Initial Clinical Signs of Naka's Grade III Lumbar Epidural Lipomatosis: A Case Series

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ABSTRACT

Introduction: Spinal epidural lipomatosis (SEL) is a rare pathology characterized by the overgrowth of nonencapsulated adipose tissue within the epidural space. This generates spinal stenosis, which might result in compression symptoms. The typical presentation is insidious pain that lasts months or years. The objective of this study was to carry out a descriptive analysis of the initial clinical signs of patients with Naka's grade III Lumbar SEL. **Materials and Methods:** Retrospective observational study in the Spinal Pathology Unit of 4 institutions, from 2010 to 2023. Patients over the age of 18, of both sexes, who consulted for low back pain with or without radiation and presented Naka's grade III lumbar lipomatosis on magnetic resonance imaging (MRI) were included. **Results:** We included 40 patients, with a mean age of 62.5 years; 75% were obese, there were no smokers. The most frequent reason for consultation was low back pain, with a median duration of 5.5 months. **Conclusion:** The most common reason for consultation was low back pain, with the exception of L3-S1 level involvement, which caused lumbar pain with radiation to the thigh. Patients with a longer period of pain (>6 months) were younger and had a lower BMI; although this was not statistically significant.

Keywords: Lumbar epidural lipomatosis, low back pain, clinical signs
Level of Evidence: IV

Manifestación clínica inicial de la lipomatosis epidural lumbar grado III de Naka: serie de casos

RESUMEN

Introducción: La lipomatosis epidural espinal es una enfermedad infrecuente caracterizada por el sobrecrecimiento del tejido adiposo no encapsulado dentro del espacio epidural. Esto genera una estenosis del conducto espinal que puede provocar sintomatología compresiva. La presentación típica es insidiosa a lo largo de meses o años. El objetivo de este estudio fue realizar un análisis descriptivo de la manifestación clínica inicial en pacientes con lipomatosis epidural espinal grado III de Naka. **Materiales y Métodos:** Estudio observacional retrospectivo en la Unidad de Patología Espinal de 4 instituciones, de 2010 a 2023. Se incluyó a pacientes >18 años, de ambos sexos, que acudieron por dolor lumbar con irradiación o sin irradiación, y presentaban lipomatosis lumbar Naka III en la resonancia magnética. **Resultados:** Se incorporó a 40 pacientes (edad promedio 62.5 años). El 75% era obeso, ninguno era fumador. El motivo de consulta más frecuente fue lumbalgia, con una mediana de evolución del dolor de 5.5 meses. **Conclusión:** La lumbalgia fue la consulta más frecuente, con excepción del compromiso de L3-S1 que fue la lumbocruralgia. Los pacientes que sufrieron dolor más tiempo (>6 meses) eran más jóvenes y tenían un índice de masa corporal menor; sin embargo, no resultó estadísticamente significativo.

Palabras clave: Lipomatosis epidural lumbar; lumbalgia; manifestaciones clínicas.
Nivel de Evidencia: IV

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INTRODUCTION

The epidural space is located between the dural sac and the spinal walls. It is a virtual space occupied by fat and veins extending from the foramen magnum to the distal end of the spinal canal.[1-3] Spinal epidural lipomatosis (SEL) is a rare disease characterized by overgrowth of unencapsulated adipose tissue within the epidural space. This generates spinal canal stenosis, which can cause compressive symptomatology.[3-5]

The first case of symptomatic SEL was described by Lee et al.[5] in 1975 in a kidney transplant patient who had received prolonged corticosteroid treatment. The prevalence is 1.1-21%, it predominates in males and patients with obesity, and is infrequent in the pediatric population.[4,6-10] The highly variable incidence is probably due to different diagnostic criteria. Borré[8] describes a mild overgrowth as SEL grade 1, while in more recent studies, the diagnosis is stricter, with SEL being considered in the presence of an evident adipose overgrowth.[6,7,8]

It may be idiopathic or associated with an excess of endogenous or exogenous corticosteroids. The most common form (26-50%) is associated with prolonged treatment with corticosteroids. Some conditions related to corticotherapy are: organ transplantation, autoimmune diseases, multiple sclerosis, diabetes mellitus, ulcerative colitis, and Crohn's disease.[7,10-15] The excess of endogenous corticosteroids causes a low percentage of SEL (3%), and is associated with diseases that generate hypercortisolemia, such as Cushing syndrome, hypothyroidism, prolactinoma, etc. The idiopathic form (17%) is that of unknown cause, in patients who were not exposed to excess corticosteroids, nor have obesity. Obesity is the most common cause of SEL when corticosteroid use is excluded (24.5%).[10-16]

The typical presentation is insidious over months or years. Lumbar or thoracic pain usually precedes the rest of the symptoms; exceptionally, patients may develop acute symptoms, and present with compressive symptoms, such as myelopathy, radiculopathy and, on rare occasions, as cauda equina syndrome. This variable presentation is largely related to the accumulation of adipose tissue in different locations of the spinal canal.[16-19]

Magnetic resonance imaging (MRI) is the preferred method of diagnosis; the images typically show hyperintense tissue in the posterior epidural space in the T1 sequence, intermediate signal in the T2 sequence and hyposignal with fat suppression, which is characteristic of fatty tissue.[14,20-23] The typical image in the axial section was described by Kuhn et al.[2] as the 'Y' sign by circumferential compression of the dural sac at the lumbar level. Borré et al.[8] and Naka et al.[21] propose different gradings for SEL, where grade III is the most severe and symptomatic, and the characteristic 'Y' image is usually observed (Figures 1 and 2).

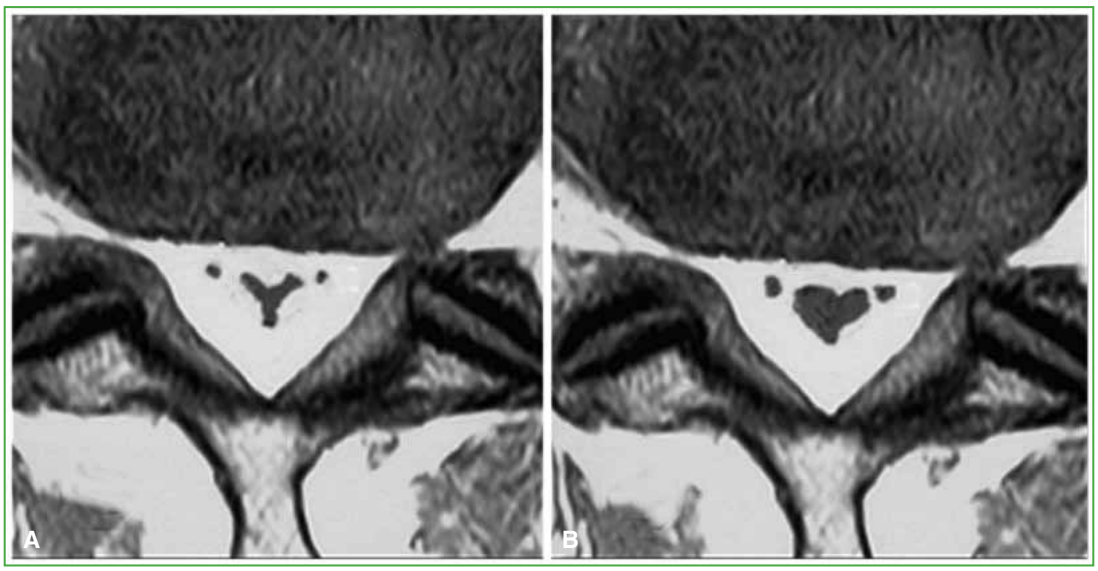

Figure 1. MRI of the lumbar spine, axial T1 sequence, axial slices. Fat accumulation in the L5-S1 intervertebral disc. **A.** 'Y' shape. **B.** Polygonal shape.

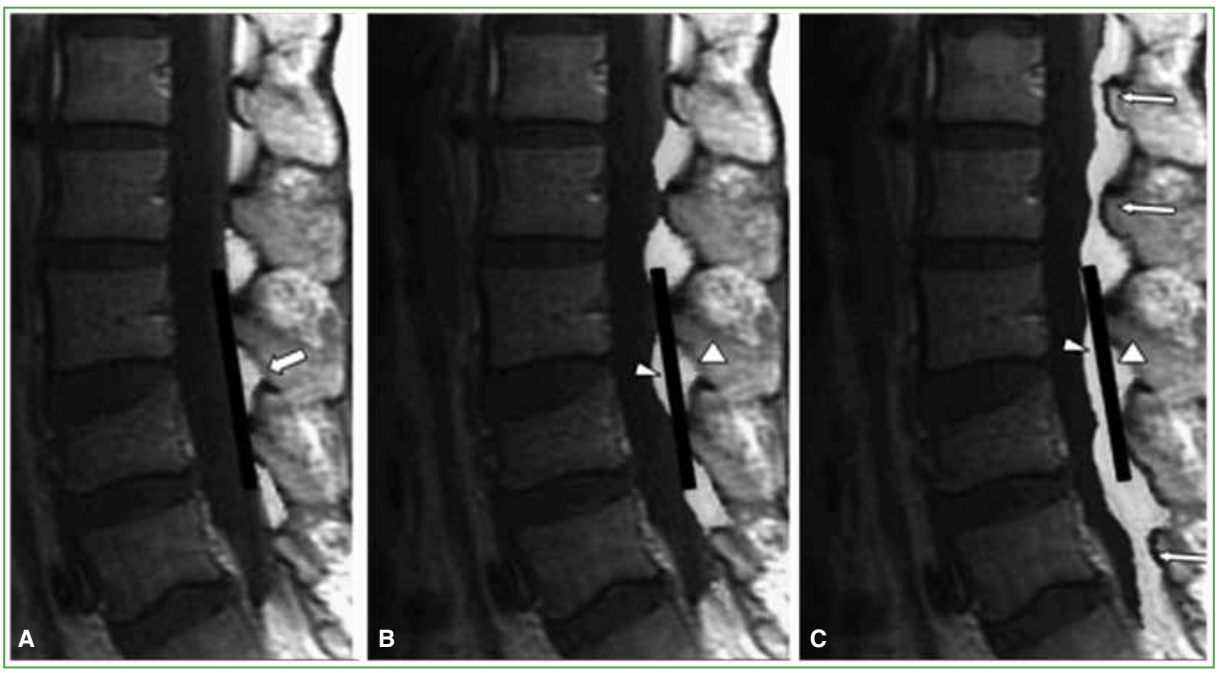

Figure 2. Classification of epidural lipomatosis according to Naka et al. MRI of the lumbar spine, T1 sequence, sagittal slices. **A.** Grade I, the fat is located between the indicated line and the extremities of the neighboring posterior arches (arrow). **B.** Grade II, the fat crosses the line, but is located between the extremes of the posterior arches (arrowheads). **C.** Grade III, fat crosses the line (arrowhead) and is seen over the extremes of the arches (arrows).

Since not all patients with grades I and II are symptomatic, SEL may be an incidental finding. For this reason, the aim of this study was to perform a descriptive analysis of the initial clinical manifestations of patients with Naka grade III lumbar spinal epidural lipomatosis (LEL).

MATERIALS AND METHODS

An observational, descriptive, retrospective study was conducted in the Spinal Pathology Unit of four institutions between January 2010 and January 2023. We included patients >18 years of age of both sexes who presented with low back pain with or without radiation, and had Naka grade III LEL on MRI.

Patients receiving corticosteroids, those suffering from endocrinopathies with cortisol alterations, those who discontinued follow-up or had incomplete records, and those with extradural disease, such as hematomas, angiolipomas or epidural metastasis, were excluded.

Data collection

Data collection was based on an exhaustive analysis of office medical records and the imaging archives of the institutions. Those who met the inclusion criteria were incorporated into a database developed by the same researchers from the medical records. The variables recorded included clinical parameters (age, sex, body mass index [BMI], comorbidities), imaging parameters (level affected on MRI), and symptomatic presentation parameters (reason for consultation, period of evolution).

The images were evaluated by two senior members of the team.

Ethical considerations

This study was approved by the corresponding Ethics and Research Committee of each institution. Given the retrospective nature of the study, participants' informed consent was not needed, and personally identifiable information was protected by coding it in a database to which only the investigators had access.

Statistical Analysis

Categorical variables are expressed as absolute number of presentation and percentage. Continuous variables that had a normal distribution are presented as mean and standard deviation; otherwise, as median and interquartile range (IQR). The Shapiro-Wilk test was used to verify the distribution of the sample.

Comparison of pre- and postintervention pain was performed with Student's t test for related samples or Wilcoxon signed-rank test for related samples, as appropriate. Comparisons between independent variable groups were performed with Student's t test for independent samples or the Mann-Whitney U test, as appropriate.

Data were analyzed using the IBM SPSS Macintosh program, version 24.0 (IBM Corp., Armonk, NY, USA).

RESULTS

Forty patients with Naka grade 3 LEL who presented for consultation to the spinal pathology team due to pain were included. Ten (25%) were women. The average age was 62.5 ± 12.9 years, none were smokers. The rest of the characteristics are detailed in Table 1.

Table 1. Sample characteristics

Variables	Total (n = 40)
Female sex, n (%)	10 (25)
Age, mean (SD)	62.5 (12.9)
BMI, median (IQR)	31.7 (29.6, 33.3)
Time of evolution, median (IQR), months	5.50 (3.75, 7.00)
Comorbidities, n (%) None Arterial hypertension Dyslipidemia Diabetes Asthma/COPD	 6 (15) 13 (32.5) 7 (17.5) 6 (15) 5 (12.5)
Other >1 comorbidity	22 (56.4) 6 (14.6)

SD = standard deviation; IQR = interquartile range; BMI = body mass index; COPD = chronic obstructive pulmonary disease

The median pain evolution was 5.50 months (IQR 3.75-7). In terms of clinical presentations, pain evolution lasted longer in radiculopathy-associated forms than in low back pain, with a statistically significant difference in low back pain (p = 0.017) (Figure 3).

The most affected level was L5-S1 (n = 22, 55%), followed by L4-S1 (35%). The L3-S1 group was older (median 60.5 vs. 59 and 59.5 years; p = 0.9), had suffered pain for longer (8.25 vs. 5.57 and 5.75 months; p = 0.32), and had a higher BMI (35.5 vs. 31.1 and 31.9 p = 0.069), but these findings were not significant. The most frequent reason for consultation was low back pain (n = 23, 56%); when the clinical presentation was analyzed according to the level affected, the most frequent clinical presentation in patients with L3-S1 involvement was lumbar and anterior thigh pain; this difference was not significant (Table 2).

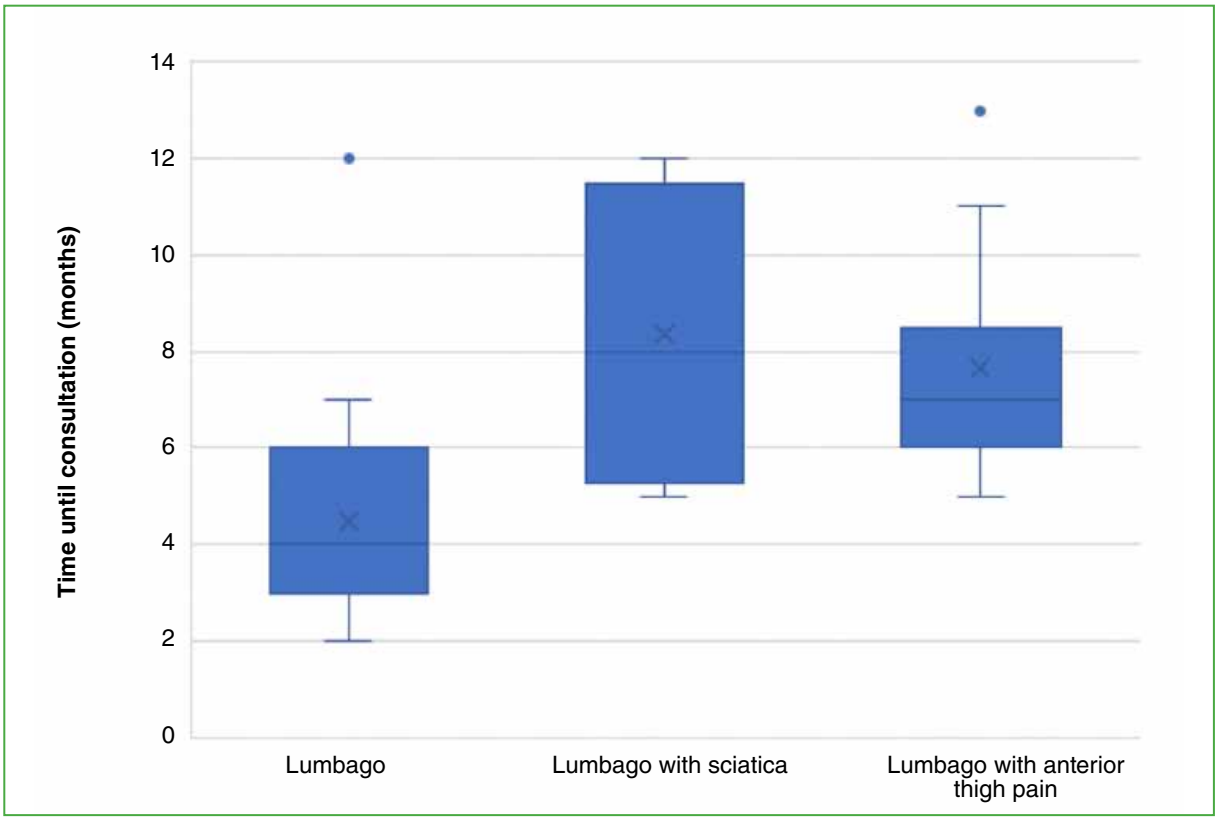

Figure 3. Distribution of time until consultation based on clinical diagnosis.

Table 2. Patient characteristics according to the compromised level.

	L3-S1 (n = 4)	L4-S1 (n = 14)	L5-S1 (n = 22)	p
Age, median [C1, C3]	60.5 [55.0, 64.8]	59.5 [51.5, 78.5]	59.0 [54.0, 71.0]	0.930
Female sex	1 (25.0)	3 (21.4)	6 (27.3)	1.000
Clinical manifestation				
Lumbago	1 (25.0)	9 (64.3)	13 (59.1)	0.366
Lumbago and anterior thigh pain	3 (75.0)	3 (21.4)	5 (22.7)	
Lumbago with sciatica	0 (0)	2 (14.3)	4 (18.2)	
Evolution, months				
Mean (SD)	8.25 (3.59)	5.57 (3.16)	5.73 (2.85)	0.322
Median [C1, C3]	7.50 [5.75, 10.0]	5.50 [3.00, 7.00]	5.00 [4.00, 6.75]	
I BMI				
Mean (SD)	35.1 (3.43)	31.9 (3.42)	31.1 (2.51)	0.069
Median [C1, C3]	34.0 [33,5; 35.6]	31.4 [29.5; 33.1]	30.8 [29.3; 33.2]	
Diabetes				
Yes	0 (0)	1 (7.14)	5 (22.7)	

SD = standard deviation; BMI = body mass index.

To facilitate the multivariate analysis, the variables time of evolution (<6 months vs. ≥6 months) and BMI (<30 vs. ≥30) were dichotomized.

When comparing the groups, bivariate analysis revealed that those with pain for a longer period of time were younger (65.8 vs. 59.1 years; p = 0.1) and had a lower BMI (31.3 vs. 32.2; p = 0.39); however, these values were not significant (Figure 4). Patients with a BMI >30 predominated (75%), who consulted mainly for low back pain (69 vs. 27%, p = 0.018), with a predominance of the male sex (86.2 vs. 45.5%, p = 0.014); the differences between age, time of evolution and affected level were not significant (Table 3).

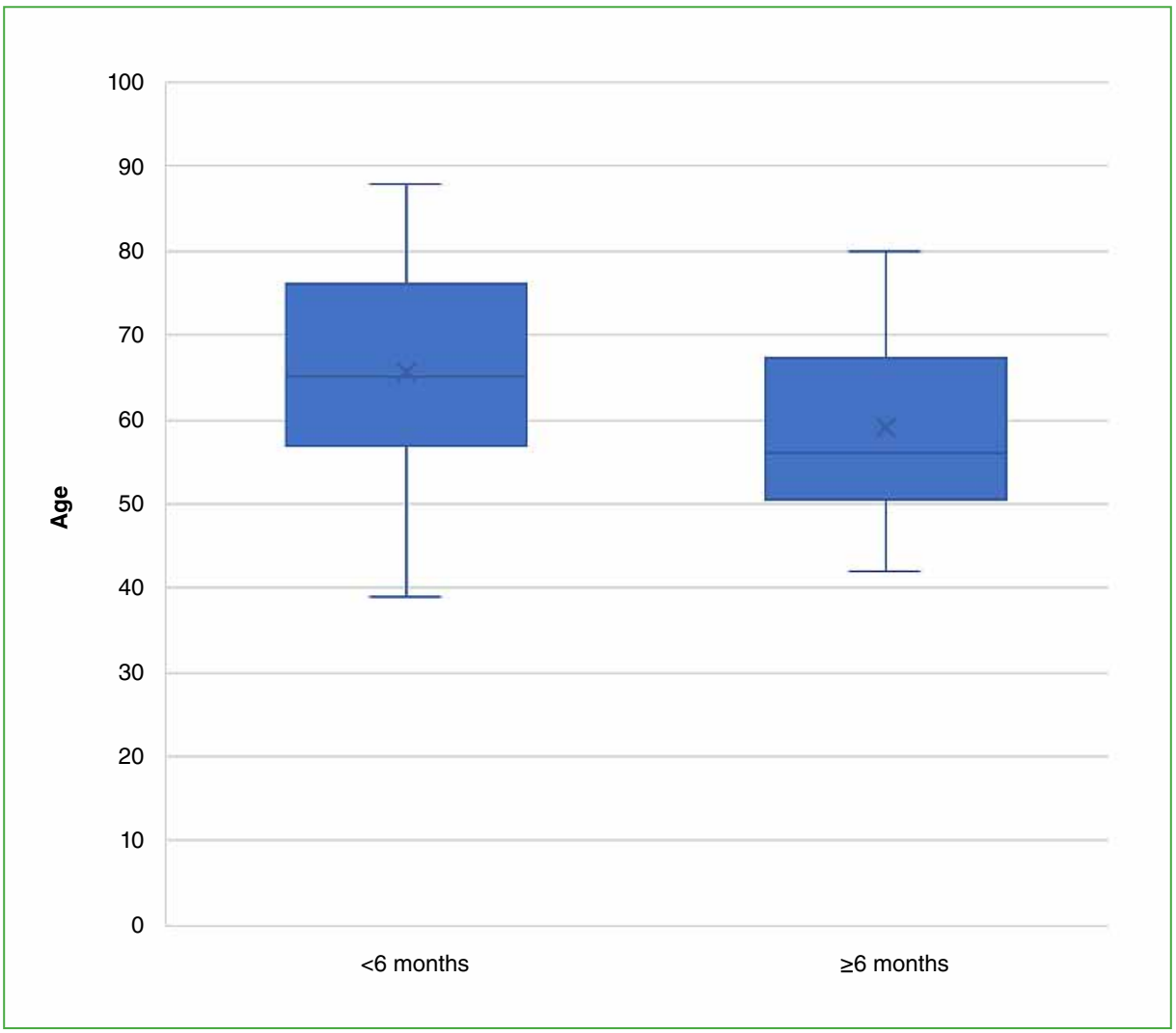

Figure 4. Distribution of age in the bivariate analysis according to time of evolution.

Table 3. Patient characteristics according to body mass index (BMI)

	IMC ≤30 (n = 11)	IMC >30 (n = 29)	p
Age, mean (SD)	59.0 (13.8)	63.8 (12.5)	0.331
Male sex	5 (45.5)	25 (86.2)	0.0144
Clinical manifestation			
Lumbago	3 (27.3)	20 (69.0)	0.018
Lumbago with sciatica (l/r)	4 (36.4)	2 (6.90)	
Lumbago and anterior thigh pain	4 (36.4)	7 (24.1)	
Time of evolution, median [C1, C3], months.	6.00 [5.00, 6.50]	5.00 [3.00, 7.00]	0.179
Level affected			
L3-S1	0 (0)	4 (13.8)	0.459
L4-S1	5 (45.5)	9 (31.0)	
L5-S1	6 (54.5)	16 (55.2)	

SD = standard deviation.

DISCUSSION

In this study, 40 patients with Naka grade III LEL and a mean age of 62.5 years, with male predominance, were evaluated. Seventy-five percent of the sample had a BMI >30. The most frequent reason for consultation was low back pain, with a median pain evolution of 5.5 months.

Idiopathic LEL accounts for approximately 17% of cases. The first case of idiopathic SEL was published by Badami and Hinck[19] in 1982. Years later, the idiopathic form was established: that which affects non-obese patients, without treatment with corticosteroids or any other underlying cause that justifies the entity; it is the least frequently reported form.[10-16] In this study, almost 25% of the patients had the idiopathic form and the rest were associated with obesity. It should be noted that this higher percentage than those published could be attributed to the exclusion of cases related to excess corticosteroids.

LEL usually predominates in the male sex (68-75%), is more frequent at 65 ± 10 years of age, and the duration of symptoms varies greatly according to the literature (4-27 months).[2-8,14,15,15,23-25] Typically, excessive adipose tissue accumulation occurs slowly and progressively, from a diminished thecal sac in the early stages to a completely obliterated one in the advanced stages. This excessive tissue generates a mass effect, causing mechanical compression and also venous compression leading to engorgement and compression of the dural sac, spinal cord, and roots.[18-22]

In our study, there were no patients with the acute form, the median evolution of pain was 5.5 months, with male predominance and a mean age similar to previous publications.

The location on the spine may vary according to the etiology of the disease, the thoracic form being the most frequent. Fogel et al.[15] reported a predominantly thoracic location (55.8%) when associated with exogenous corticosteroids, thoracolumbar involvement in cases of endogenous hypercortisolism, and a predilection for the lumbosacral area in idiopathic SEL and in those associated with obesity.[11-15] Naka et al. described the predilection of the middle thoracic and lower lumbar area in idiopathic cases, the thoracic forms being the ones most associated with neurological alterations (up to 70% of cases of myelopathy).[21]

In our study, as in previous reports, the low lumbar level (L5-S1) predominated in patients with idiopathic LEL and LEL associated with obesity.

There are several gradings according to MRI, but none correlates with clinical manifestations.[2,8,20-22] Borré et al.[8] proposed a classification of SEL on MRI from 0 to 3 according to the percentage of the spinal canal occupied by fatty tissue; grade 3 is always symptomatic (with canal occupation >70%). Naka used axial and sagittal MRI slices for classification from 0 to III, advanced forms are associated with the 'Y' image described by Kuhn et al.[2]

In our study, the clinical presentation of patients with Naka grade III LEL was analyzed, with a predominance of low back pain (56%).

The weaknesses of the study are its retrospective design and relatively small number of patients. On the other hand, non-probability sampling may be subject to possible selection bias. In spite of this, we highlight as a strength that it is the first to analyze the clinical presentation of Naka grade III LEL in our field and that the results obtained were similar to those of international publications on the subject, indicating an original contribution from our setting.

CONCLUSIONS

Our sample included 40 patients with Naka grade III LEL. Male sex predominated, the average age was 62.5 years, and there was greater involvement of the lumbosacral segment.

The most common presentation was low back pain, with the exception of L3-S1 involvement, which caused lumbar and anterior thigh pain. Seventy-five percent of the sample was obese, with no significant differences in terms of age, pain evolution and level affected with respect to patients with a BMI ≤30. Patients who reported pain for >6 months were younger and had a lower BMI; however, these findings were not statistically significant.

Conflict of interest: The authors have no conflicts of interest to declare.

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