due to the projection of the body because of a fall from a motorcycle, bicycle or horses with the head crashed on the ground and in one case there was a head injury resulting in precipitation from the stairs. Zielinski [23] described the case of a patient who rammed his head into a wall; even in this case the forces are directed axially as the other cases. Uncertain remain the case of a patient described by Rebattu who was beaten because the description is not clear.

We found that a p-CSS is caused by isolated condyle or atlas fractures, and that the mechanism and the

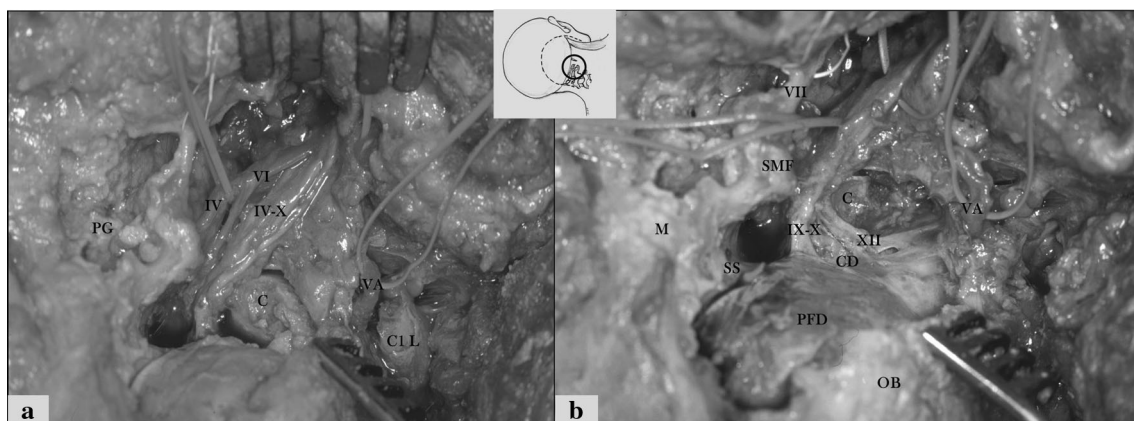


Fig. 6 Photographs obtained during cadaveric studies. On the *upper side* of the picture we put a schematic drawing of the skin incision (*dashed line*) and the *red circle* shows the photographed region. It shows a posterolateral approach in 3/4 prone position to expose the right paracondylar–C1 region extended to upper neck. Paracondylar region dissection before (a) and after (b) an almost complete resection

of the occipital condyle to show the intracondylar portion of the XII cranial nerve. For anatomical details see text. SS sigmoid sinus, C condyle, Cl L atlas lamina, VA vertebral artery, PG parotid gland, IV, VI, VII, X and XII cranial nerve, PFD posterior fossa dura, M mastoid, OC occipital bone, CD condyle dura, SMF stylomastoid foramen

biomechanics of trauma in these two cases are different and must be examined separately.

Atlas fractures

Jefferson [30] in his classic work reports that the atlas fractures are the result of an axial load or a vertical compression of the atlas. Subsequently, numerous biomechanical models have been proposed. Sköld [27] and Panjabi [31] have shown that a perfectly axial load on C1 capable of fracturing the vertebra is rare, while in most cases it is combined to mechanisms of flexion, extension, lateral bending and rotation.

Zielinski et al. [23] through their cadaver study found that there was only 8–10 mm of space between the transverse process of the atlas and the styloid process and the CN IX–XII pass through this narrow space. If the lateral mass of the atlas is abnormally displaced laterally after a fracture, this space would be reduced to nearly nothing and lower CN palsies would be likely to develop. In our case, we believe that the biomechanics of the trauma consisted of a fall with hyperflexion of the head and lateral transmission of the forces on the lateral mass of C1 that was fractured as well as its anterior and posterior arches. This possibility is supported by the fact that in all other fractures of C1 with p-CSS, shown in Table 1, the mechanisms of trauma are similar with massive rupture of the lateral mass and of the arches at the side of clinical manifestation. This type of fracture can be classified as Jefferson type III or Landells type II or III.

This mechanism of trauma is able to explain how the lower four cranial nerves are compressed and stretched by the approach of the transverse process to the styloid one. After the traumatic event the realignment of the head and the reduction of the avulsion of the articular mass would lead to a partial normalization of the space. Detting [32] reports that in a 16-year-old patient with cranial nerve IX, X and XI deficit the distance from the styloid process to the transverse process of the atlas was only 5 mm on the side of the fracture. So far these few observations do not allow to affirm that it is possible to evaluate a possible anatomical damage in case of p-CSS based on radiological measurements of the space between the C1 transverse and styloid process.

Biodynamics of the traumatic event may also explain clinical improvement with complete recovery as in the case of Zielinski et al. [23] or the improvement of the neurological deficits in the remaining reported cases. This hypothesis is supported by the fact that the hypothesized traumatic stretching and compression of the nerves do not determine a final tearing.

The anatomical dissection of the region between the atlas and the styloid process (Fig. 5) we performed further defines these considerations. In fact from the dissection of this region

it is evident that the glossopharyngeal, vagus and the accessory spinal nerves pass in a narrow space between the styloid process and its stilojoid ligament and the transverse process of the atlas. The hypoglossal nerve is not visible because it transits on the anatomical plane immediately below.

Occipital condyle fracture

It is generally the result of the condyle dislocation. In fact, in our series all cases are classified as types 2A and 2B according to Tuli et al. [29] with displaced OCF with or without intact ligaments. Only the case described by Wani et al. [21] which was studied only on the basis of plain radiographs and not on tomographic exams is associated to linear fracture without displacement of OCF. The biomechanics of these fractures, in which the nerves are damaged by the displaced condyle fragment, explains the serious and often complete injury of the lower four cranial nerves, and explains the precocity of the symptoms and the slight possibility of subsequent recovery after trauma. In fact, the cases of condyle fracture associated to p-CSS reported in Table 1 achieved only modest improvements, and the case of Wani et al. [21] was so modest that patient still continued to use a nasogastric tube because of a severe impairment of swallowing when discharged.

The cadaver dissection we performed helps us understand the anatomical region. It is evident (Figs. 5, 6) as the lower four cranial nerves pass close to the condyle and as the dislocation of a fragment in a very small space between the bony and rigid structures can tear them.

The long-term results are different in those fractures; in fact, while in the C1 fractures we can assist to a significant improvement of symptoms in the p-CSS associated to condyle fracture generally there is no recovery of the deficit or only a modest improvement of them.

Surgical treatment of the cranial nerve lesions in p-CSS is not considered an option of choice. In fact in the case described by Sharma et al. [20] on a condyle fracture and the one reported by Kwon et al. [17] regarding the atlas fracture, surgery was performed to decompress spinal cord or to restore the vertebral stability. All the other cases of p-CSS were treated by immobilization with Halo vest or cervical collar, which in our case was the Philadelphia type. Often it is necessary the use of a nasogastric tube or performing a tracheotomy to remedy the risks associated with swallowing and breathing disorders. In the series reported these procedures were carried out only in some condyle fractures.

Conclusion

The p-CSS is caused by occipital condyle fractures with dislocation of a fragment or by atlas fractures with the

articular mass dislocation, following a trauma with force directed through the top of the head. In the first case the lesion of the lower four cranial nerves is severe and causes a complete tearing. In atlas fracture the nerves are stretched when they pass between the lateral mass of the atlas and the styloid process with a minimum damage of them. Therefore, better long-term results are observed in p-CSS associated to atlas fractures. These findings are supported by the anatomical dissection that shows how these nerves transit in a little space but compressible next to the articular mass of the atlas and instead in a bony and ligamentous box at the condyle level.

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