

Traumatic Acute Spinal Cord Injury Associated with Ossification of the Cervical Posterior Longitudinal Ligament without Tomographic Evidence of Bone Trauma. Case Series and Literature Review

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ABSTRACT

Objectives: to present a case series of traumatic acute spinal cord injury associated with ossification of the cervical posterior longitudinal ligament (OPLL) without tomographic evidence of bone trauma and to perform a narrative review of the literature on its treatment and postoperative prognosis. **Materials and methods:** We described a case series of patients with traumatic spinal cord injury and association of cervical OPLL, treated by our team during the period January 2012 - December 2019. We excluded patients with an association of vertebral fractures and/or dislocations, and those referred to another center before treatment and with incomplete records. Additionally, a narrative review of the literature on postoperative treatment and prognosis of this association was carried out. Ten articles were obtained with our search strategy for the narrative review. **Results:** A sample of 5 cases was formed, all males, with an average age of 62.2 (+/- 9.36), 4 cases were surgically treated by a posterior approach and 1 case was conservatively treated. **Conclusion:** we presented a case series of a rare association in our region, preceded only by an isolated case report. The review of the current literature suggests timely surgical treatment over conservative treatment, but controversies persist in this regard.

Keywords: Ossification; posterior longitudinal ligament; spinal cord injury; cervical trauma.

Level of Evidence: IV

Trauma medular cervical en pacientes con osificación del ligamento longitudinal posterior sin evidencia de fractura. Serie de casos y revisión bibliográfica

RESUMEN

Objetivos: Presentar una serie de casos de lesión medular aguda traumática asociada a osificación del ligamento longitudinal posterior cervical sin evidencia tomográfica de trauma óseo y realizar una revisión narrativa de la bibliografía sobre su tratamiento y pronóstico posoperatorio. **Materiales y Métodos:** Descripción de una serie de pacientes con lesión medular aguda traumática y osificación del ligamento longitudinal posterior cervical, tratados por nuestro equipo, entre enero de 2012 y diciembre de 2019. Se excluyó a pacientes con fracturas o luxaciones vertebrales asociadas, aquellos derivados a otro centro antes del tratamiento y con registros incompletos. Además, se llevó a cabo una revisión narrativa de la bibliografía sobre el tratamiento y el pronóstico posoperatorio de esta asociación en la última década. Con nuestra estrategia de búsqueda, se obtuvieron 10 artículos, a partir de los cuales se desarrolló la revisión narrativa. **Resultados:** Se conformó una muestra de 5 casos, todos hombres, con una edad promedio de 62.2 años (DE ± 9,36), 4 pacientes fueron operados por vía posterior y uno recibió tratamiento conservador. **Conclusión:** La lesión medular aguda traumática asociada a osificación del ligamento longitudinal posterior cervical es infrecuente en países no asiáticos, precedida, en nuestra región, por un reporte de caso aislado. La revisión de la bibliografía actual sugiere al tratamiento quirúrgico oportuno por sobre el tratamiento conservador, pero persisten las controversias al respecto.

Palabras clave: Osificación; ligamento longitudinal posterior; lesión medular aguda; trauma.

Nivel de Evidencia: IV

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INTRODUCTION

Traumatic spinal cord injury still represents a challenge in the care of polytraumatized patients due to the controversies associated with diagnostic-therapeutic aspects and because, once established, it constitutes a “devastating” injury with great morbidity and mortality and socioeconomic impact.^{1,2}

We know that a timely diagnostic evaluation conditions the therapeutic opportunity, in which the finding of unstable vertebral fractures or elements that compress the neuraxis alerts the team of spinal surgeons to the possibility of operating to release neurological elements and to perform reduction and stabilization of spinal injuries.³

Within the spectrum of traumatic spinal cord injuries in adults, there are those without evidence of fracture or vertebral dislocation on radiographs and tomography, called SCIWORA, SCIWOCTET or SCIWORET.⁴⁻⁹ The term SCIWORA is an acronym for Spinal Cord Injury Without Radiological Abnormality and it was coined in 1982 by Pang in pediatric patients evaluated with radiographs and defines traumatic spinal cord injury without radiographic or tomographic evidence of bone injury.^{4,5} Computed tomography and magnetic resonance imaging (MRI) evidenced, in adult patients, the association of non-traumatic injuries linked to the injury mechanism (ligament calcification, degenerative changes, central disc herniation, spinal canal stenosis), suggesting that the acronym SCIWORA was a “misnomer”.^{6,7} In fact, authors with similar considerations proposed alternative terms, such as SCIWOCTET (*Spinal Cord Injury Without CT Evidence of Trauma*) and SCIWORET (*Spinal Cord Injury Without Radiographic Evidence of Trauma*).^{8,9}

The ossification of the posterior longitudinal ligament (OPLL) is a pathological process of lamellar bone deposition in the aforementioned structure.¹⁰ It has a high prevalence in Asian countries, affecting 2% of the Japanese, and represents one of the main causes of narrow cervical canal. For this reason, the publications on acute traumatic spinal cord injury complicated by OPLL are mostly of Asian origin.¹¹⁻²¹

According to our literature search in journals indexed in national and international biomedical databases, we found a single reported case of Hispanic-American origin.²² Consequently, we consider our contribution a relevant experience in the treatment of this serious injury in a non-Asian country.

Our objective is to present a series of cases of cervical spinal cord trauma associated with OPLL without tomographic signs of vertebral fractures and dislocations.

The secondary objective is to carry out a narrative review of the literature on the treatment and postoperative prognosis of this group of patients.

MATERIALS AND METHODS

Case series

We conducted a descriptive study of a series of cases of cervical spinal trauma associated with OPLL, treated by the same surgical team, in two centers: a hospital of the public health system of the Autonomous City of Buenos Aires and a referral center of the same area. The study lasted from January 2012 to December 2019.

As inclusion criteria, we considered patients with acute traumatic spinal cord injury observable on MRI and tomographic evidence of calcification of the posterior vertebral common ligament closely related to the affected spinal area. Patients with evidence of vertebral fractures or dislocations, those referred to another center after the primary revision, and those with incomplete medical records were excluded.

The following study variables were described: age, sex, comorbidities, traumatic history, preoperative time (> 72 h or <72 h), initial neurological status according to the American Spinal Injury Association (ASIA) scale,²³ type of OPLL according to the morphological classification: (A) continuous, (B) segmental, (C) combined, and (D) limited to the disc space (Figure 1),²⁴ extent of myelopathy according to cervical levels (hyperintense image in MRI T2-weighted sequence), treatment performed (surgical or conservative), type of surgery (technique and approach), post-therapeutic neurological evolution (stable, neurological improvement or deterioration) and complications (related to trauma and intervention).

In the description, the individual data, their respective summary and dispersion measures were included. The average and standard deviation were considered for the numerical variables, and the absolute value and the percentage for the nominal ones.

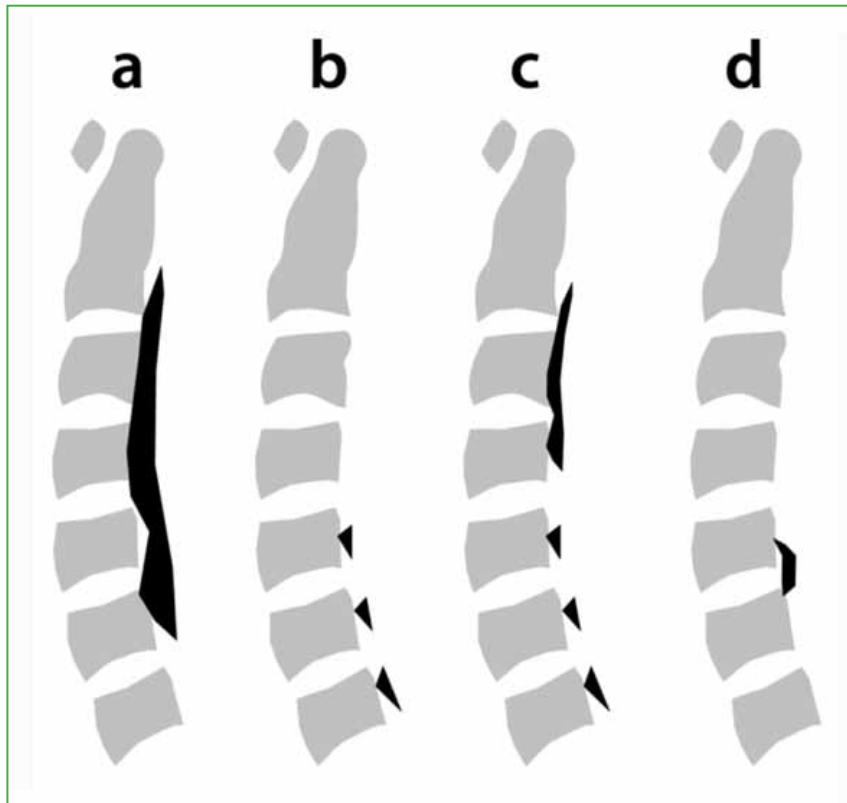


Figure 1. Morphological classification of the ossification of the posterior longitudinal ligament into 4 types: type A or continuous (extended along several vertebrae), type B or segmental (areas of segmented ossification in the posterior wall of the vertebral body), type C or mixed (combination of continuous and segmented) and type D (limited to the disc space).

Narrative review of the literature on the treatment and postoperative prognosis of acute spinal cord injury complicated by OPLL

A literature review was carried out in the main biomedical databases MEDLINE, EMBASE, LILACS, Cochrane Library through the PubMed search engines, VHL portal and Google Scholar, considering articles published between January 2010 and September 2020, using the following keywords and search strategies:

- Strategy 1: (((("cord injury") AND ("ossification of the posterior longitudinal ligament")) AND (cervical)) AND (trauma)) AND (treatment)

- Strategy 2: (((("cord injury") AND ("ossification of the posterior longitudinal ligament")) AND (cervical)) AND (trauma)) AND ("outcomes")

In the PubMed search engine, 22 articles were collected (strategy 1: 20; strategy 2: 10; coincidences: 8). Case reports, studies of patients with associated vertebral fractures or dislocations, and expert opinions (13 articles) were excluded. Nine of all the articles obtained were selected.^{11,18-20,25-29} An additional article was obtained through Google Scholar with the same keywords.³⁰ Finally, 10 articles were included in the review. The following variables were recorded: lead author, year of publication, country of origin, study design and level of evidence.

FINDINGS

Case series

During the study period, seven patients with acute traumatic spinal cord injury and OPLL were included. Two patients were excluded: one due to referral to another center after initial care and one due to death in the first 24 h before treatment, with no data being recorded in the clinical record. A sample of five patients was formed. All were men, with an average age of 62.2 years (standard deviation, 9.36).

As a traumatic history, two (40%) had suffered a fall from their own height; two (40%), a motor vehicle collision and one (20%), an equestrian accident.

All had a severe initial neurological deficit according to the ASIA scale: four (80%) ASIA A and one (20%) ASIA C (central cord syndrome).

As relevant antecedents, three (60%) patients had comorbidities (obesity: 2 cases, high blood pressure and diabetes: 2 cases) and one (20%), had injuries associated with trauma (closed chest trauma); none had a history of myelopathy.

According to the OPLL morphological classification, one case (20%) was type A or continuous (case 1); one (20%), type B or segmental (case 2); one (20%), type C or combined (case 5); two (40%), type D or limited to the disc space (cases 3 and 4) (Figures 2 and 3).

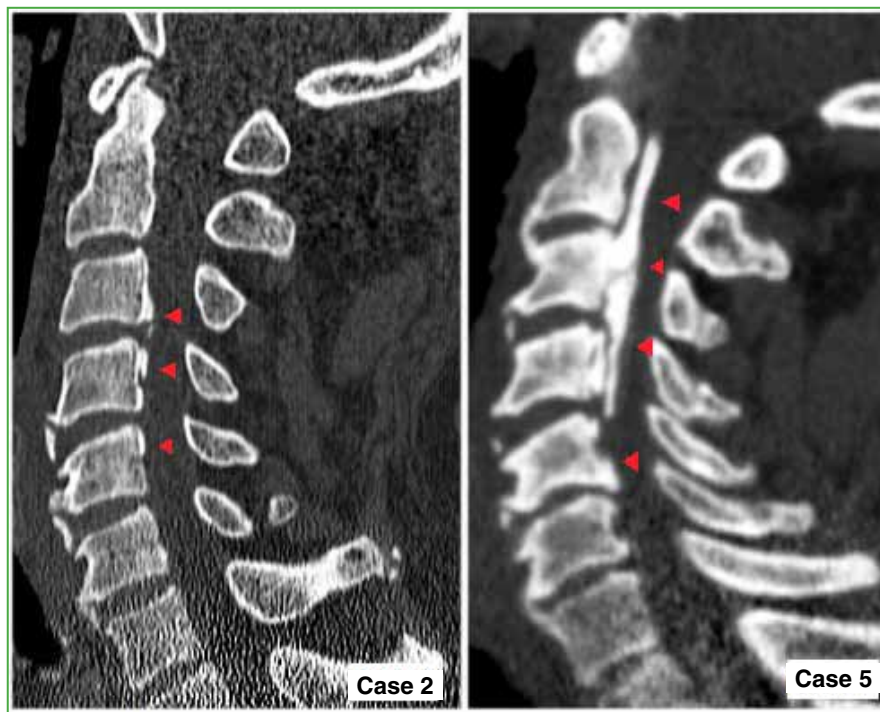


Figure 2. Computed tomography, sagittal plane. Characteristic images of ossification of the posterior longitudinal ligament (arrows), segmental (Case 2) and mixed (Case 5) are observed.

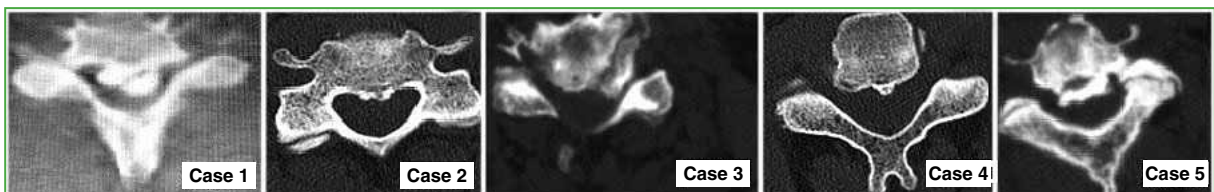


Figure 3. Computed tomography, axial plane. Cases 1-5. Images compatible with ossification of the posterior longitudinal ligament.

All patients had hyperintense spinal topography images on large-area MRI T2-weighted sequence, with involvement of multiple cervical levels ([Figure 4](#)).

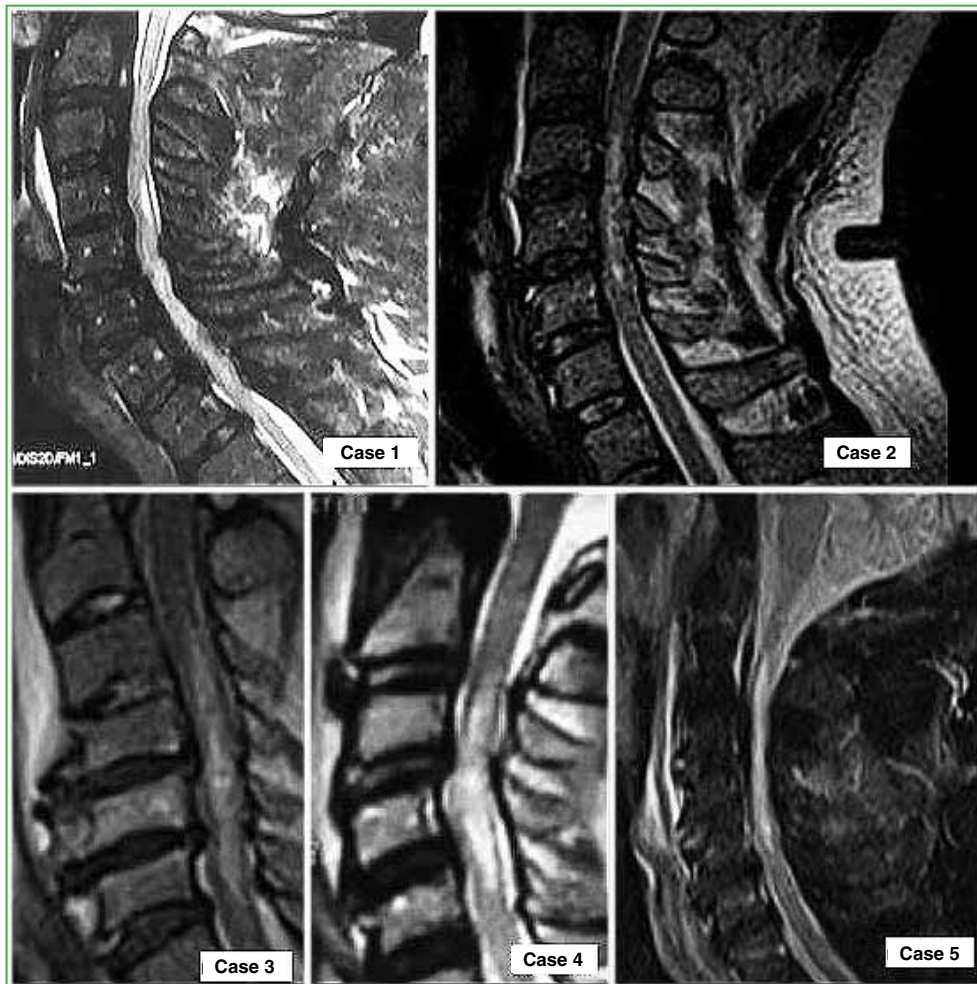


Figure 4. Initial MRI. Cases 1-5. Increased medullary signal in T2-weighted (Case 2) and STIR (Cases 1, 3, 4 and 5) sequences.

Four (80%) patients treated by surgery and one (20%) who received conservative treatment were described. Three were operated on before 72 h and, in the remaining patient, it was necessary to postpone surgery beyond this period due to associated respiratory distress complicated by nosocomial infection. All surgeries were posterior. The surgical techniques implemented were laminoplasty (1 case), laminectomy and arthrodesis (2 cases) and laminectomy without fixation (1 case). The choice of surgical technique was based on the analysis of complementary studies, the availability of implants as soon as possible in the emergency room, and the surgeon's preference ([Figure 5](#)).

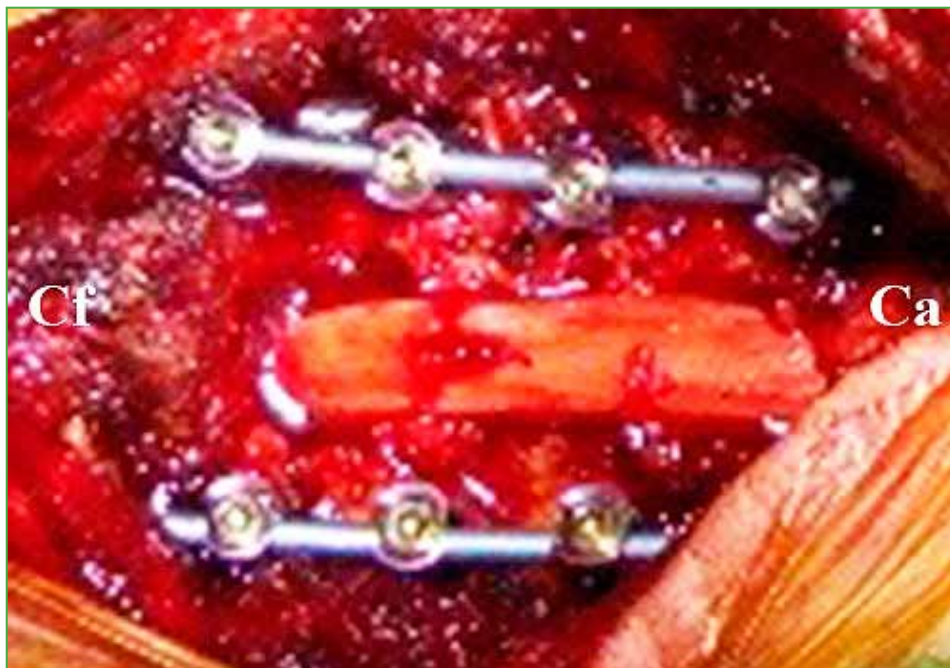


Figure 5. Intraoperative image. Laminectomy and arthrodesis. Cf = cephalic, Ca = caudal.

One patient decided to receive conservative treatment given the risks of surgery and the prognosis of spinal cord injury (case 5, ASIA A).

During the immediate postoperative period, two patients presented significant hemodynamic compromise (neurogenic shock) that caused the death of one of them 48 hours after surgery and the other one week later.

One patient died at six months from a gastrointestinal condition not associated with trauma (perforated diverticulum and fecal peritonitis). The total mortality rate was 60%, 40% (2 cases) died in the immediate postoperative period. Two patients continue in rehabilitation and follow-up to date.

The neurological status did not improve in any case, in an average follow-up time of 3.9 years (± 3.78), in three of the five cases.

All received an intravenous corticosteroid regimen with methylprednisolone upon admission, according to the NASCIS II protocol.

Table 1 summarizes the description of the variables and Table 2, the patient data.

Narrative literature review

Table 3 lists the articles included in the literature review.

Risk factors for acute traumatic spinal cord injury in OPLL patients

Wu et al. conducted a retrospective cohort study of patients with OPLL treated conservatively versus a control group without OPLL over an 8-year follow-up period. The incidence of cervical spinal cord injury was significantly higher in the OPLL patient cohort than in the control group, with an incidence of 4.81 versus 0.18 patients per 1000 people-year. This study did not assess the risk of myelopathy associated with specific aspects of OPLL (type, canal diameter, age of the patients).²⁹

Onishi et al. retrospectively estimated a greater association between OPLL and acute spinal cord injury in elderly patients, those with a mixed or segmental ossification pattern and the additional presence of ossification of the anterior common vertebral ligament.²⁰

Table 1. Series description

Average age (SD)	62.2 (\pm 9.36)
Sex, n (%)	
Male	5 (100)
Female	0 (0)
Traumatic history	
low energy, n (%)	2 (40)
high energy, n (%)	3 (60)
Initial neurological injury, n (%)	
ASIA A	4 (80)
ASIA B	0 (0)
ASIA C	1 (20)
ASIA D	0 (0)
OPLL classification, n (%)	
Type A	1 (20)
Type B	1 (20)
Type C	1 (20)
Type D	2 (40)
Extension of the spinal cord image on magnetic resonance imaging (T2-weighted), n (%)	
From C4 to T1	1 (20)
From C3 to C5	2 (40)
From C2 to C5	1 (20)
From C3 to C6	1 (20)
Surgical treatment, n (%)	
Yes	4 (80)
No	1 (20)
Surgery, n (%)	
Laminoplasty	1 (20)
Laminectomy	1 (20)
Laminectomy and arthrodesis	2 (60)
Neurological evolution, n (%)	
Improvement	0 (0)
Stable	5 (100)
Deterioration	0 (0)

OPLL = Ossification of Posterior Longitudinal Ligament.

Table 2. Patient data

Case	Age	Sex	Trauma	Initial ASIA	OPLL/C	MRI extension (T2) (levels)	Treatment	Surgery	Neurological evolution
1	54	M	Fall from own height	A	A	C4 to T1	Surgical	Laminoplasty	Stable
2	55	M	Fall from own height	A	B	C3-C5	Surgical	Laminectomy and arthrodesis	Stable
3	65	M	Traffic	C	D	C3-C6	Surgical	Laminectomy	Stable
4	77	M	Equestrian	A	D	C3-C5	Conservative	-	Stable
5	60	M	Traffic	A	C	C2-C5	Surgical	Laminectomy and arthrodesis	Stable

COLLP =Ossification of Posterior Longitudinal Ligament Classification, MRI = magnetic resonance imaging, M = male.

Table 3. Articles on the ossification of the posterior longitudinal ligament and acute traumatic spinal cord injury.

Year	Author	Origin	Type of study	Level of evidence
2011	Yan et al. ²⁵	China	Case series	IV
2011	Chikuda et al. ¹¹	Japan	Retrospective observational analysis. Multicenter	III
2012	Onishi et al. ²⁰	Japan	Retrospective observational analysis. Comparison of 3 groups. 1) acute spinal cord injury and OPLL, 2) myelopathy and OPLL, and 3) control	III
2012	Wu et al. ²⁹	Taiwan	Retrospective observational analysis.	III
2014	Choi et al. ²⁸	South Korea	Retrospective observational analysis, correlation.	III
2014	Gu et al. ¹⁸	China	Retrospective observational analysis, comparison of 2 groups.	III
2015	Kwon et al. ²⁶	South Korea	Retrospective observational analysis, multivariate analysis.	III
2016	Gu et al. ¹⁹	China	Retrospective observational analysis.	III
2020	Li and Jiang ³⁰	China	Retrospective observational analysis, multivariate analysis.	III
2020	Hollenberg and Mesfin ²⁷	United States	Retrospective observational analysis, comparison	III

Surgical versus conservative treatment

The treatment of acute traumatic spinal cord injury associated with OPLL remains controversial. There are no published studies with a high level of evidence to guide its therapy.

In a retrospective multicenter study of 34 centers in Japan, Chikuda et al. evaluated 94 patients. Neurological improvement was greater in patients treated with surgery; however, it was not statistically significant. They emphasize that, when the sample was segmented according to the existence of compromise of the gait prior to the trauma (previous myelopathy), in this subgroup, the improvement with surgical treatment was significant.¹¹

In a retrospective study of 60 patients, Gu et al. obtained a higher proportion of cases with neurological recovery in the follow-up of patients treated with surgery compared to those who received conservative treatment.¹⁸

Yang et al. published a series of 25 patients with improved neurological status in 21 of the 25 cases. Twenty patients in the series had Frankel C or D neurological conditions.²⁵

Prognostic factors for the outcome of surgical treatment

In the last decade, five retrospective studies have been published which estimate the prognostic factors of surgical treatment in patients with spinal cord injury complicated by OPLL.

Kwon et al. retrospectively evaluated the factors associated with postoperative neurological recovery in a series of 38 patients treated posteriorly (laminoplasty or laminectomy) over a seven-year period. Using a multivariate analysis, they estimated that advanced age, a lower motor score on the ASIA scale upon admission, the severity of spinal cord hyperintensity in the T2-weighted sequence of the preoperative MRI and a smaller space available for the spinal cord (SAC) were associated with worse neurological outcomes.²⁶

In a study of similar design with 36 patients operated on using different approaches, Gu et al. estimated the presence of an area of hyperintensity of the medullary signal in the T2 sequence of the MRI as the main factor associated with worse outcomes in the postoperative neurological recovery.¹⁹

In a retrospective multivariate analysis of 69 patients with acute traumatic spinal cord injury without evidence of vertebral fracture or dislocation that included 10 patients with OPLL, Li and Jiang did not demonstrate a relationship between the prognosis of spinal cord injury and the presence of OPLL. The significant association factors with postoperative prognosis were: 1) baseline ASIA score (patients with ASIA C and D scores on admission had a higher rate of neurological recovery), 2) length of spinal cord injury on MRI (≥ 45 mm was associated with worse neurological outcomes), 3) the Pavlov relationship (< 0.65 was associated with poor results) and 4) the type of spinal cord injury on MRI (the coexistence of bleeding and edema was associated with worse outcomes).³⁰

Choi *et al.* compared the results obtained with posterior release surgery (laminectomy or laminoplasty) in patients with chronic cervical myelopathy and those with a history of low-energy trauma as a trigger. In their sample, a traumatic history was not significantly associated with worse outcomes. The factors that had a significant association were: preoperative neurological status, magnitude of spinal cord compression, diabetes mellitus, and increased spinal signal on the initial MRI.²⁸ Recently, Hollenberg *et al.* conducted a similar study in a cohort of North American patients and reported, as a significant finding, worse initial and postoperative motor neurological values in patients with acute spinal cord injury compared with those with myelopathy.²⁷

DISCUSSION

We presented a series of five cases of cervical spinal cord trauma associated with OPLL that, although it represents a low number of patients compared to previous publications, has as its only precedent a case report of this association by Spanish-speaking authors.²² As an additional noteworthy antecedent, Rendó *et al.* published a case of OPLL-associated myelopathy.³¹

In our setting, the prevalence of OPLL in acute traumatic spinal cord injury without evidence of fracture is unknown. According to Asian publications, 34% of these injuries are associated with OPLL.¹⁰ Bazan *et al.* reported a series of 13 patients with acute traumatic spinal cord injury without radiographic and tomographic signs of trauma, none of them had OPLL.³²

The clinical profile of this condition includes elderly patients with a minor traumatic history.¹¹ Two of our cases had suffered falls from their own height as a traumatic event.

According to the literature consulted, patients with OPLL have a higher risk of suffering a traumatic spinal cord injury, in addition to the possible development of chronic myelopathy.²⁹ The traumatic history would not have a direct impact on the results compared to other factors that are known to have a significant association, such as the severity of the initial neurological state, advanced age, increased intensity of the spinal cord signal on MRI and magnitude of compression.^{19,26-29} The suggested treatment is early surgical release, and reports of the posterior approach (laminectomy or laminoplasty) predominate.^{11,18-20,25-30}

In our series, emergency surgical treatment did not provide benefits based on neurological recovery and the mortality rate in the immediate postoperative period was high. It should be noted that all the included cases had a high ASIA score upon admission (4 ASIA A and 1 C). Clinically, four patients had a complete spinal cord injury and one had a central cord syndrome. Likewise, in accordance with the factors associated with worse outcomes in the literature, all of our cases were associated with extensive hyperintense spinal cord images on preoperative MRI and evidence of significant spinal canal stenosis.

The main weaknesses of our study are its descriptive nature and the low number of cases, which is why the contribution of the data of the series is limited. However, we consider it a novel report in the Hispanic-American literature on a rare association in our region.

CONCLUSION

We presented a series of patients with cervical spinal cord trauma associated with OPLL, which is rare in our setting; there is only one case report registered in the Spanish-American literature. The recent literature considers surgical release via the posterior route in time as the predominant treatment. Severe initial neurological injury, advanced age, increased intensity of the spinal cord signal on MRI, and high spinal cord compression are associated with poor postoperative outcomes.

Conflict of interests: The authors declare they do not have any conflict of interests.

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