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(CASE REPORT)

The longest documented recurrence of a Nondysraphic intramedullary spinal cord lipoma: A case report and review of literature

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Abstract

Introduction: Intramedullary lipomas are rare benign spinal cord tumors and even rarer are nondysraphic intramedullary spinal cord lipomas, accounting for less than 1% of all intraspinal tumors. Recurrence after surgical resection is documented, but long-term natural history data remain limited.

Case Presentation: A 54-year-old male presented with progressive bilateral lower limb weakness and posterior column syndrome. He had undergone subtotal resection of a thoracic intramedullary lipoma 30 years previously at age 24 and remained asymptomatic for 28 years. MRI demonstrated a lipomatous lesion extending from D1 to D2. He underwent complete macroscopic resection. Histopathology confirmed benign lipoma. The patient showed excellent functional recovery at six-month follow-up.

Conclusion: This represents the longest documented recurrence interval for nondysraphic intramedullary lipomas. Complete resection can be safely achieved in recurrent cases using modern microsurgical techniques. Lifelong surveillance is mandatory regardless of asymptomatic duration.

Keywords: Intramedullary lipoma; Nondysraphic; Spinal cord tumor; Recurrence; Long-term follow-up

1. Introduction

Nondysraphic intramedullary spinal cord lipomas are exceptionally rare benign lesions, comprising less than 1% of all intraspinal tumors. Unlike dysraphic forms associated with spina bifida, these lesions occur without spinal dysraphism and typically present in the second to fifth decade of life with slowly progressive myelopathy. Mostly reported in the cervical or cervico-thoracic junction.

The pathogenesis remains controversial. The developmental error theory, most widely accepted, suggests misplaced mesenchymal cells during neural tube development differentiate into adipocytes. Recently, some authors proposed that nondysraphic forms may represent true neoplasms rather than static malformations, explaining their tendency to recur after resection.

Surgical management traditionally favors conservative subtotal resection to preserve neurological function, as recurrence rates appear similar regardless of resection extent. Limited long-term data exist on recurrence patterns. The longest documented interval between surgery and recurrence is 17 years in nondysraphic forms and 15 years in dysraphic forms.

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We present a unique case of thoracic intramedullary lipoma recurring 30 years after initial resection. This case provides exceptional natural history data.

2. Case presentation

We report the case of a 54-year-old male who presented with a 2 years history of progressive bilateral lower limb weakness and sensory disturbances. The patient had undergone subtotal resection of an upper thoracic intramedullary lipoma 30 years ago in 1995 (Figure 1) at the age of 24, with complete recuperation of his symptoms, following which he remained asymptomatic for 28 years with no regular imaging follow-up.

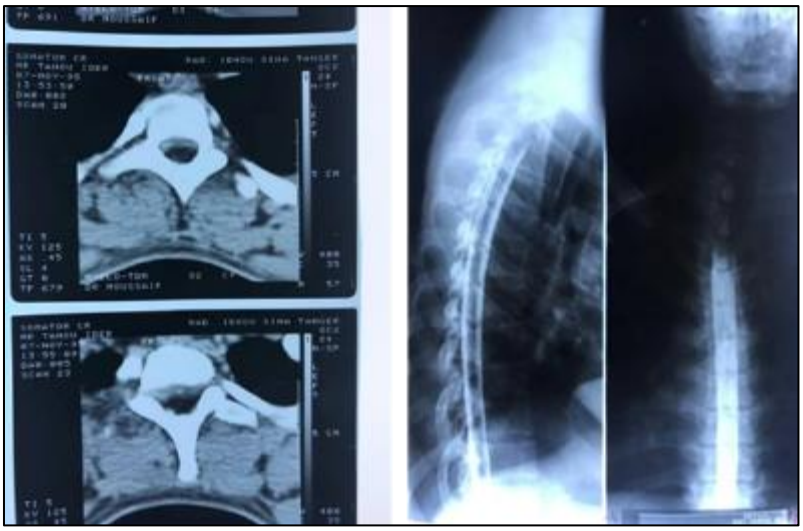

Figure 1 1995 pre-operative Myelo-CT showing anterior upper thoracic spinal cord displacement and compression

His current symptomatology began insidiously with balance difficulties, particularly in low-light conditions, progressing to bilateral lower limb weakness, progressing over the course of 2 years. Though bowel and bladder continence were maintained.

Neurological examination of the upper limbs found normal MRC muscle power scale, tone and reflexes. Lower limb examination revealed weakness with power of 3/5 bilaterally. Associated with all pyramidal signs : tone was spastic with mild atrophy of the right quadriceps, deep tendon reflexes of the lower limbs were hyperactive with sustained ankle clonus and bilateral Babinski sign, confirming upper motor neuron involvement. Sensory examination demonstrated a classic posterior column syndrome.

Magnetic resonance imaging revealed a well-defined intramedullary lesion extending from D1 to D2 vertebral levels **(Figures 2&3)**. The lesion demonstrated marked hyperintensity on both T1- and T2-weighted sequences, complete signal suppression on STIR images and no enhancement after GADO injection corresponding to classical features of lipoma. The spinal cord was pushed ventrally and no sign of spinal dysraphism was observed. Based on his past history, the clinical presentation and classic radiological features, the diagnosis of recurrent thoracic nondysraphic intramedullary lipoma with cord compression was established.

Figure 2 Sagittal MRI cuts showing the dorsal tumors extending from D1 to D2 with typical characteristics of a lipoma hyper signal T1 and T2, hypo signal in STIR WI and no contrast enhancement after T1 injection

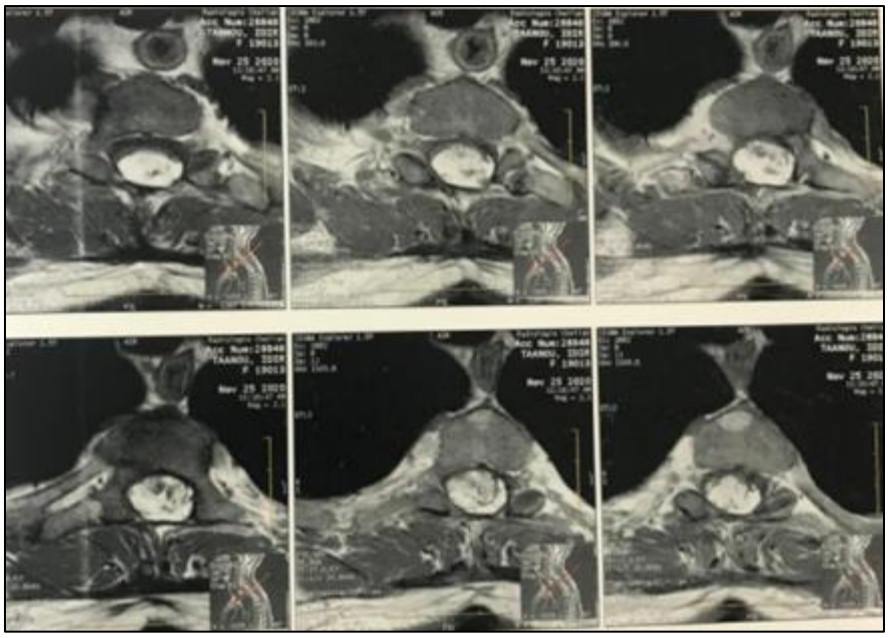

Figure 3 Coronal T2WI cuts showing the intramedullary lipoma in hyper signal pushing the spinal cord anteriorly

The patient underwent surgical intervention with posterior approach for maximal safe resection. The previous laminectomy site at C7-T2 was identified with dense fibrous tissue adhesions. The dura was opened, we identifying a white-yellowish fibrous mass with lobulated fatty tissue in close contact with the medulla (Figure 4). Using microsurgical techniques and meticulous bipolar coagulation for hemostasis we could achieve complete macroscopic monobloc resection (Figure 5).

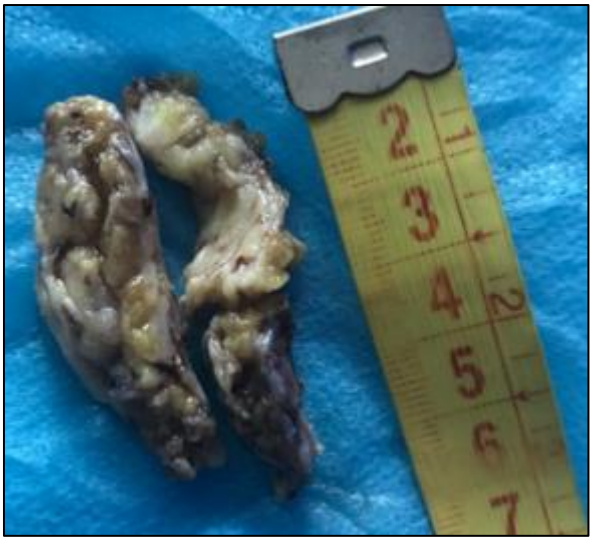

Figure 4 Tumor aspect after resection

Figure 5 Per-op imaging showing the laminated aspect of the spinal cord after resection

Following surgery patient presented a worsening of his motor weakness going from 3/5 to 1/5 MRC power scale, patient was put under corticosteroids and after 3 months of intensive reeducation he has regained an acceptable muscle power (4+/5 in both lower limbs). Histopathological examination of the tumor confirmed the diagnosis of intramedullary lipoma.

3. Discussion

Intramedullary lipomas can be differentiated into dysraphic (the most common) and nondysraphic depending on the presence or not of spinal dysraphism features (cutaneous stigmata, vertebral anomalies, teetered cord syndrome…).

Our case is particularly noteworthy for several reasons: the remarkably prolonged latency period of 30 years between initial surgery and symptomatic recurrence, the achievement of complete macroscopic resection in a recurrent lesion with dense adhesions and the excellent functional outcome following surgery. To our knowledge, this represents one of the longest documented intervals between primary surgery and recurrence in the literature for nondysraphic intramedullary lipomas.

3.1. Epidemiology and Natural History

Nondysraphic intramedullary lipomas account for <1% of all spinal cord tumors characteristically, only a few case reports have been reported no big series. The extreme rarity of nondysraphic intramedullary lipomas is underscored by multiple large series: Endo et al.(**1**) documented zero lipomas among 1,033 intramedullary tumors, while a Chinese single-center series of 1,321 intradural tumors (**2**) similarly reported lipomas as rare or absent.

From many case reports we could deduce a bimodal age distribution with symptoms occurring either :

During the first years of life, congenital lesions with early mass effect in this case always search for associated dysraphism,

During the second to fifth decades more common presentation for nondysraphic forms, the same congenital lesions presenting with delayed symptoms. Our patient's initial presentation at age 24 and recurrence at age 54 aligns with this pattern, though the 30-year asymptomatic interval is exceptional.

Table 1 Epidemiology and anatomical location from different case series

Study	N	Age mean (range)	Anatomical location
Bhatoe et al (2005) (3)	14	30-35 y (20s- 40s)	Cervical & thoracic n=13 Lumbar n = 1
Fleming et al. (2010) (4)	5	7 y (2-12)	Cervical and thoracic
Lee et al. (1995) (5)	6	Not stated (8 - 45y)	Cervical & thoracic n = 5 Lumbar n = 1
Srinivasan et al. (2014) (6)	3	25.3 y (25-26)	Thoracic and lumbar
Andrew M. Hersh (2024) (7)	6	35 y (28-57)	Thoracic n= 4, cervicothoracic n= 1, lumbar n=1

3.2. Pathogenesis

The pathogenesis of nondysraphic intramedullary lipomas is still controversial, three theories have been proposed by Ammerman et al (**8**). and reinforced by some authors later

The developmental theory (**8,9**) (the most widely accepted), postulates that mesenchymal cells - precursors of adipocytes - migrate abnormally into the developing neural tube in the neurulation phase developing into a lipoma later.

The metaplasia theory (**8**) suggests transformation of existing neural or glial tissue into adipose tissue,

The hamartomatous theory (**8,10**) proposes that these represent congenital malformations rather than true neoplasms, this has been supported by histopathological findings of lipomas often containing a mixture of mature adipose tissue / fibrous connective tissue / blood vessels / peripheral nerves….

This theory have been challenged by Fleming's article (**4**) who made an important distinction differentiating dysraphic lipomas considered true hamartomas and nondysraphic lipomas thought to be true neoplasms which explains their recurrence (like in our case).

We can find across some articles the influence of body fat in some patients, with reports of size reduction of the lipoma with the decrease of body mass index **(11, 12, 13,14),** while this may be true in some form of epidural lipomatosis, most of the case reports for intramedullary are lacking data on body mass index to establish the correlation (in our case the patient was not overweight).

3.3. Clinical and radiological features

Our patient presented with progressive spastic paraparesis and posterior column dysfunction is characteristic of cervicothoracic intramedullary NDIL. Indeed, across reported series, nondysraphic intramedullary lipomas exhibit a characteristic central expansion within the spinal cord, often posteriorly or posterolaterally :

- Corresponding to the pattern of the embryologic inclusion of mesenchymal elements along the dorsal midline during neural tube closure.
- Leading to progressive pyramidal and posterior column dysfunction.
- This expansion results in ventral flattening of the cord on MRI.
- MRI remains the diagnostic modality of choice, with characteristic findings that are virtually pathognomonic: hyperintensity on both T1- and T2-weighted sequences with complete signal suppression on fat-saturated images (STIR). With minimal to no enhancement after GADO injection.

Always search for spinal dysraphism features that differentiate the two variants mainly :

- Clinical : dermal sinus, dimple, hairy patch, hemangioma
- Radiological : defect of posterior vertebral elements, normal vertebral segmentation, subcutaneous lipoma continuous with the spinal canal, tethered cord, diastematomyelia, syrinx, scoliosis…

3.4. Surgical Management

The surgical management of nondysraphic intramedullary spinal cord lipomas remains challenging due to the intimate relationship between lipomatous tissue and functioning neural elements. Contemporary surgical philosophy favors subtotal resection over aggressive gross total removal, as subtotal tumor excision (approximately 70-80% resection) has demonstrated good outcomes while minimizing the risk of iatrogenic neurological deterioration.

The approach principles follow all the rules of intramedullary tumors surgery, including posterior midline exposure with laminectomy / laminoplasty extending one level above and below the tumor, followed by dural opening under microscopic visualization, then a midline myelotomy is performed at the point of maximal cord expansion or yellowish discoloration, with pial sutures applied for gentle retraction. The lipoma-cord interface cannot typically be established intraoperatively due to dense adhesions between the fatty tissue and neural parenchyma, limiting the extent of safe resection. Intraoperative neurophysiological monitoring (MEPs / SSEPs / D waves) or intraoperative ultrasound provide valuable information making tumor debulking safer.

3.5. Postoperative Outcomes and Prognosis

Table 2 Summarizes outcome and prognosis based on some articles

Series	**Extent of resection**	**Immediate postop outcome**	**Follow-up**	**Recurrence**
Bhatoe et al(3) (2005) (n=14)	Partial to subtotal	Transient decline	No motor deterioration Motor and/or sensory improvement in majority	Not detailed
Fleming et al.(4) (2010) (children n=5)	Variable	Transient decline	Variable recovery	2/5 cases
Lee et al.(5) (1995) (n=6)	Variable	No improvement	Poor outcomes	2/6 cases
Srinivasan et al.(6) (2014) (n=3)	Radical subtotal	2 cases improved 1 case remained stationary	The 2 cases : complete improvement The 1 case : no improvement	The 2 cases = no recurrence 1 case = recurrence after 6 years

Andrew M. Hersh (2024) (7) (n=6)	Subtotal resection	All cases experienced mild neurological decline	5 cases improved 1 case of aggravation (Special case : already operated 17 years ago and experienced recurrence after 4 years)	1 case = recurrence after 4 years

What we can conclude is :

- Majority of patients experience transient neurological decline in the immediate postoperative.
- Preoperative neurological status is the most important prognostic factor
- Subtotal resection (70-80%) provides good outcomes without excessive risk
- Recurrence is common (mean 11±7 years) but similar across all resection extents

3.6. Implications for Follow-up and Surveillance

Our case has important implications for surveillance protocols following resection of intramedullary lipomas. The 30-year latency period before symptomatic recurrence challenges the practice of discontinuing follow-up after 5-10 years, as is common for many benign spinal tumors. We propose that patients who have undergone resection of intramedullary lipomas, regardless of extent of resection, should be counseled regarding lifelong surveillance with periodic MRI examinations. While annual imaging may be reasonable in the first decade following surgery, the frequency might be reduced to every 2-3 years subsequently if the patient remains asymptomatic and stable on imaging. However, any new or progressive neurological symptoms should prompt immediate imaging evaluation.

The finding that our patient remained asymptomatic for 28 years before developing symptoms over a 6-month period suggests that tumor regrowth may follow a similar pattern of slow accumulation followed by rapid clinical deterioration once a critical size is reached. The alternative approach of clinical surveillance alone, relying on symptom development to trigger imaging, risks delaying diagnosis until significant neurological compromise has occurred, potentially limiting recovery potential. Patients should be informed.

This also raises the question of management of asymptomatic patients, as no consensus on surgical intervention is available, should we go for a prophylactic surgery? Or wait until the onset of symptoms? The latter is our policy in our department for all benign lesions.

4. Conclusion

We present a rare case of recurrent nondysraphic intramedullary lipoma, the longest documented latency period after initial subtotal resection. The patient's classic presentation, MRI findings, and successful complete resection highlight important teaching points. Nondysraphic intramedullary lipomas, though benign, have inherent growth potential and require lifelong surveillance. Complete macroscopic resection can be safely achieved in experienced hands using modern microsurgical techniques and intraoperative neuromonitoring, even in challenging recurrent disease. Early surgical intervention for symptomatic lesions before irreversible neural damage is crucial for functional recovery. Distinguishing nondysraphic intramedullary lipomas from complex dysraphic lipomas is critical, as they represent distinct entities with different pathogenesis, natural history, and surgical strategies.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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