Neurol Med Chir (Tokyo) 45, 375～378, 2005

# Mature Spinal Teratoma Associated With Thickened Filum Terminale

## —Case Report—

Özkan ATES, Süleyman R. ÇAYLI, Ayhan KOÇAK, Alpay ALKAN*, Çagatay ÖNAL, and Ufuk USTA**

*Departments of Neurosurgery, *Radiology, and **Pathology, Inönü University School of Medicine, Malatya, Turkey*

### Abstract

**A 30-year-old man presented with an intradural spinal teratoma with thickened filum terminale manifesting as urinary and sexual disturbances, and low back pain persisting for 4 years. Spinal magnetic resonance imaging revealed thickened filum terminale containing a heterogeneously enhanced intradural lesion extending from the L-3 to L-4 levels and in contact with the conus medullaris. The filum terminale was incised and the tumor was totally resected. The histological diagnosis was mature teratoma consisting of three germ cell layers. The patient's complaints had completely resolved 6 months later.**

Key words: intradural teratoma, thickened filum terminale, tethered cord, spinal cord tumor, filum terminale teratoma

## Introduction

Intraspinal teratoma was first described in 1863[17] and then in 1888.[4] Teratoma is generally believed to originate from three germ cell layers.[2,8,9] The age at occurrence varies widely from newborn to 57 years.[7,13] Intramedullary teratomas may occur along the whole spinal cord with predominance in the thoracolumbar region.[13] The clinical symptoms and findings are related to the location of the tumor as for other intramedullary tumors.

The conventional view of the pathogenesis of teratoma is that primordial germ cells failed to migrate properly during early embryonic development.[16] A clinicopathological study based on seven cases of spinal cord teratoma suggested that teratomas could originate from misplacement of a multipotential germ cell.[1] Pluripotential embryonic caudal mesenchymal cells could give rise to teratomas due to dysfunction of several factors that probably involve gene functions and cellular inductive interactions, suggesting that development of spinal teratoma is not neoplastic but the result of multiple dysembryogenic mechanisms.[9] Spinal teratomas are well known to associate with dysraphic congenital malformations.[2,3,5–7,9,10,12] The co-occurrence of spinal dysraphism and intramedullary teratoma indicates a developmental fault in the same period as suggested by dysembryogenic theory.

Here we describe a unique case of filum terminale teratoma associated with thickened filum terminale.

Received June 29, 2004; Accepted January 6, 2005

## Case Report

A 30-year-old man was admitted to our department on December 3, 2001. The patient's complaints were low back pain, sensation of incomplete voiding, constipation, and problems with erection and ejaculation. These complaints had started 4 years ago and the severity had increased.

Physical examination revealed bladder distension up to the pubic symphysis, normal external genitalia, and normal prostate gland function. Flow cystometry study revealed residual urine volume of 400 ml. Motor, reflex, and sensory examinations, and routine urine, hematological, and serum tests found no abnormalities. Somatosensory evoked potential (SEP) study revealed prolonged latency and decreased amplitude.

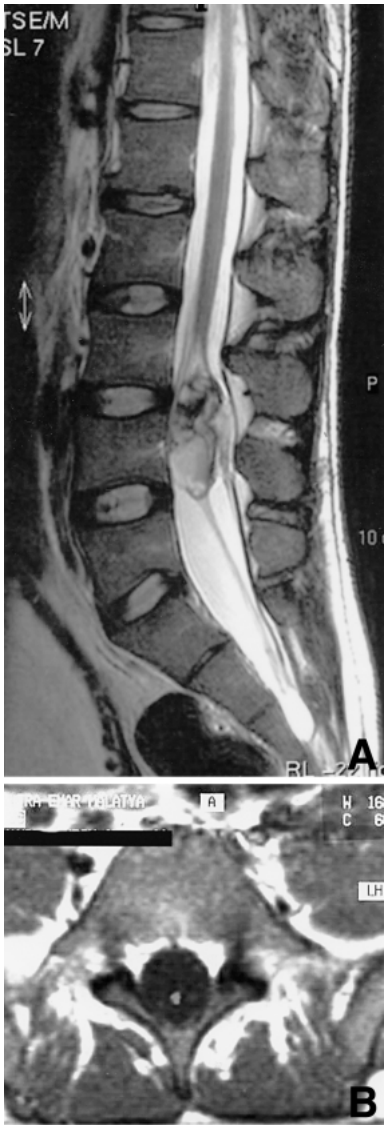


**Fig. 1** **A: Preoperative sagittal $T_2$-weighted magnetic resonance (MR) image showing a solid mass with fatty and cystic components causing expansion of the spinal canal. B: Preoperative axial $T_1$-weighted MR image showing thickened filum terminale passing through cross-sectional images at the L5–S1 levels.**

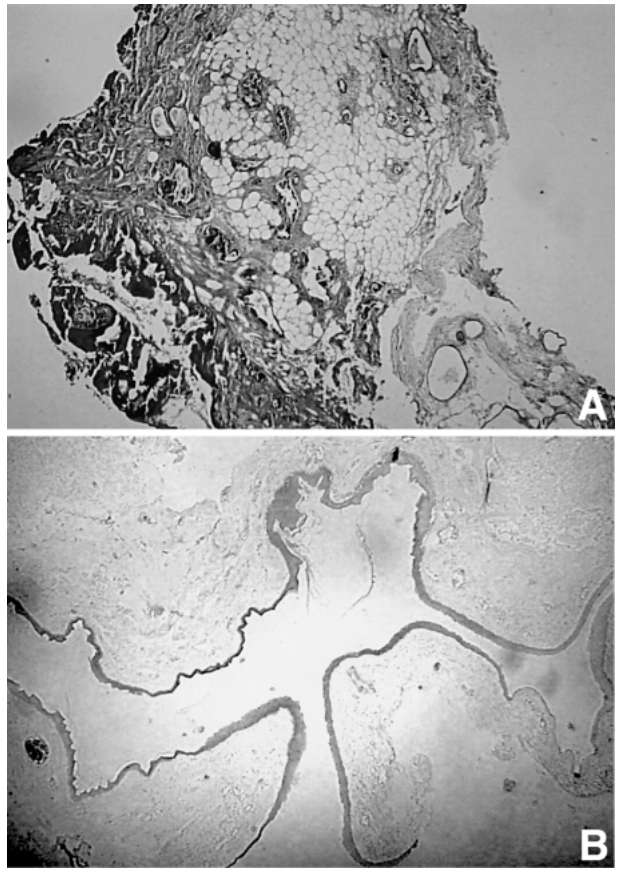


**Fig. 2** **A: Photomicrograph showing a large number of glandular formations covered by prism and flattened cubic epithelial cells and embedded in fibrovascular tissue and mature fat tissue which contain some cystic formations. HE stain, ×100. B: Photomicrograph of a cystic formation covered by multiple levels of keratinous squamous epithelial cells containing keratin lamellae. HE stain, ×100.**

Computed tomography demonstrated an intradural space-occupying lesion and widening of the spinal canal at the L3–4 levels. Sagittal $T_2$-weighted magnetic resonance (MR) imaging showed thickened filum terminale containing a heterogeneously enhanced mass lesion extending from the L-3 to L-4 levels and in contact with the conus medullaris (Fig. 1A). Axial $T_1$-weighted MR imaging showed a thickened fatty filum terminale passing through cross-sectional images at the L5–S1 levels (Fig. 1B).

Total laminectomy was performed at the L-3 and L-4 levels. The dura mater was opened with a midline incision to expose the enlarged medulla. The yellowish tumor found located adjacent to the conus medullaris in the filum terminale. Fatty substance was present outside the tumor, and mucous substance, bony fragments, and hair were found inside the tumor. Dissection and total removal of the tumor were easy, but fibro-adipose tissue was adherent adjacent to the conus medullaris in some places. The adherent tissue was dissected carefully and totally removed. The thickened filum terminale was then released.

Histological examination found a large number of

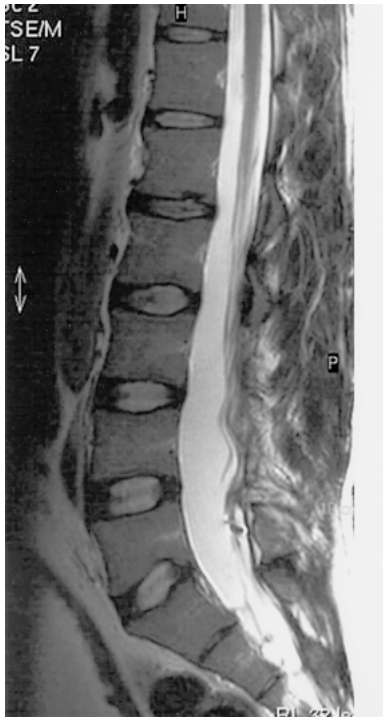


**Fig. 3 Postoperative sagittal $T_2$-weighted magnetic resonance image showing total resection of the mass and the conus medullaris at the L2-3 disc space.**

glandular formations covered by mature prism and flattened cubic epithelial cells (Fig. 2). These structures were embedded between fibrovascular tissue and mature fat tissue, which included some cystic formations. Three germ cell layers including ectodermal, mesodermal, and endodermal elements were observed. The histological diagnosis was mature teratoma.

One week after the operation, the patient's low back pain improved but his urinary and sexual complaints continued. Six months after the operation, neurological findings and urinary functions were exactly normal. Problems with erection and ejaculation were also improved. SEP study revealed normal latency amplitude. Postoperative $T_2$-weighted MR imaging revealed total resection of the mass and the conus medullaris at the L2-3 disc space without evidence of residual or recurrent tumor (Fig. 3).

## Discussion

Thickened filum terminale is a different pathological entity from other dysraphic congenital malformations, such as spina bifida, split cord malformation, meningomyelocele, and lipomeningomyelocele, so originates at a different stage of embryogenesis.[14] Almost all neural tube defects other than thickened filum terminale occur as a result of faults in the primary neurulation period. The filum terminale is considered the caudal part of medulla. Thickened filum terminale is an abnormality that arises in the canalization period of the embryogenesis.[14] In our case, the teratoma originated from the filum terminale and grew adjacent to the conus medullaris. The localization of the tumor in the filum terminale indicates the occurrence of a secondary neurulation period developmental anomaly. We suggest that misplacement of pluripotential embryonic caudal mesenchymal cells during the secondary neurulation period before closure of neural tube could result in teratoma formation. It is not clear whether the thickened filum terminale in our case formed due to the teratoma or was an independent anomaly that coincidentally occurred during the secondary neurulation period.

This case involved the unique association of teratoma and thickened filum terminale without other dysraphic malformation originating in the primary neurulation period. This case supports the dysembryogenic theory for the pathogenesis of teratomas.[9] Total surgical resection is the best choice of treatment for spinal intradural teratoma. However, total resection should not be attempted in cases with tight adherence to the neural tissue. Recurrence has been observed in incomplete resections.[6,7,11,13,15] In our case, the tumor was totally resected and the thickened filum terminale was incised. Postoperative MR imaging observed no residual tumor.

## References

1) Al-Sarraj ST, Parmar D, Dean AF, Phookun G, Bridges LR: Clinicopathological study of seven cases of spinal cord teratoma: a possible germ cell origin. *Histopathology* 32: 51–56, 1998
2) Caruso R, Antonelli M, Cervoni L, Salvati M: Intramedullary teratoma: case report and review of the literature. *Tumori* 82: 616–620, 1996
3) Etuş V, Ceylan S: Spinal cord teratoma associated with dysraphism: An unclear pathogenesis. *Journal of Neurological Sciences* (*Turkish*) 20: 46–50, 2003
4) Gowers WR, Horsley VA: A case of tumor of the spinal cord: removal and recovery. *Trans med Chir Soc Edinb* 71: 379–430, 1888
5) Hader WJ, Steinbok P, Poskitt K, Hendson G: Intramedullary spinal teratoma and diastematomyelia: case report and review of the literature. *Pediatr Neurosurg* 30: 140–145, 1999
6) Hamabuchi M, Hasegawa R, Murase T: Teratoma of the spinal cord: a case report with CT scans. *J Bone Joint Surg Br* 71: 390–392, 1989

7) Hamada H, Kurimoto M, Hayashi N, Hirashima Y, Matsumura N, Endo S: Intramedullary spinal teratoma with spina bifida. *Childs Nerv Syst* 17: 109–111, 2001
8) Hejazi N, Witzmann A: Spinal intramedullary teratoma with exophytic components: report of two cases and review of the literature. *Neurosurg Rev* 26: 113–116, 2003
9) Koen JL, McLendon RE, George TM: Intradural spinal teratoma: Evidence for a dysembryogenic origin. Report of four cases. *J Neurosurg* 89: 844–851, 1998
10) Muthukumar N: Split cord malformation and cystic teratoma masquerading as lipomeningomyelocele. *Childs Nerv Syst* 19: 46–49, 2003
11) Nonomura Y, Miyamoto K, Wada E, Hosoe H, Nishimoto H, Ogura H, Shimizu K: Intramedullary teratoma of the spine: report of two adult cases. *Spinal Cord* 40: 40–43, 2002
12) Pelissou-Guyotat I, Sindou M, Pialat J, Goutelle A: Teratome mature intramedullaire associe a une moelle attachee et a un lıpome ıntradural: a propos d'un cas opere. Revue de la litterature. *Neurochirurgie* 34: 205–209, 1988 (Fre, with Eng abstract)
13) Poeze M, Herpers MJHM, Tjandra B, Freling G, Beuls EAM: Intramedullary spinal teratoma presenting with urinary retention: case report and review of the literature. *Neurosurgery* 45: 379–385, 1999
14) Selçuki M, Coşkun K: Management of tight filum terminale syndrome with special emphasis on normal level conus medullaris (NLMC). *Surg Neurol* 50: 318–322, 1998
15) Seol HJ, Wang KC, Kim SK, Kim H, Choe G, Chi JG, Cho BK: Intramedullary immature teratoma in a young infant involving a long segment of the spinal cord. *Childs Nerv Syst* 17: 758–761, 2001
16) Teilum G: Classification of endodermal sinus tumor (mesoblastoma vitellinum) and so-called 'embryonal carcinoma' of the ovary. *Acta Pathol Microbiol Scand* 64: 407–429, 1965
17) Virchow R: *Die Krankhaften Geschwülste, vol 1*. Berlin, A Hirschwald, 1863, p 514 (Ger)

*Address reprint requests to*: Ö. Ates, M.D., Inönü Üniversitesi, Turgut Özal Tip Merkezi, Nörosirürji Anabilim Dali, 44069 Malatya, Turkey.
*e-mail*: atesozkan@hotmail.com