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Case Report

Ossification of the Ligamentum Flavum: Thoracic Myelopathy in Non-Oriental Patient. Case report

Mielopatia Torácica por Ossificação do Ligamento Amarelo em Paciente não Oriental. Relato de caso

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ABSTRACT

Ossification of the ligamentum flavum (OLF) of thoracic spine occurs along with a progressive spine lesion, and is a rare myelopathy in non-Oriental patients. Computed Tomography or Magnetic Resonance imaging of spine are used for diagnosis, and early decompressive laminectomy is the treatment of choice. We report a case of female patient, 73-year-old, who presented progressive and symmetrical paraparesis, and painful tactile hypoesthesia at T4 level with two months of evolution of the case. After OLF diagnosis, with myelopathy on T1-T2 segment, we performed spine decompression with significant functional better results. According to our research and knowledge, this is the third case to be reported on Brazilian medical literature of a patient with non-Oriental descent.

Key words: *Ligamentum flavum; Spinal cord; Spine disease; Spinal stenosis*

RESUMO

Ossificação do ligamento amarelo (OLA) da coluna torácica é causa rara de mielopatia em pacientes não orientais, que cursa com lesão medular progressiva. O diagnóstico é feito através de tomografia computadorizada ou ressonância magnética da coluna vertebral. Laminectomia descompressiva precoce é o tratamento de escolha. Relata-se o caso de uma paciente de 73 anos, que apresentou quadro progressivo de paraparesia simétrica e hipoestesia tátil e dolorosa com nível sensitivo em T4 com 2 meses de evolução. Após o diagnóstico de OLA com mielopatia no segmento T1-T2, realizou-se descompressão medular com importante melhora funcional. Acredita-se tratar do terceiro caso relatado na literatura brasileira de uma paciente não descendente oriental.

Palavras-chave: *Ligamento amarelo; Medula espinhal; Doenças da coluna vertebral; Estenose espinal*

Introduction

Ossification of the ligamentum flavum (OLF) is an uncommon cause for thoracic myelopathy which demands surgical treatment. This condition usually is described in Far East countries, fairly common in Japanese people[1], even though rare in Western countries[2-5]. Two cases were reported in Brazil[4,5]. Polgar was the first to describe OLF, in 1929 [6], and Yamaguchi et al. [7], in 1960, reported the first case of OLF causing thoracic myelopathy. Prevalence of thoracic OLF is 3.8% in asymptomatic patients[8], and up to 63.9% in symptomatic patients [9]. After Computer Tomography (CT) and Magnetic Resonance imaging (MRI) advent, this disease could be better understood. CT is the golden standard exam for OLF diagnosis.

Although physiopathology still remains not well understood, the calcification in the posterior portion of vertebral canal can emerge as secondary to a mechanical stress, progressing to ligament hyperplasia until its replacement by a compact lamellar bone [10].

The main signals and symptoms involve sensitive and motor compromise, below the affected segment. Along with the disease

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evolution can occur lower limbs spastic paralysis, hyperreflexia and sphincter dysfunction. In general, treatment demands surgical approach due to the progressive nature of symptomatology and absence of response to conservative treatment [2].

This report is the third case of thoracic myelopathy due to ossification of ligamentum flavum described in Brazil.

Case Report

Female patient, 73-year-old, non-Oriental descendant, presented a spastic progressive paraparesis, reporting falls and significant gait ataxia since two months ago. She also complained of sensitive disturbance of lower limbs which progressed to walking incapacity a month after admission at the hospital. Due to severe joint arthrosis, she reported a previous surgery for ankle prosthesis placement on the left side, a year ago. Also, the patient presented diabetes mellitus. On physical exam were found paraparesis Grade II, spasticity, lower limbs hyperreflexia, painful tactile hypoesthesia on T4 level, proprioceptive disturbance on lower limbs, and Babinski's sign to the right. On clinical evaluation 4 points were reached from a total of 11 according to the modified scale of Japanese Orthopedics Association (mJOA).

T2-weigthed sequence of MRI sagittal images depicted hypointense signal in the posterior portion of the vertebral canal extending from C5 to T4. Moreover, it was observed changes on degenerative discs and rectification of the cervical spine. Axial views also showed spinal cord compression, more evident on T1-T2 segment (Figure 1). CT-Scan confirmed the ligamentum flavum ossification, as well as the spinal canal stenosis increased on T1-T2 segment (Figure 1). These aspects corroborated to the clinical condition of the patient.

Posterior surgical spinal decompression of T1-T2 segment was proposed to the patient (Figure 2). At first, was performed a midline incision, with subperiosteal dissection of paraspinal musculature and T1 and T2 laminas were exposed. After laminectomy was observed a hardened and calcified ligamentum flavum, adhered to dura mater (Figure 3). Cautious dissection was performed and ligamentum flavum was resected on all extent of laminectomy. Due to severe adherence of ossified ligamentum to the dura mater, occurred a small dural injury which was promptly corrected with muscle fascia flap.

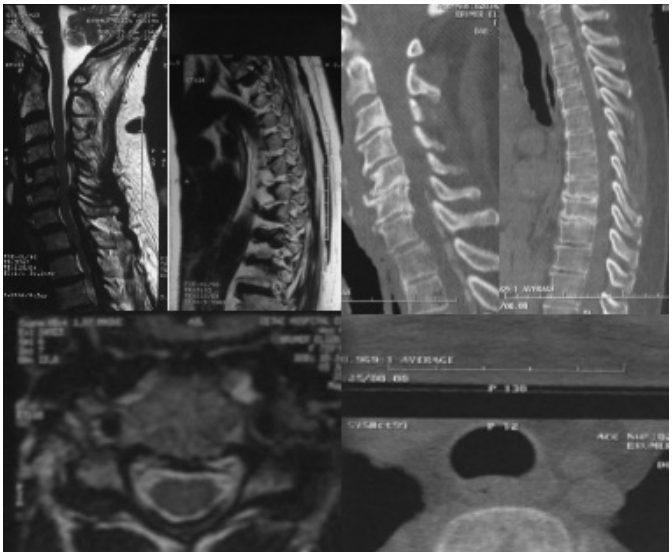

Figure 1. A. MRI with hypointense signal in the longitudinal posterior portion of the vertebral canal in cervicothoracic transition on sagittal T2 sequence. **B.** Hyperdensity of posterior portion of spinal canal resembling a bone, evidence of ossification of ligamentum flavum on cervicothoracic transition.

Figure 2. Spine X-ray and CT depicting laminectomy on T1-T2. Ligamentum flavum resection with ossification, evidence of spinal canal enlarged and with no stenosis in this segment.

In the immediate postoperative period, the patient referred tactile and proprioceptive sensitivity improvement in the lower limbs. On the first day of post-op, the patient ambulated, still with difficulty, but without the need for support aid. According to the mJOA scale, she evolved functionally to 8 points for a total of 11.

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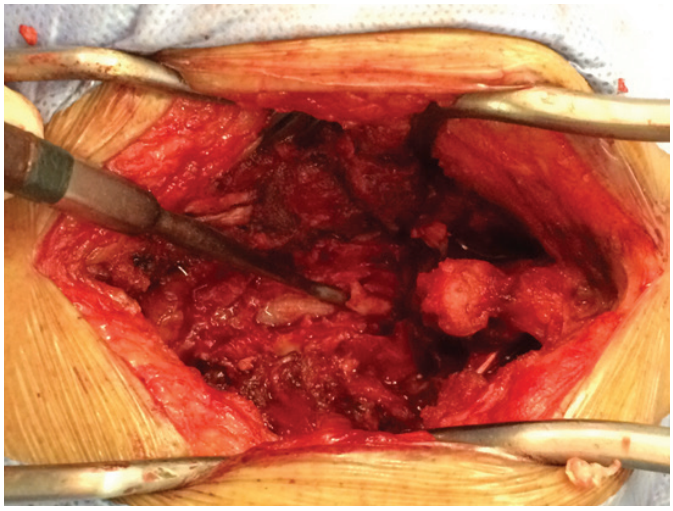

Figure 3. Laminectomy of T1 and T2 showing calcified ligametum flavum removal, and exposure of thecal sac.

Discussion

Since the first report by Polgar, in 1929 [6], a few cases of myelopathy due to OLF were reported. The majority (more than 90%) of diagnosis was in Japanese patients[1]. Myasaka et al. showed OLF occurrence up to 20% of individuals > 65 years of age, and was considered as a normal feature of aging[11]. In a study recruiting 1,736 healthy volunteers of Southern China, Guo et al. found an incidence of 3.8% of thoracic OLF [8]. Lang et al. analyzed retrospectively CT images from 993 patients with thoracic pain symptoms, and 63.9% had OLF diagnosis, but no significant spinal stenosis [9]. Ossification generally occurs as a lesion, oval shaped, in the posterior spinal canal. In the physiopathological view, it can be secondary to mechanical stress. Patients with genetic or systemic predisposition evolve to ligamentous hyperplasia, vascular neoformation, cartilaginous metaplasia, ossification, and finally replacement by compact lamellar bone [10].

Okada et al. [12] reported major incidence in the lower third of thoracic spine, at T10-T12 level, and, in second place, upper thoracic spine (T1-T4)[13]. Image diagnosis can be performed using CT scan which shows intense ossification of ligamentum flavum, hyperattenuation of the bone[13]. Therefore, CT scan reveals location, morphology, spinal canal invasion grade, and dural ossification extent [2]. According to Sato's classification, there are 5 types of OLF which are shown in CT images, lateral, extended, enlarged, fusioned and tuberous [11]. On MRI images, OLF shows low signal intensity in T1- and T2-weigthed sequences, oval or beak-shaped, protruding the spinal canal. This is used to identify all the segments involved and signal changes in spinal cord, thus identifying multi-segment lesions[13].

In the majority of studies, the most important prognostic factors are severity of pre-op symptoms, and the period of time before surgical treatment. Therefore, earlier diagnosis and immediate surgical intervention with posterior decompressive laminectomy, and OLF exeresis are essential to a better prognosis. After decompression, the result can be notably favorable in the absence of signal change of spinal cord on MR image [14]. Some authors describe a rapid neurological improvement in all patients after surgery[2,15,16]. Posterior decompression and, consequently, the preservation of anterior spinal circulation, have been suggested as the motive for a rapid postoperative recovery.[2] Surgical result was evaluated using the modified scale of the Japanese Orthopaedic Association (mJOA)[16] generating a recovery rate for the difference of pre-and post-operative score. In the case we described above, the patient did not ambulate in the previous 30 days, with mJOA 4. In the first day post-decompression, walked with significant functional recovery, mJOA of 8, a recovery score of 57%, which was considered as good. A recovery score of 75-100% was considered as excellent; 50-74% as good, 25-49% as regular, and 0-24% as poor [16]. Poor prognosis is indicated by long duration of pre-op symptoms, as well as compressive level, and spinal cord compromising [14]. Yet, other studies observed radiological progression in 36%-86%, with clinical recurrence of compressive myelopathy in 2%-53% of the cases[17].

Another treatment option, which has been advocated by some authors, is the laminoplastia or laminectomy with lateral fusion, due to late neurological deterioration on OLF recurrence on the same site or because of kyphotic deformity of spine [17].

In our case, the radiological finding had extensive OLF of seven spinal segments in the cervicothoracic transition, and we opted for decompression in the level of higher stenosis and according to the patient's clinical evaluation. The patient achieved an important functional improvement. If we had chosen laminectomy and OLF resection, in all affected segments, it would lead to higher surgical morbidity and fixation would be needed. On the other hand, Yu et al. performed decompression and OLF resection through posterior decompressive laminectomy in 49 patients. The overall average increase in kyphosis was only 2.9° and none of the patients required additional surgery due to spinal deformity[16].

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The dura mater might be manipulated very carefully to avoid CSF leakage and damage to spinal cord in intraoperative period. In locations with greater adherence, the ligamentum flavum can be can excised with the outer layer of the dura mater, leaving its inner layer intact. This technique rarely leads to CSF loss and fistula formation[15]. The pathophysiological mechanisms of the OLF and the reasons of its peculiar prevalence in Eastern countries are mostly unknown. Since the scarcity of papers describing this condition in non-Asian patients, we believe it is important to report this case, besides the fact that it is the third case reported in Brazilian medical literature with OLF and non-Asian descent.

Conclusion

The OLF is one of the rare causes of thoracic myelopathy in patients of Western origin and should be remembered as a cause. Spine CT is the exam of choice for a better visualization of ligamentum flavum ossification. The early diagnosis and enough surgical decompression can improve the patient's functional achievements.

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