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Case Report

Intradural calcifying pseudoneoplasm of the neuraxis presenting as a cauda equina syndrome

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Abstract

Background: Calcifying nonneoplastic pseudoneoplasms of the neuraxis (CAPNON) have been reported in 59 cases in literature, however, they rarely involve the spinal cord. Owing to the advances in immunohistochemical markers, their structure and origin are better understood now.

Case Report: We present the case of a 72-year-old female who had longstanding history of low back pain that exacerbated 20 days prior to the presentation to the emergency room with a frank cauda equina syndrome.The lumbar computed tomography scan showed a hyperdense lesion, suggestive of calcified tumor, whereas the magnetic resonance imaging revealed a hypointense lesion on theT1 and T2-weighted images, without contrast enhancement or edema on fluid-attenuated inversion recovery. She underwent an emergent L2-L4 laminectomy and L3-L4 discectomy with resection of L2 intradural tumor, following which she regained normal function.

Conclusion: A 72-year-old female presented with a cauda equina syndrome attributed to an L2 intradural CAPNON. Following gross total resection, the patient was neurologically intact.

Key Words: Calcifying nonneoplastic pseudoneoplasms (CAPNON), differential diagnosis, intradural, neoplasm, spine

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INTRODUCTION

Calcified pseudoneoplasms of the neuraxis (CAPNON) are nonneoplastic lesions that rarely involve the central nervous system (CNS) and particularly the spine. Clinical features depend on the site of the lesion and may vary from asymptomatic to severely symptomatic; surgical resection is an effective treatment modality.[1,2]

Although the clinical presentation is often characterized by pain, with no neurological impairment, neurological deficits may occur if there is significant compression of the neural structures. Here, we describe a 72-year-old

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female who presented with an acute cauda equina syndrome attributed to CAPNON; following gross total excision, the patient was neurologically intact.

CASE REPORT

A 72-year-old female, with a longstanding history of low back pain, noted exacerbation of pain 20 days prior to admission with a cauda equina syndrome (2/5 proximal and 0/5 distal strength). A lumbar spine computed tomography (CT) scan showed a hyperdense intravertebral lesion of uncertain etiology (e.g., calcified tumors/ectopic calcifications inside vertebral canal vs. calcified embryological remnants). A lumbar magnetic resonance (MR) revealed with a lesion that was hypointense on T1 and T2-weighted images and showed no enhancement with contrast and no edema on fluid-attenuated inversion recovery studies [Figures 1 and 2]. She underwent an L2-L4 laminectomy and L3-L4 discectomy, as well as the resection of an L2 intradural tumor [Figure 3]. During surgery, the lesion was not adherent to the lumbosacral roots but displaced the nerve roots while contributing to cauda equina compression. Gross total resection of this extremely calcified lesion was accomplished [Figure 4].

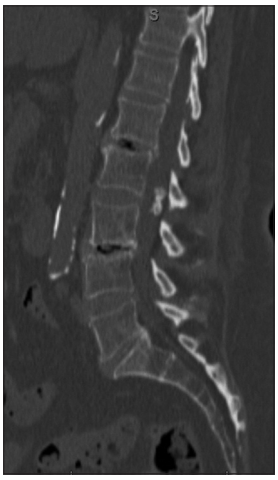

Figure 1: Lumbar computed tomography scan: hyperdense lesion, suggestive of a calcified tumor

Figure 3: Intraoperative image: intradural calcified lesion

Postoperatively, the patient was discharged on postoperative day 4 with residual proximal 4/5 and distal 2/5 residual motor deficits.

DISCUSSION

History and etiology of calcifying nonneoplastic pseudoneoplasms

Although common in the limbs, calcifying pseudoneoplasms (CAPNON) are rare lesions in the CNS (brain or spine).[1,2] They be found at any age but are more common in patients over 50 years old, and are more typically noted in males.[2,5] In some publications, they are associated with neurofibromatosis type 2.[3] To date, 59 cases have been described in the literature; 32 in the brain and 27

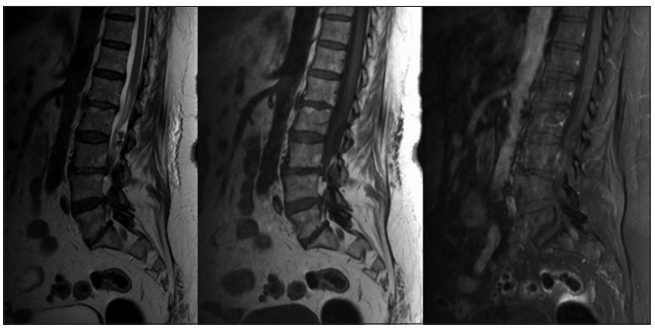

Figure 2: T1- and T2-weighted magnetic resonance imaging shows hypointensity in both series and no enhancement by contrast

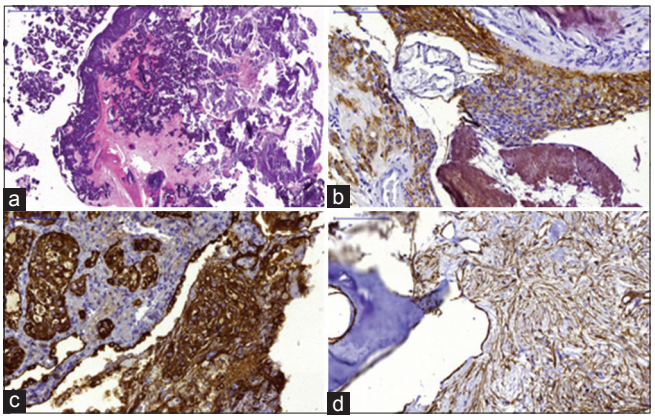

Figure 4: Histological section of calcifying nonneoplastic pseudoneoplasms stained for hematoxilin and eosin (a) and immunohistochemical expression for glial fibrillary acidic protein (GFAP), epithelial membrane antigen (EMA), and smooth muscle actin (b, c, and d, respectively). Optical microscopy, augmented ×200 (blue bar); (a) extensive laminating and concentric arranged calcified lesion with bone formation trabecules associated with fibrous tissue; (b) small and round cell proliferation with minimal atypia surrounding the calcified and fibrous lesion which expressed GFAP; (c) strong and diffuse expression for EMA in the same surrounding cells which denotes ependymal differentiation; (d) fibro-osseous lesion immunostained for smooth muscle actin that demonstrated fibroblastic differentiation

in the spine [Tables 1 and 2].[5-7,4] They are more frequent in the cervical segment (7), followed by thoracic (5) and lumbar (4) regions. For the spinal lesions, 14 were extradural involving a vertebral body, but only 2 were intradural.

Clinical presentation and magnetic resonance/computed tomography characteristics

All spinal cord lesions presented with back or cervical pain; only 4 had neurological deficits involving the upper and/or lower extremities.[4,9] CT studies showed that these lesions are a hyperdense lesion while MR studies demonstrated hypointense T1- and T2-weighted images with limited edema and contrast enhancement.[1] Differential diagnoses include meningioma, granulomatous lesions, astrocytoma with calcification, oligodendrogliomas, and hamartomas.[7]

Histopathology and immunology

The common histopathologic features of CAPNON include (1) typical chondromyxoid matrix in a nodular pattern; (2) palisading spindle-to-epithelioid cells; (3) variable amounts of fibrous stroma; (4) calcification, osseous metaplasia, and scattered psammoma bodies; and (5) foreign-body reaction with giant cells.[5] The presence of each component is highly variable, and some examples may not show all of the abovementioned elements.[7]

In general, it stains positive for epithelial membrane antigen (EMA) and vimentin and negative for S100 protein and glial fibrillary acidic protein (GFAP). Smith *et al*. found positive immunoreactivity to GFAP and S-100 protein in their report of an "unusual fibro-osseous lesion."[8]

Surgical management and histopathology and immunological assessment

Gross total resection of this calcified intradural lesion adherent to the cauda equina and *filum*

Table 1: Calcifying nonneoplastic pseudoneoplasm

Case	Author and Year	Age/Sex	Localisation	Treatment	Follow-up (months)	Recurrence
1	Duque, 2016	48 F	Left atrium	GT	18	No
2	Kerr, 2011	56 M	Right cerebellopontine angle	ST	6	No
3	Rhodes and Davis, 1978	27 F	Right frontal lobe	ST	84	No
4	Jun and Burdick, 1984	55 M	Corpus callosum	GT	N	No
5	Garen, 1989	44 M	Trigeminal ganglion region	GT	N	No
6	Bertoni, 1990	31 M	Left jugular foramen	ST	156	Yes
7	Bertoni, 1990	48 M	Right cerebellar tonsil	GT	228	No
8	Bertoni, 1990	32 M	Frontal lobe	GT	360	No
9	Bertoni, 1990	58 M	Jugular foramen	ST	N	N
10	Tsugu, 1999	22 F	Right parietal lobe	GT	96	No
11	Shrier, 1999	32 F	Left temporal lobe	GT	12	No
12	Qian, 1999	33 F	Left temporal lobe	GT	31	No
13	Qian, 1999	47 F	Parasagittal frontal	GT	72	No
14	Qian, 1999	49 M	Clivus	GT	90	No
15	Tatke, 2001	6 M	Left temporal medial region	ST	6	No
16	Aiken, 2009	16 M	Right temporal horn	GT	N	No
17	Aiken, 2009	35 M	Right temporal lobe	GT	N	N
18	Aiken, 2009	49 F	Left hippocampus	GT	N	N
19	Aiken, 2009	59 M	Right parietal lobe	GT	N	N
20	Montibeller, 2009	67 F	Right inferior colliculus	GT	18	N
21	Mohapatra, 2010	48 M	Right temporabasal region	GT	N	No
22	Hodges, 2011	36 M	Left cerebellopontine angle	ST	7	No
23	Stienen, 2011	46 M	Right parietal lobe	ST	10	No
24	Stienen, 2011	56 F	Left frontoparietal lobe	ST	22	No
25	Muccio, 2012	55F	Cervicomedullary junction	GT	14	No
26	Nonaka, 2012	56 M	Right temporal lobe	GT	N	N
27	Nonaka, 2012	35 M	Left occipital condyle	ST	6	No
28	Grabowski, 2013	49 F	Pineal region	GT	21	No
29	Fatih, 2014	59 F	Cerebellomedullary cistern	GT	N	N
30	Wisniewski, 2015	29 M	Foramen magnum	GT	2	No
31	Tan, 2016	45 M	Superior medullary velum	GT	3	No
32	Alshareef, 2016	59 F	Cervicomedullary junction	ST	12	No

GT: Gross resection; ST: Subtotal resection; N: Not described

Table 2: Spinal calcifying nonneoplastic pseudoneoplams

Case	Author and Year	Age/Sex	Localisation	Treatment	Follow-up (months)	Recurrence
1	Brock, 2016	72 F	L2id	GT	6	No
2	Duque, 2016	51 F	L2ie	GT	39	No
3	Duque, 2016	46 F	C3io	GT	27	No
4	Duque, 2016	73 M	D2ie	GT	12	No
5	Bertoni, 1990	50 M	FMe	ST	42	No
6	Bertoni, 1990	23 M	Th10e	ST	N	No
7	Bertoni, 1990	58 M	C2e	ST	112	No
8	Bertoni, 1990	12 M	C6e	ST	39	No
9	Bertoni, 1990	32 M	L4e	ST	83	No
10	Bertoni, 1990	33 F	Th9e	ST	N	No
11	Bertoni, 1990	68 F	L4e	ST	16	No
12	Bertoni, 1990	20 F	C2e	ST	N	No
13	Bertoni, 1990	56 F	L4e	ST	N	No
14	Smith, 1994	48 M	L2e	GT	N	No
15	Shrier, 1999	59 M	FMe	GT	24	No
16	Qian et al., 1999	59 M	C1e	GT	46	No
17	Chang, 2000	60 M	C2io	ST	24	Yes
18	Mayr, 2000	58 M	Th10e	ST	48	No
19	Mayr, 2000	63 M	C3e	ST	60	No
20	Liccardo, 2003	40 M	Th8e	GT	36	No
21	Park, 2008	59 F	C7e	GT	N	No
22	Tong, 2010	67 F	L4e	Laminectomy	N	No
23	Ozdemir, 2011	53 M	FMid	GT	N	No
24	Muccio, 2012	57 M	T10-11e	GT	2	N
25	Song, 2015	77 F	T12e	GT	5	No
26	Song, 2015	67 F	L2-3e	GT	N	N
27	Song, 2015	78 F	L1e	GT	N	N
28	Singh, 2016	90 F	C7-D1id	ST	N	N

GT: Gross resection; ST: Subtotal resection; N: Not described; FM: Foramen magnum; TH: Thoracic spinal segment; C: Cervical spinal segment; L: Lumbar spinal segment; E: Epidural; IE: Intradural extramedullary; ID: Intradural; IO: Intraosseous

terminale (size 2.0 × 1.5 × 0.5 cm) was accomplished. Histopathologically, it was calcified (e.g., laminar radiated distribution), accompanied by fibro-osseous metaplastic tissue and a rhyme of small epithelioid cells through the neuropil. Immunohistochemistry demonstrated focal expression of GFAP, S-100 protein, and EMA ("dot" pattern).

With immunohistochemistry, this was a primary CNS lesion composed of glioneuronal cells with probable ependymal origin that differentiated to benign fibro-osseous calcifying tissue. Therefore, the possibility of a neoplastic nature cannot be excluded.

To date all reported cases have been treated surgically, either by complete or incomplete resection.[7] Notably, partially resected tumors showed no regrowth.[4]

Financial support and sponsorship
Nil.

Conflicts of interest
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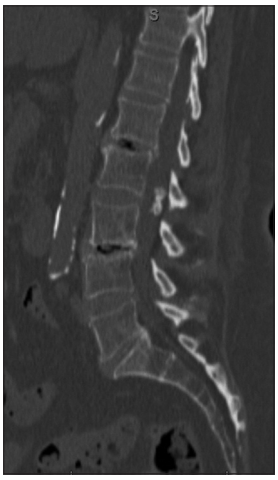

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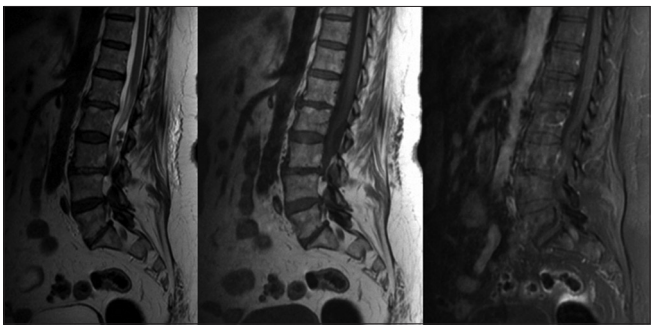

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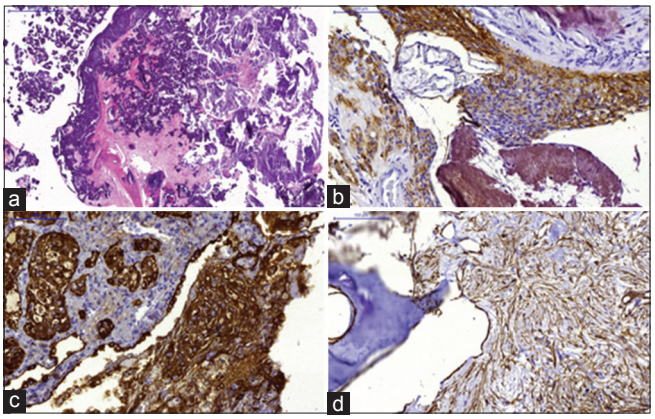

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