

# Symptomatic Triple-Region Spinal Stenosis: Case Report and Narrative Literature Review

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## ABSTRACT

Simultaneous spinal stenosis across all three regions of the spine is an uncommon condition that requires proper clinical and imaging assessment. There are currently no established guidelines for its diagnosis and treatment. The objective of this paper is to describe, based on a case report, the clinical presentation, treatment, and progression of triple stenosis in a patient and compare it with available evidence through a narrative literature review. A 69-year-old woman presented with progressive paraparesis associated with right sciatica and positive signs of upper motor neuron involvement. Imaging confirmed triple stenosis: cervical, thoracic, and lumbar. She underwent thoracic decompression and tumor resection, as well as conservative treatment for cervical and lumbar stenoses, and showed favorable progress one year after surgery.

Symptomatic triple-region spinal stenosis is an uncommon condition. Proper clinical and radiological assessments will enable an accurate diagnosis with appropriate and timely intervention.

**Keywords:** Symptomatic triple-region spinal stenosis; tandem spinal stenosis; cervical, thoracic, and lumbar stenosis.

**Level of Evidence:** IV

## Estenosis espinal de triple región sintomática: reporte de un caso y revisión narrativa de la bibliografía

## RESUMEN

La estenosis espinal de las tres regiones de la columna en simultáneo es un cuadro infrecuente que requiere una adecuada valoración clínica y de las imágenes. En la actualidad, no existen guías establecidas para su abordaje diagnóstico y terapéutico. El objetivo de este artículo es describir la presentación clínica, el tratamiento y la evolución en un paciente con triple estenosis y contrastarlos con la evidencia disponible a través de una revisión narrativa de la bibliografía. Se presenta a una mujer de 69 años de edad que consultó con un cuadro de paraparesia progresiva asociado a cialgia derecha y signos de motoneurona superior positivos. En los estudios por imágenes, se constató una triple estenosis: cervical, torácica y lumbar. Se procedió a la descompresión y la resección tumoral torácica asociadas al tratamiento conservador de las estenosis cervical y lumbar. La evolución era favorable al año de la cirugía. La estenosis espinal de triple región sintomática es una condición rara, las valoraciones clínicas y radiológicas adecuadas permitirán un diagnóstico correcto con un abordaje adecuado y oportuno.

**Palabras clave:** Estenosis espinal de triple región sintomática; estenosis espinal en tándem; estenosis cervical, torácica y lumbar.

**Nivel de Evidencia:** IV

## INTRODUCTION

Tandem spinal stenosis (TSS) is defined as synchronous narrowing of the diameter of the spinal canal in at least two regions of the spine, with a prevalence ranging from 0.2% to 11% in imaging studies.<sup>1,2</sup> Clinically, it manifests with associated upper and lower motor neuron symptoms. Dagi et al. described a typical triad characterized by: 1) claudication and loss of lower limb strength; 2) gait disturbance; and 3) upper motor neuron signs, such as hyper-reflexia, clonus, and positive Babinski sign.<sup>3</sup>

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**How to cite this article:** Latallade V, Pereira Duarte M, Gamsie S, Kido GR, Petracchi MG, Gruenberg MF. Symptomatic Triple-Region Spinal Stenosis: Case Report and Narrative Literature Review. *Rev Asoc Argent Ortop Traumatol* 2024;89(1):64-69. <https://doi.org/10.15417/issn.1852-7434.2024.89.1.1862>

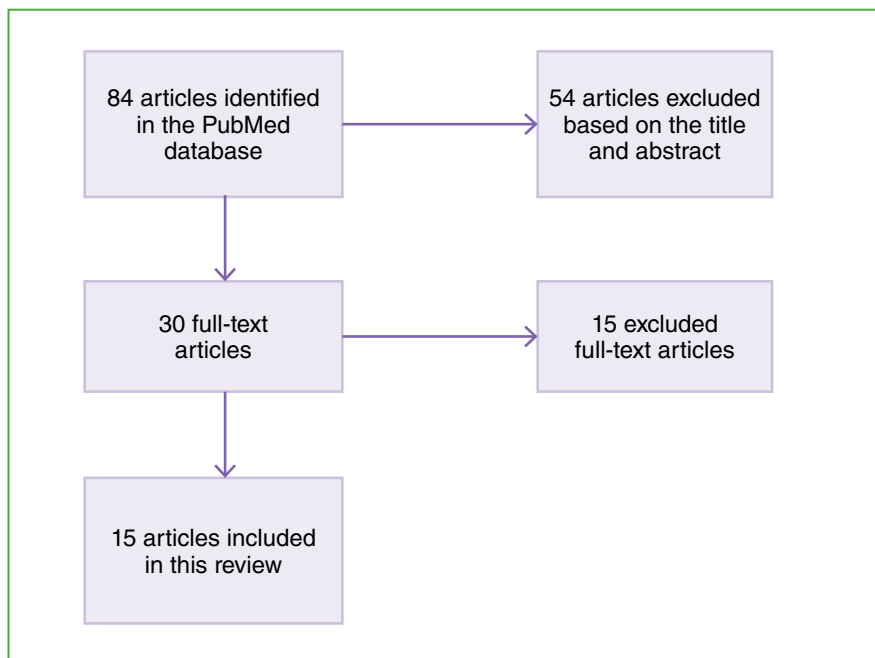
Evidence regarding the treatment of TSS is scarce and the casuistry is even more limited when the stenosis occurs in three segments synchronously, termed symptomatic triple-region spinal stenosis (TRSS). Its low frequency, combined with the variety of possible signs and symptoms, creates an extremely complex picture, making diagnosis and resolution challenging and presenting numerous therapeutic alternatives (single or multiple region decompression, in stages or simultaneously).<sup>4</sup>

The aim of this article is to describe the clinical presentation, treatment and evolution in a patient with triple stenosis and to contrast them with the available evidence by means of a narrative review of the literature.

This study had the prior approval of the Ethics Committee of our institution. A literature search was performed in PubMed, Web of Science, Scopus and Lilacs databases using the following terms: “Tandem spinal stenosis”, “Tandem stenosis” or “Cervical, thoracic and lumbar stenosis”.

All case reports, case-control studies, cohort studies, and reviews published in the last 10 years, evaluating adult patients with TSS and TRSS, and a minimum follow-up of one year, were included. No reports of similar cases were found in Latin American databases. Articles that did not evaluate spinal disease, included pediatric patients or disease of a single region were excluded.

Fifteen articles were selected for this narrative review. **Figure 1** shows the details of the literature search and the selection of articles.



**Figure 1.** Flow chart of the bibliographic search.

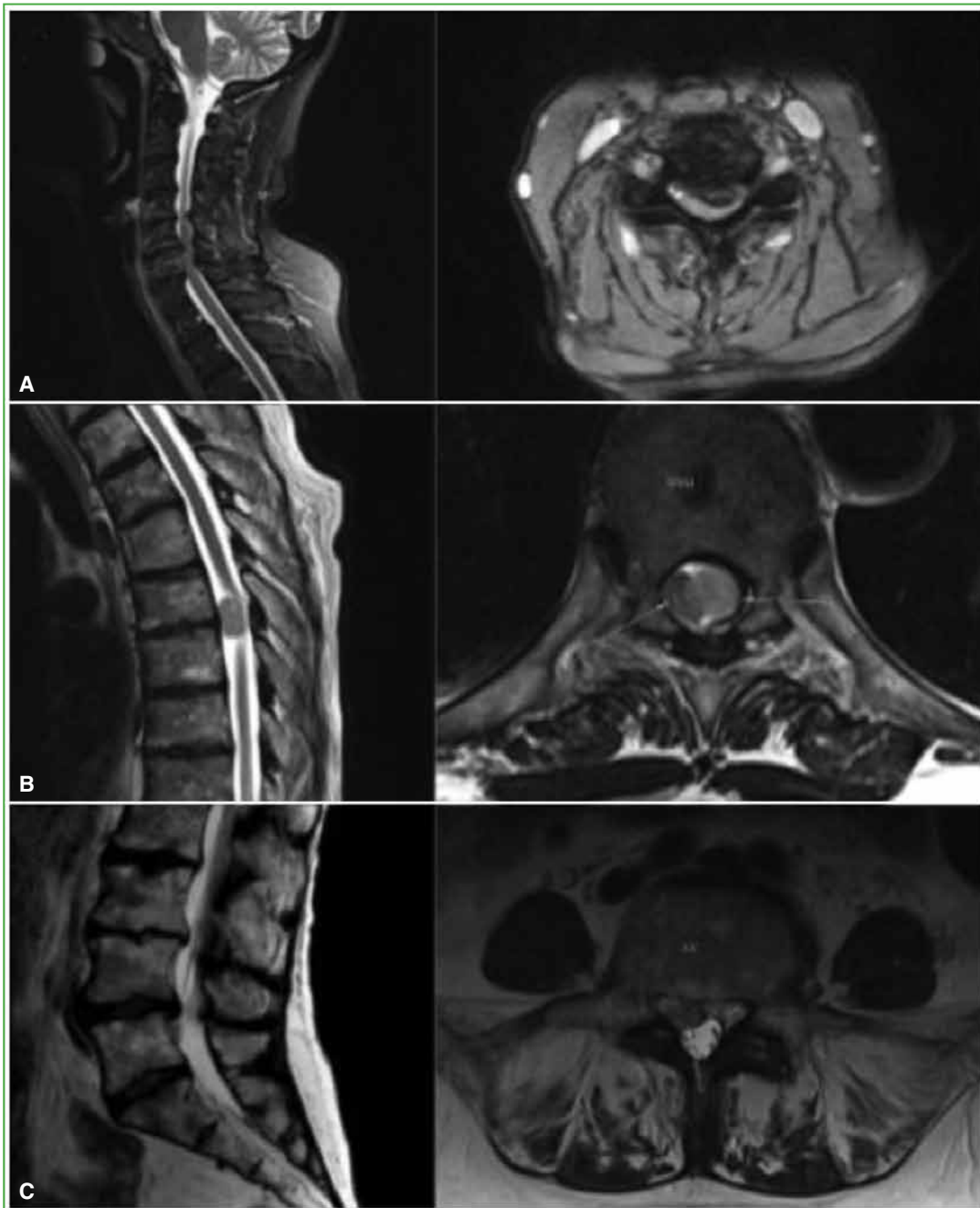
## CLINICAL CASE

A 69-year-old woman, with no relevant medical history, consulted the emergency room for progressive and incapacitating paraparesis (grade C on the ASIA scale,<sup>5</sup> score 5 on the Nurick scale<sup>6</sup>) of one year of evolution associated with right sciatica.

Physical examination revealed paraparesis with motor strength M3/5<sup>7</sup> in both lower limbs from L2 to S1, with bilateral patellar and achilles hyperreflexia, negative Hoffman sign, positive Babinski sign and clonus in the lower limbs, together with right sciatica of 9/10 intensity according to the visual analog scale in the territory of the L5 root, and positive Lasègue and Bragard signs. Cervical myelopathy scores were: 9/17 on the modified Japanese Orthopaedic Association scale<sup>8</sup> and 5 on the Nurick scale.

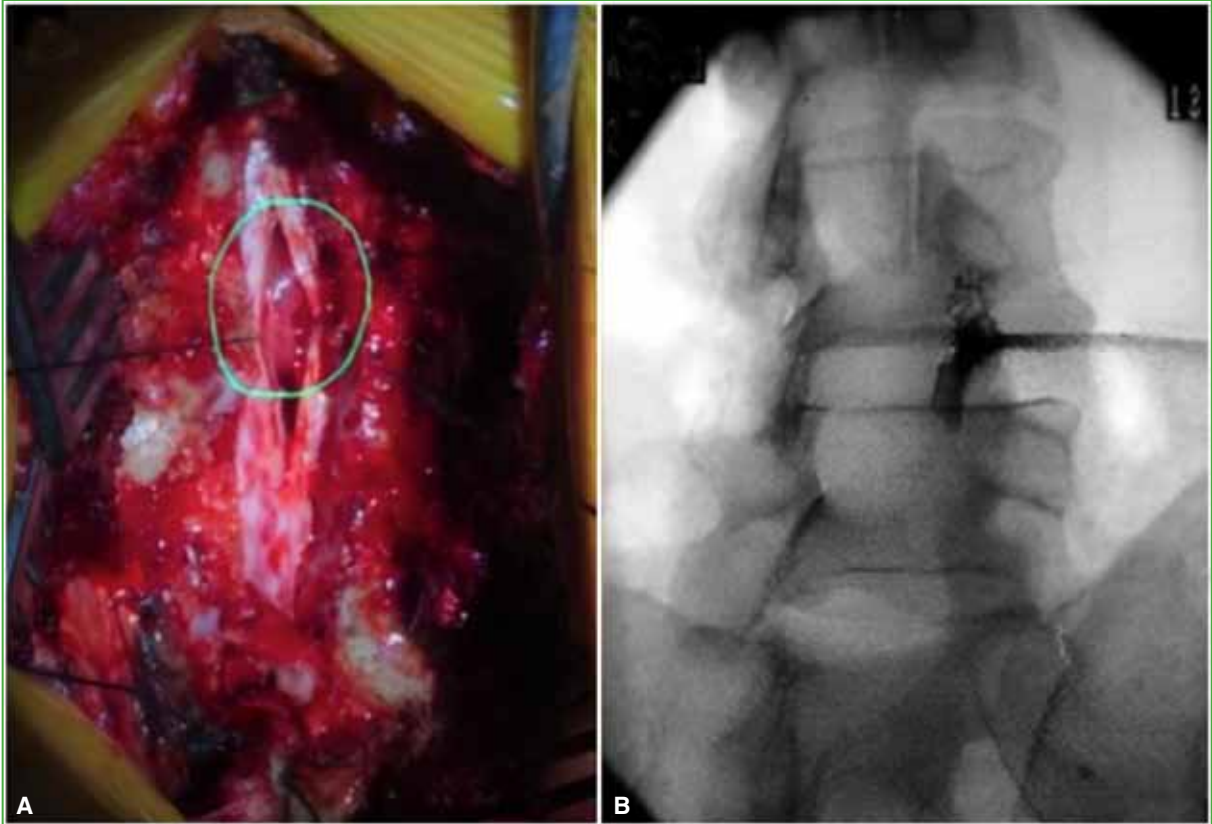
### Imaging studies

In an emergency MRI, three points of stenosis of the spinal canal were detected: 1) a degenerative narrow cervical canal at C5-C6 with focal myelomalacia; 2) an intradural extramedullary tumor at T6-T7 impacting the spinal cord, measuring 15 mm x 8 mm x 7 mm; and 3) a right posterolateral disc extrusion of L4-L5 (Figure 2).



**Figure 2.** Spinal MRI without contrast. **A.** Sagittal and axial slices of the cervical spine showing left posterolateral C5-C6 stenosis due to disc protrusion with signs of myelomalacia. **B.** Sagittal and axial slices of T6-T7 with an intradural-extramedullary tumor. **C.** Sagittal and axial lumbar slices with right posterolateral disc extrusion at L4-L5.

Once the diagnosis of TRSS was confirmed, after four days of hospitalization, it was decided to perform a posterior decompression of T6-T7 by laminectomy and excision of the thoracic intradural extramedullary tumor, and a right intraoperative block of L4-L5 (Figure 3).



**Figure 3.** **A.** Intraoperative image of the same patient. The opening of the dura mater with an intradural extramedullary dorsal tumor is observed through a central T6-T7 laminectomy. **B.** Intraoperative fluoroscopic image of the right L4-L5 periradicular block.

Surgery was performed under general anesthesia, with intraoperative monitoring of somatosensory and motor evoked potentials, and lasted 2 hours and 20 minutes. Pathological examination revealed a WHO grade I transitional meningioma.<sup>9</sup>

The evolution was favorable during the immediate postoperative period, with remission of the sciatica (3/4 on the visual analog scale). On the third day after surgery, the patient moved with a walker and was discharged.

At the day 7 and day 14 follow-up visits, there was a notable improvement in motor strength and a 2/10 level of sciatica; after one month, the patient was able to walk with a cane, and three months after surgery, she was no longer using it. One year after surgery, at the last follow-up, residual pain of 1/10 intensity was detected, with an M4 deficit for right L5, no gait instability and decreased upper motor neuron signs, with scores of 17/17 on the modified Japanese Orthopaedic Association scale, and 1 on the Nurick scale.

## DISCUSSION

We present the case of a patient who consulted for progressive spastic paraparesis associated with right sciatica and a TRSS diagnosis, and underwent surgery for one of the lesions, which resulted in a good postoperative evolution and almost complete remission of symptoms.

The term TSS was introduced by Dagi et al. in 1987 to describe concurrent symptomatic cervical and lumbar spinal stenosis.<sup>3</sup>

In 2016, Uehara et al. classified TSS into four subtypes according to region: cervicothoracic, thoracolumbar, cervico-thoraco-lumbar, and cervicolumbar.<sup>10</sup>

Our patient had a cervico-thoraco-lumbar TSS, a rare condition, as most reports address tandem stenosis with compression at two levels only.

The clinical manifestations of TSS are highly variable and depend on the location of the stenotic areas and their severity. LaBan et al. and Kenneth et al. reported difficulty in reaching a correct diagnosis due to the combined clinical presentation of upper and lower motor neuron symptoms.<sup>11,12</sup> In their series of 33 patients, Bhandutia et al. reported a late diagnosis rate of 45%, which had serious consequences on outcome.<sup>13</sup>

Epstein et al. noted that the correct order for surgical treatment of TSS depended on the severity of myelopathy and radiculopathy.<sup>14</sup> In their series, cervical decompression improved lumbar radicular symptoms. Similarly, in our patient, thoracic decompression may have favored symptomatic improvement associated with lumbar periradicular block. We hypothesized that cervical stenosis, as a chronic degenerative process, may have developed adaptive mechanisms that prevented it from manifesting clinically, but the tumor, with increased compression velocity, would have been responsible for nearly all upper motor neuron symptoms.

Most published reports address TSS with compression at the cervical and lumbar levels. We found few case reports of TSS involving the thoracic region, and none with an associated tumor. Chen et al. and Hu et al. published retrospective reviews of 15 and 16 cases, respectively, on cervicothoracic tandem ossification.<sup>15,16</sup> Both reported surgical resolution using a single posterior incision that achieved significant clinical improvements, but at the expense of a high intraoperative and postoperative complication rate.

Finally, Schaffer et al. published a case of TRSS treated with simultaneous surgery of the three regions, and warned that this may be associated with a prolonged hospital stay.<sup>17</sup> Jannelli et al. presented a patient with TRSS who underwent staged decompression, first cervical, then lumbar, and finally thoracic, with a final improvement of the paraparesis that was delayed by two and a half years.<sup>18</sup> In this last case, initially, only images of the cervical spine were taken. In our patient, however, due to the disparity between clinical and imaging findings, it was considered of vital importance to order an MRI of the complete spine.

Unlike what is described in the literature, in our case, we started with the thoracic region because the tumor was causing severe stenosis, which was compatible with the patient's symptoms. The results were an early hospital discharge and a very good postoperative evolution, with a significant improvement of the neurological deficit.

This article is a narrative review of the literature on triple tandem spinal stenosis based on a case report. Its limitations are mainly related to the low incidence of this disease, since almost all the bibliographic citations included are case reports and small series of patients, as well as its retrospective nature and low level of evidence. In addition, the modality of reporting of these cases and series of patients is very heterogeneous, each publication emphasizing a different aspect of the subject. Nevertheless, this is, to the best of our knowledge, the first narrative review on the subject and we believe we have developed each of the aspects inherent to this rare entity in a harmonious and engaging manner.

## CONCLUSIONS

We present a patient with TRSS and progressive neurological compromise, who required thoracic decompression, and had a favorable evolution. Although multiple stenosis is a rare condition, diagnostic suspicion through a correct clinical assessment allows for timely treatment and monitoring. TRSS should be considered within the differential diagnoses in patients with signs and symptoms of spinal and upper and lower motor neuron stenosis.

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Conflict of interest: The authors do not declare conflicts of interest.

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