Isolated Intramedullary Lumbar Spine Neurocysticercosis: A Rare Occurrence and Review of Literature

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Introduction

Neurocysticercosis (NCC) is the most common parasitic infection of the central nervous system (CNS), caused by the larval form of Taenia solium. Pigs are the intermediate hosts, and humans are the definitive/intermediate hosts. Common risk factors are poor personal hygiene and unsanitary pig raising practices. Spinal cysticercosis is a rather rare clinical occurrence. Intramedullary (IM) spinal NCC is rarer still. Furthermore, cases of IM-NCC at lumbar levels are few and far between. We present a case of a 35-year-old male patient who was diagnosed to have IM-NCC at L2-3 level and was managed surgically with no recurrence at 2 years of follow-up. A systematic literature review (1992–2020) highlights it to be only the third case reported with exclusive lumbar involvement.

Case Report

A 35-year-old man presented with the complaints of low back ache for 12 years, radiating to right leg for 4 months and numbness extending to lateral side of the sole of right foot. On examination, there was a 30% sensory loss in right S1 dermatome as compared with contralateral limb, with no bladder bowel involvement. Patient had no motor deficit. Magnetic resonance imaging (MRI) of the lumbosacral spine was suggestive of IM cystic lesion at L2-3 hypointense on T1-weighted images and hyperintense on T2-weighted images. MRI brain did not reveal any abnormality. Lumbar puncture and serologic studies were not performed.

With the differential diagnosis of neoplastic lesion, the patient was taken up for posterior laminectomy. L2-3 laminectomy was done. A dural bulge was identified. On durotomy, the cord was found to be enlarged. Under microscopic guidance, posterior longitudinal myelotomy was done, the cysts were approached, and subtotal resection of cysts was done. Intraoperatively, three grayish white cysts were identified. Cysts were found to be adherent to the nerve roots causing their inflammation. As a result, one of the cysts could not be excised and was only decompressed. The remaining two cysts were completely excised. Histopathology revealed it to be NCC.

The patient improved postoperatively. Back pain was relieved, and there was significant reduction in radiating pain. He was started on albendazole (15 mg/kg body weight)
for 4 weeks and steroids for 2 weeks. The patient was discharged on the 4th post-operative day. He was followed-up biweekly for the first month. Thereafter, monthly follow-up was done for the next 2 months. MRI done at 6 months confirmed resolution of the cystic lesion. Thereafter, 6 monthly follow-up was done. Patient is symptom free and not on any medication at 2 years of follow-up.

**Discussion**

Spinal NCC was originally reported by Rockitansky. Its infrequent incidence (0.7–5.85%), is attributed to the sieve effect provided by subarachnoid layer which filters cysticerci, thus preventing them to pass. IM involvement occurs in less than one-fifth of the cases with intradural pathology. The cysticerci migrate via hematogenous and ventriculoependymal pathways, thus afflicting mainly the dorsal segment of spinal cord primarily as a consequence of high-blood flow.

In 2017, the Infectious Diseases Society of America (IDSA) and American Society of Tropical Medicine and Hygiene (ASTMH) recommended clinical practice guidelines for the diagnosis and treatment of NCC. The said guidelines strongly advice prescription of corticosteroids in cases of spinal NCC with spinal cord dysfunction and also as an adjunct to antiparasitic treatment. As evidence to recommend one modality of

![Diagram](image-url)
treatment (medical or surgical) over the other is lacking, authors suggested treatment be planned on case basis and surgical expertise available.

**Review of Literature and Results**

The authors searched the PubMed database using keywords “Spinal neurocysticercosis” and “spinal cord neurocysticercosis.” A total of 213 results were obtained which included articles pertaining to both IM and extramedullary (EM) spinal cord NCC lesions.

Research papers reporting exclusive extra spinal involvement, no MRI assessment, published in non-English vernacular, and conducted in nonhuman subjects were excluded from this review (Fig. 1). In the final analysis, 77 articles were shortlisted, encompassing both EM and IM involvement of spinal cord NCC (Table 1). The cumulative number of cases was 147. These include 100 (EM), 46 (IM), and 1 (EM + IM).8

The literature review done by authors revealed only 33 articles (case reports and series) pertaining to the IM-NCC, the earliest being published in 1996. It translates to three articles being published from 1996–2000, followed by nine articles being in print from 2001–2010, and finally 21 articles from 2011–2020. Evidently, there has been increased scientific interest in spinal NCC. Increase in data availability will help to make more evidence-based treatment guidelines possible.

The review brings forth the fact that such cases were found not only in countries of Asia, Mexico but also in countries of the developed world. Eighteen such publications originate from India, followed by eight in the United States, three in Brazil, and one each in China, Guatemala and Spain. A 30-year long (1980–2010) combined research study was undertaken by clinicians from Mexico and India. In most instances, the patients in the developed nations have a history of travel to the endemic region or a history of immigration.10–12 It not only highlights the significance of cultural and environmental impact in this parasitic disease but also the need to consider this rare entity as a differential in such patients by clinicians in the developed world.

The patients with IM-NCC ranged in age from 5 to 70 years with an average of 31.06 years. Among the 46 patients of IM-NCC, 30 were male and 15 were female. In one of the study, this information was not provided.9

Spinal NCC has been reported to occur most commonly in the dorsal spine. The authors found majority (n = 32) of the IM-NCC has been reported to be located in dorsal or dorso-lumbar levels, including two cases occurring at D11-L1 and D12-L1.13,14 It is followed by cervical and cervicodorsal region (n = 12). Highly sporadic occurrence has been reported in the lumbar region. Two of 46 cases of IM-NCC occur purely in the lumbar region.9,15 Our patient is only the third reported case with pure lumbar involvement. With such an unusual occurrence, the diagnosis of the NCC was overlooked, and intraoperative findings were contrasting to our preoperative assessment, compelling us to share our experience.
| S.no | Authors | Country | Year | Age (yr) | Gender | Compartment | S.C level | Symptoms | Investigations | Mx | Remarks |
|------|---------|---------|------|----------|--------|-------------|-----------|----------|----------------|----|---------|
| 12   | Yacoub et al \(^8\) | USA     | 2017 | 49       | M      | EM          | C4-D4, D6-D9 | M, S     | M, S           | M  | P.O albendazole |
| 13   | Muralidharan et al \(^1\) | India   | 2017 | 56       | M      | EM          | CMJ-C4     | M, S, B  | M, EITB        | S  | P.O Albendazole |
| 14   | Pal et al \(^5\)    | India   | 2017 | 44       | M      | EM          | D1-2, D3, D1-9 | M, P, S  | M              | S  | Operated twice. P.O Albendazole given, mimics arachnoid cyst |
| 15   | Yadav et al \(^6\)  | India   | 2017 | 8        | M      | IM          | C5–6      | M, P     | M              | M  | M        |
| 16   | Sharma \(^7\)      | India   | 2017 | 48       | F      | EM          | L2-S2     | P        | M, Eo          | S  | P.O albendazole |
| 17   | Bansal et al \(^6\) | India   | 2017 | 40       | M      | EM          | L5-S1     | P        | M              | S  | P.O Albendazole |
| 18   | Ranga \(^8\)       | India   | 2017 | 6        | M      | IM          | C4-6      | P        | M, S, ELISA    | M  | P.O Albendazole |
| 19   | Hansberry et al \(^8\) | USA     | 2016 | 49       | M      | EM          | Post fossa to C2 | M, S     | M, WBA        | S  | Cranial + |
| 20   | Pant et al \(^9\)  | India   | 2016 | 60       | M      | IM          | D11       | M, R, B  | M              | S  | P.O albendazole |
| 21   | Torous et al \(^10\) | USA     | 2016 | 40       | M      | EM          | L4-S1     | P, M, S, B | M  | S        |
| 22   | Valsangkar \(^11\) | India   | 2015 | 40       | M      | EM          | D10-12    | P, M, B  | M              | S  | P.O Albendazole |
| 23   | Salazar Noguer \(^12\) | Guatemala | 2015 | 43       | M      | IM          | C7-D1     | M, S     | M              | S  | M        |
| 24   | Hackius \(^13\)    | Switzerland | 2015 | 46       | F      | EM          | C1-C2, L4-5 | H, N, D | M, Eo, E      | M  | M        |
| 25   | Cárdenas \(^9\)    | Mexico, India | 2015 | 64       | M      | EM          | CMJ       | M        | S              |   | 30 year study, 19 Mexican, 8 Indian |
| 26   | 57 | M      | EM          | CMJ     | M        | C           |           |          |                |    |          |
| 58   | 60 | F      | EM          | D1-D7   | M, B     | C           |           |          |                |    |          |
| 59   | 64 | M      | EM          | D1-2-3  | M, B, R   | C           |           |          |                |    |          |
| 60   | 65 | F      | EM          | C3-4    | M, B      | C           |           |          |                |    |          |
| 61   | 66 | F      | EM          | C7-D2   | M, B, R   | C           |           |          |                |    |          |
| 62   | 67 | F      | EM          | D5-7    | S        | C           |           |          |                |    |          |
| 63   | 21 | M      | EM          | D5-7    | S        | C           |           |          |                |    |          |
| 64   | 50 | M      | EM          | L4-5    | M, B, R   | C           |           |          |                |    |          |
| 65   | 48 | M      | EM          | L3-4    | M        | C           |           |          | VP diversion   |    |          |
| 66   | 49 | M      | EM          | L3-5    | S, B      | S           |           |          |                |    |          |
| 67   | 52 | M      | EM          | L3-5    | S, B      | S           |           |          |                |    |          |
| 68   | 45 | F      | EM          | C1-5, C5-D8 | M, B, R | S           |           |          |                |    |          |
| 69   | 64 | F      | EM          | D1-2    | S        | S           |           |          |                |    |          |
| 70   | 32 | M      | EM          | C2-3    | S, Z      | M           |           |          |                |    |          |
| 71   | 38 | F      | EM          | L2-4    | M        | M           |           |          |                |    |          |
| 72   | 49 | F      | EM          | CMC-C2  | M        | C           |           |          |                |    |          |
| 73   | 33 | F      | EM          | C3-4, L2-4 | S      | S           |           |          |                |    |          |
Table 1 (Continued)

| S.no | Authors | Country    | Year  | Age (yr) | Gender | Compartment | S.C level | Symptoms | Investigations | Mx | Remarks                                      |
|------|---------|------------|-------|----------|--------|-------------|-----------|----------|----------------|----|---------------------------------------------|
| 62   | F       | EM         | D5-8  | M        | M      | L4          | M         | M        | M, E (CSF, serum) | M  | Brown–Sequard Syndrome                     |
| 50   | F       | EM         | L4    | M        | C      |              |           | M        |                |    |                                             |
| 16   | F       | IM         | D11   | M        | S      |              |           | M        |                |    |                                             |
| 35   | F       | EM         | D12-L1| M        | C      |              |           | M        |                |    |                                             |
| 45   | M       | IM         | CMJ   | M        | S      |              |           | M        |                |    |                                             |
| NA   | NA      | IM         | D2    | M        | S      |              |           | M        |                |    |                                             |
| 16   | M       | IM         | L1    | M, B     | S      |              |           | M        |                |    |                                             |
| 39   | M       | IM         | D12   | M        | S      |              |           | M        |                |    |                                             |
| 28   | M       | IM         | D1–2  | M        | S      |              |           | M        |                |    |                                             |

26 Chaurasia et al21, 2015, India

M, E

Brown–Sequard Syndrome

27 Wang et al44, 2015, USA

45 M

EM

CMJ

H, P

M, serum antibody +

S

P.O albendazole

28 Ganesan45, 2015, India

32 M

EM

L2-S1

P, B, S

M

S

29 Han et al46, 2014, South Korea

59 M

EM

L1-5

P, S, M

M

S

Cranial +

P.O Albendazole

30 Vecchio et al47, 2014, Italy

23 M

EM

L3-4

H, D

M, E

M

31 Amelot et al48, 2014, France

48 M

EM

CVJ

M, H, PS

M

C (VP shunt) Case series of 3 cases, 2 had spinal involvement

25 F

EM

L4-S2

H, P, AMS

M

EVD, S

32 Kim et al49, 2014, South Korea

64 M

EM

D12-L1, L3-4

H, M, B

M

S (VP shunt + laminectomy) Hydrocephalus +, P.O albendazole

33 Qazi4, 2014, India

19 M

EM

D11-L1

M, B

M

S

34 Yoo et al50, 2014, South Korea

42 M

EM

D11-S1

P

M, S.E

S

P.O albendazole

35 Lacoangeli51, 2013, Italy

44 F

EM

L4-5

P, M, S, B

M

S

P.O Albendazole

36 Chandramohan15, 2013, India

15 M

IM

L1

M, B, R

M, Western blot

M

37 Rice et al52, 2012, USA

42 M

IM

D10-D11

M, S

M

S

Brown–Sequard Syndrome

38 De Deo et al52, 2012, Italy

49 M

EM

D6-8, D10-11

H, M

M

S

Cranial +, P.O Albendazole

39 Callacondo et al53, 2012, Peru

NA

NA

All 18 EM

LS M.C

Out of 55 patients with cranial NCC (intraparenchymal + basal cisterns) 18 pt had spinal involvement all EM

40 Jain et al16, 2012, India

20 M

IM

C2

Z, M, B

M, IgG

M

Lost to follow up, Cranial+

41 Shin et al54, 2012, South Korea

48 M

EM

D12-S1

M, B

M

S

P.O Albendazole

42 Agale10, 2012, India

38 M

IM

D11-11

M

M

S

P.O Albendazole

43 Kapu et al55, 2012, India

38 F

EM

D12-L1

P, M, S

M

S

P.O Albendazole, Cysticercal abscess (Continued)
| S.no | Authors            | Country | Year | Age (yr) | Gender | Compartment | S.C level | Symptoms | Investigations | Mx | Remarks                                |
|------|--------------------|---------|------|----------|--------|-------------|-----------|----------|----------------|----|----------------------------------------|
| 44   | Bin & al.          | China   | 2011 | 40       | F      | IM          | D4-5      | M, B, R  | S              |    | P.O anticysticercal agents            |
| 45   | Seo et al.         | South Korea | 2011 | 59       | M      | EM          | D12-L1, L4-5 | H, V    | M, Eo         |    | P.O albendazole, ocular symptoms, Visual defects persisted |
| 46   | Jongwutives et al. | USA     | 2011 | 59       | F      | EM          | L1-4      | M, S, B  | M, S.E         |    |                                        |
| 47   | Park et al.        | South Korea | 2011 | 72       | M      | EM          | L5-S1     | P, M     | M             |    | Cranial + HCP +                        |
| 48   | Lambertucci et al. | Brazil  | 2011 | 23       | M      | IM          | C3-5      | P, M     | M             |    | P.O albendazole                        |
| 49   | Azfar et al.       | India   | 2011 | 10       | F      | IM          | D2        | M, S, B, R | CSF E       | M  |                                        |
| 50   | Vij et al.         | India   | 2011 | 20       | M      | IM          | D10-11    | P, M, S, B | M             | S  | Coexisting IM Schwannoma               |
| 51   | Jang et al.        | South Korea | 2010 | 50       | F      | EM          | L5-S1     | P         | M             | S  | Reoperated, P.O Albendazole            |
| 52   | Boulos et al.      | Canada  | 2010 | 35       | F      | EM          | CMJ       | H, S     | M             | S  | P.O albendazole, Cranial leptomeningeal enhancement + |
| 53   | Lim et al.         | South Korea | 2010 | 42       | M      | EM          | C2-L2     | P         | M             | S  | (D3-5, L1-3) H/O HCP +, P.O Albendazole |
| 54   | Choi et al.        | South Korea | 2010 | 43       | F      | EM          | L5-S1     | P, M, S  | M             | S  | PIVD L5-S1, P.O Albendazole            |
| 55   | Gonçalves et al.   | USA     | 2010 | 62       | M      | IM          | D11       | P, M, S, B | M             | S  |                                        |
| 56   | Chibber et al.     | India   | 2009 | 38       | F      | IM          | D5-S6     | P, M, B  | M, E          | M  |                                        |
| 57   | Shin et al.        | South Korea | 2009 | 45       | M      | EM          | C1-L1     | M, B     | M, CSF ELISA | S  | Cranial + HCP +, P.O albendazole       |
| 58   | Kasliwal et al.    | India   | 2008 | 34       | M      | EM          | C1-C2     | P, M, H  | M             | S  | P.O albendazole                        |
| 59   | Izc et al.         | USA     | 2008 | 70       | M      | IM          | D11-L1    | M, B, R  | M             | S  | P.O albendazole                        |
| 60   | Paterakis et al.   | Greece  | 2007 | 60       | M      | EM          | L3, L5-S1 | P, M, B  | M             | S  | P.O Albendazole                        |
| 61   | Ahmad et al.       | India   | 2007 | 8        | F      | IM          | D1-2      | P, M, B  | M             | S  | P.O albendazole                        |
| 62   | Guedes-Correa et al| Brazil  | 2006 | 53       | F      | IM          | D12-L1    | P         | M             | S  |                                        |
| 63   | Delobel et al.     | France  | 2004 | 45       | M      | EM          | L3-4      | P, M, B  | M, E (CSF, serum) | S  | HIV + cranial +, Brown Sequard syndrome, P.O Albendazole |
| 64   | Torabi et al.      | USA     | 2004 | 35       | M      | IM          | C4, D4-9  | H, P, M, B | M             | M  | Cranial + HCP +, Multilevel            |
| 65   | Alsina et al.      | USA     | 2002 | 38       | M      | EM          | L2-3      | M, B     | M             | S  | Cranial +                              |
|      |                    |         |       | 14       | F      | EM          | C5-D1     | M         | M             | S  |                                        |
|      |                    |         |       | 36       | M      | EM          | C5        | M         | M             | S  | Cranial+                                |
|      |                    |         |       | 40       | M      | EM          | FM        | H         | M             | C  |                                        |
|      |                    |         |       | 28       | F      | IM          | C1        | M         | M             | M  | Cranial +                              |
|      |                    |         |       | 80       | M      | EM          | D4-S, D7-9 | P        | M             | S  |                                        |
| 66   | Colli et al.       | Brazil  | 2002 | 15       | F      | EM          | D9        | M         | M             | S  | HCP +                                  |
|      |                    |         |       | 23       | F      | EM          | D2-L1     | P, M     | M             | S  | HCP +                                  |
|      |                    |         |       | 24       | F      | EM          | L2-S      | P, S     | M             | S  |                                        |
|      |                    |         |       | 36       | F      | EM          | D11-L5    | M, B, R  | M             | O  | HCP +                                  |
|      |                    |         |       | 40       | F      | EM          | C5-6      | M, B     | M             | S  | HCP +                                  |
| S.no | Authors | Country | Year | Age (yr) | Gender | Compartment | S.C level | Symptoms | Investigations | Mx | Remarks |
|------|---------|---------|------|----------|---------|--------------|-----------|----------|---------------|----|---------|
| 43   | F       | EM      |      |          |         | L3-5         | P, M      | M        | S             | HCP+|         |
| 46   | F       | EM      |      | D1-2     | P, M, S | M            | M         | S        | HCP+         |
| 46   | F       | EM      |      | D9-L1    | P, M, B, R | M           | S        | HCP+        |
| 22   | M       | EM      |      | D1       | P        | M            | O         |         | HCP+         |
| 24   | M       | EM      |      | D8-L2    | P        | M            | S         |         |               |
| 24   | M       | EM      |      | L3-451   | P, S, M | M            | S         | HCP+      |
| 51   | M       | EM      |      | C3-7     | M        | S            |           |         |               |
| 67   | Sheehan | USA     | 2002 | 16       | F       | IM           | C1-2      | S        | M             | S  | P.O praziquantel |
| 68   | Homans  | USA     | 2001 | 5        | F       | IM           | D1-1-12   | P, B     | M, EITB      | S  | Cranial + operated twice |
| 69   | Mathuriya et al | India | 2001 | 28       | M       | IM           | D1        | P, M, S, B | M            | S  |
| 55   | M       | IM      |      | D1-2     | P, M, S, B | M          | S        |               |
| 50   | M       | IM      |      | D11      | P, M, S, B | M          | S        |               |
| 70   | Gaur et al | India | 2000 | 22       | F       | IM           | D8        | M, S, B   | CSF ELISA | M  |
| 27   | F       | IM      |      | D3-6     | M, S, B, R | CSF ELISA  | M        |           |
| 71   | Ciftçi et al | USA | 1999 | 30       | F       | EM           | C2-4      | H, P     | M            | NA | HCP+      |
| 72   | Garg et al | India | 1998 | 11       | M       | IM           | D9        | M, S, B   | CSF, ELISA | M  |
| 10   | M       | IM      |      | D8       | M, S, B    | M          | M        |               |
| 73   | Lau et al | Hong Kong | 1998 | 35       | M       | EM           | D1-1-51   | M, S, H, L| M            | S  | Cranial+ |
| 74   | Davies  | Australia| 1996 | 40       | M       | EM           | C3-6      | M, H, P  | M            | S  | Cranial + CSF diversion done multiple times, P.O Praziquantel |
| 75   | Corral  | Spain   | 1996 | 20       | F       | IM           | C         | Z, S, M  | M            | E  | Cranial + |
| 76   | Isidro-Llorens | Spain | 1993 | 30       | F       | EM + IM      | C7-L2, IM at D | M    |               | Operated twice, P.O Praziquantel |
| 77   | Bandres et al | USA | 1992 | 34       | M       | EM           | C2, S1-L3 | P        | M            | M  | A case series of 5 patients, ventricular dilation + |

Abbreviations: CSF, cerebrospinal fluid; CMJ, cervicomedullary junction; CVJ, craniovertebral junction; EM, extramedullary; EITB, enzyme-linked immunoelectrotransfer blot; ELISA, enzyme-linked immunosorbent assay; IgG, immunoglobulin G; IM, intramedullary; VP, ventriculoperitoneal.
Isolated spinal NCC is not a common occurrence. Concomitant cranial lesions are usually present. Of the 147 cases thus reported, 39 patients were known to have a concurrent or a history of cranial involvement. Six of the patients with IM-NCC had such a finding.\(^5,16–20\)

Eighteen of patients with spinal NCC were reported to have hydrocephalus. Only one of these patients had IM-NCC,\(^17\) who also had an evidence of cranial NCC.

The most common symptoms in patients with IM-NCC were those of motor involvement (40), followed by bladder involvement (26), back pain (21), sensory involvement (17), and bowel involvement (6). Two patients had complaints of seizures while headache was seen in one patient. These patients had cervical spine and brain lesions. Two patients had complaints of bladder and bowel involvement (6). Two patients had complaints of sensory and bowel involvement (26), back pain (21), sensory involvement (17), and bowel involvement (6). Two patients had complaints of seizures while headache was seen in one patient. These patients had cervical spine and brain lesions. Two patients (dorsal level IM-NCC) presented with Brown–Séquard syndrome (dorsal lesion).\(^21,22\) One of the patients was found to have a coexisting schwannoma at D10–11 lesion.\(^23\) Patients with pure lumbar involvement had motor symptoms and bladder bowel involvement.\(^9,15\)

MRI is the investigation of choice for spinal NCC. Research papers with no MRI assessment were excluded from this review. MRI is the most essential to make diagnosis of spinal pathology, its level, and compartment involved. Other investigations may not always be helpful. In only 12 of the cases of IM-NCC, antibodies were detected by various techniques, including enzyme-linked immunosorbent assay (ELISA), Western blot, and enzyme-linked immunoelectrotransfer blot (EITB).\(^4,24,25\)

In the review, it was concluded that surgery was the main modality of treatment \((n=29)\), while 16 patients were managed medically with anticysticercal agents (albendazole, praziquantel). One of the patients refused any treatment. Postop medical treatment was given to 12 patients. Redo surgery was required in two cases, both were in dorsal region.\(^4,26\) There is no conclusive evidence pointing to advantage of one modality over the other. Case-based decisions are made, and patients are treated, according to clinical expertise available.

**Conclusion**

IM spinal NCC is a rare occurrence, even scarcer in the lumbar regions. To the author's knowledge, this case study is only the third to be reported in global data, thus adding up to the current literature.

Given its rarity, it is highly likely that such a diagnosis be ignored at the outset. It is prudent to consider this differential, especially in relation to patient history, travel history, personal history, and cultural background, to avoid any surprise. Advocated by clinical judgement, although medical treatment has been followed by similar results, surgical intervention remains the mainstay of treatment of spinal NCC. Although clinicians do prescribe steroids and antiparasitic agents in postop period, strong evidence-based guidelines are needed, necessitating more high-quality research. Steady follow-up is crucial to detect recurrence.

Sources of Support

Nil.

Note

The authors declare in consensus that the information provided is true to the best of their knowledge and currently this manuscript is not under submission in any other journal.

Conflict of Interest

None declared.

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