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2 **Running Title:** Spinal Intradural Neurenteric Cyst in a Septuagenarian

3 **Title of the article:** A Rare Case of Spinal Intradural Neurenteric Cyst in a Septuagenarian
4 Causing Acute Paraparesis

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43 **Abstract**

44 *Background and Aim:* Neurenteric cysts (NCs) account for 0.7 to 1.3% of all spinal tumors and
45 are most commonly present during the first three decades of life. Only two cases have been
46 reported in individuals in their 7th and 8th decades of life. This study described a rare case of
47 an NC in a septuagenarian (70 – 79 years of age) who presented with atypical features of the
48 disease.

49 *Case Presentation:* A 77-year-old female patient presented with acute onset paraparesis.
50 Magnetic resonance imaging revealed a 13.6 × 14.1 × 15.4 mm intradural extramedullary cystic
51 lesion, which was anteriorly located at the C7-D1 level. It was T1 hypointense, T2
52 hyperintense, and showed faint contrast enhancement at the junction between the cyst and the
53 spinal cord. At surgery, a cyst with a greyish-white thin wall was noted in the spinal canal,
54 displacing the spinal cord posteriorly. The cyst wall was partially excised, and a small portion
55 adhering to the spinal cord was left behind. The histopathology report was consistent with an
56 NC. At the 2-week follow-up, the patient's lower limb strength had improved.

57 *Conclusion:* Although rare, NC can present in the seventh decade of life. Magnetic resonance
58 imaging may reveal unusual findings, like T1 hypointensity and faint contrast enhancement of
59 the cyst wall. In elderly patients, a portion of the cyst wall may be left behind if it adheres to
60 the spinal cord to avoid the risk of neurological deterioration.

61 **Keywords**

62 Intradural-extramedullary

63 Magnetic resonance imaging

64 Neurenteric cyst

65 Spinal cord

66 **Highlights**

- 67 • Among all spinal cord tumors, neurenteric cysts (NCs) account for 0.7 to 1.3%.
- 68 • NCs primarily occur during the first three decades of life, with the most common
69 location being the cervicothoracic region.
- 70 • In the majority of cases, NCs are non-contrast enhancing lesions and exhibit isointensity
71 or mild hyperintensity on T1-weighted MRI.
- 72 • We reported a rare case of an NC occurring in a 77-year-old woman, which exhibited
73 atypical findings on the MRI scan, like T1 hypointensity of the cyst and faint contrast
74 enhancement at the junction between the cyst and the spinal cord.
- 75 • Total excision is the treatment of choice; however, in older patients, partial excision of
76 the cyst is safer.
- 77 • **Plain Language Summary**

78 Neurenteric cysts are rare congenital lesions of the spinal axis. The majority of these cysts are
79 located in the cervical region. Patients typically present with symptoms during their first three
80 decades of life, making presentations in the elderly very rare. These cysts are slowly
81 progressive, and symptoms usually have a gradual onset; acute onset symptoms are uncommon.
82 MRI is the investigation of choice and typically shows the cyst located anterior to the spinal
83 cord within the spinal canal. We presented a rare case of an NC at C7-D1 level in an elderly
84 woman who exhibited symptoms of acute onset and showed atypical MRI findings. Surgery
85 is considered for symptomatic patients, and a partial excision of the cyst was performed due to
86 its adherence to the spinal cord. An anteriorly placed intradural cystic lesion at the C7-D1 level
87 should raise suspicion for a neurenteric cyst, even if the patient presents in the seventh decade
88 of life.

89 **Introduction**

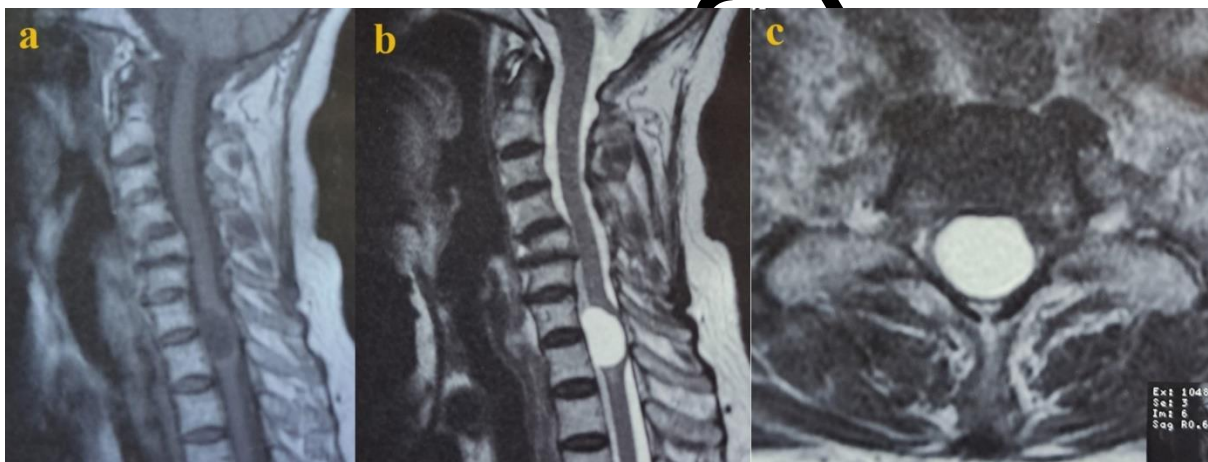
90 Neurenteric cysts (NCs) are rare spinal malformations that account for 0.7 to 1.3% of spinal
91 tumors and 16% of cysts.¹ During embryogenesis, endodermal remnants fail to separate from
92 ectodermal counterparts, resulting in the formation of NCs. These cysts are more common in
93 men and typically present during the first three decades of life. They occur rarely in elderly
94 patients, with only two cases ever described in individuals over 70 years of age.² The majority
95 of NCs show isointensity or mild hyperintensity on T1-weighted magnetic resonance imaging
96 (MRI) scans. The intensity pattern depends on the protein content of the cyst. The cyst usually
97 shows no contrast enhancement, and there is an absence of a mural nodule. These findings help
98 differentiate NCs from more common spinal cord tumors.¹ Wilkins and Odom described three
99 histopathological presentations of NC.³

100 Here, we reported a case of a 77-year-old female patient presenting with acute onset of
101 paraparesis due to spinal cord compression by an NC. This case exhibited hypointensity on T1-
102 weighted imaging and faint contrast enhancement at the interface between the spinal cord and
103 the cyst, which are atypical findings. An NC presenting in an elderly woman in her seventh
104 decade of life with acute paraparesis is very rare.

105 **Case report**

106 A 77-year-old female patient presented to the emergency department with acute onset left-sided
107 chest pain of 1 day's duration. She had a known history of hypertension and coronary artery
108 disease for which she was receiving treatment. The patient was evaluated by a cardiologist and
109 diagnosed with myocardial infarction. She was advised to start antiplatelet medications. While
110 in the hospital, the patient developed acute onset weakness in both lower limbs within 24 hours
111 of admission. Examination revealed Medical Research Committee (MRC) grade 1 power in the
112 right lower limb and grade 2 power in the left lower limb. There was a decrease in sensation
113 over the trunk and both lower limbs. The patient's urinary bladder was catheterized. She

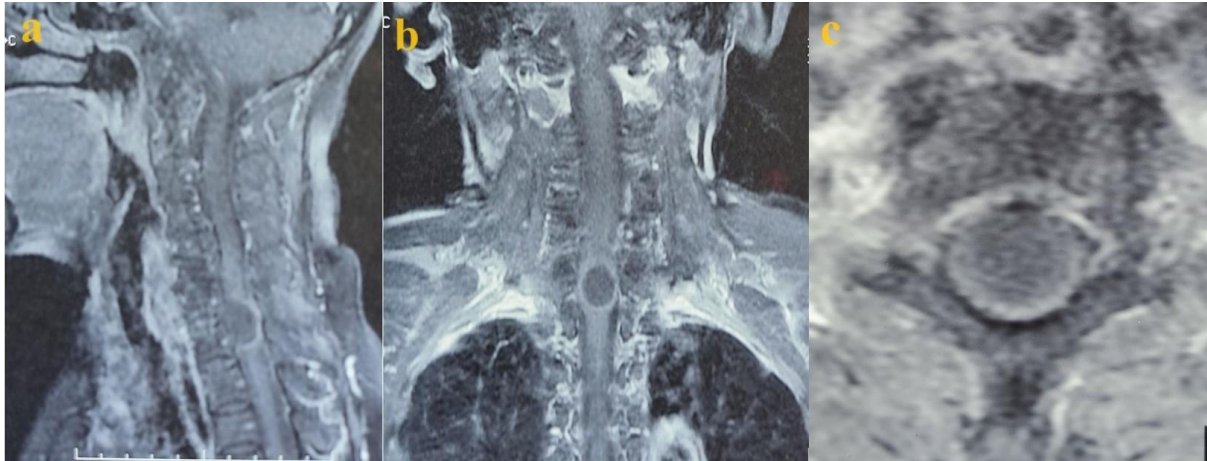
114 underwent an MRI of the brain, which showed a $13.6 \times 14.1 \times 15.4$ mm intradural
115 extramedullary cystic lesion that was anteriorly located at the C7-D1 level. It was T1
116 hypointense and T2 hyperintense on MRI (Figure 1). Contrast MRI showed faint enhancement
117 at the junction between the cyst and the spinal cord (Figure 2). Contrast enhancement was not
118 observed in the anterior part of the cyst, where the spinal cord was absent. The lesion was noted
119 to be almost completely obliterating the spinal canal. The spinal cord was displaced posteriorly
120 and thinned out, with adjacent cord signal changes. No other congenital abnormalities were
121 observed on the MRI of the spine. In light of the anteriorly located cystic lesion at the C7-D1
122 level, the authors' initial diagnosis was neurenteric cyst (NC), with arachnoid cyst as the
123 differential diagnosis.



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125
126 Figure 1. MRI scan of the spine.

127 (a) T1-weighted sagittal scan showing a hypointense lesion at the C7-D1 level. (b) T2-weighted
128 sagittal scan showing an intradural extramedullary hyperintense cystic lesion located anteriorly
129 at the C7-D1 level. (c) T2-weighted axial scan showing the lesion almost completely
130 obliterating the spinal canal.

131 Abbreviations: C: Cervical, D: Dorsal, MRI: Magnetic Resonance Imaging.



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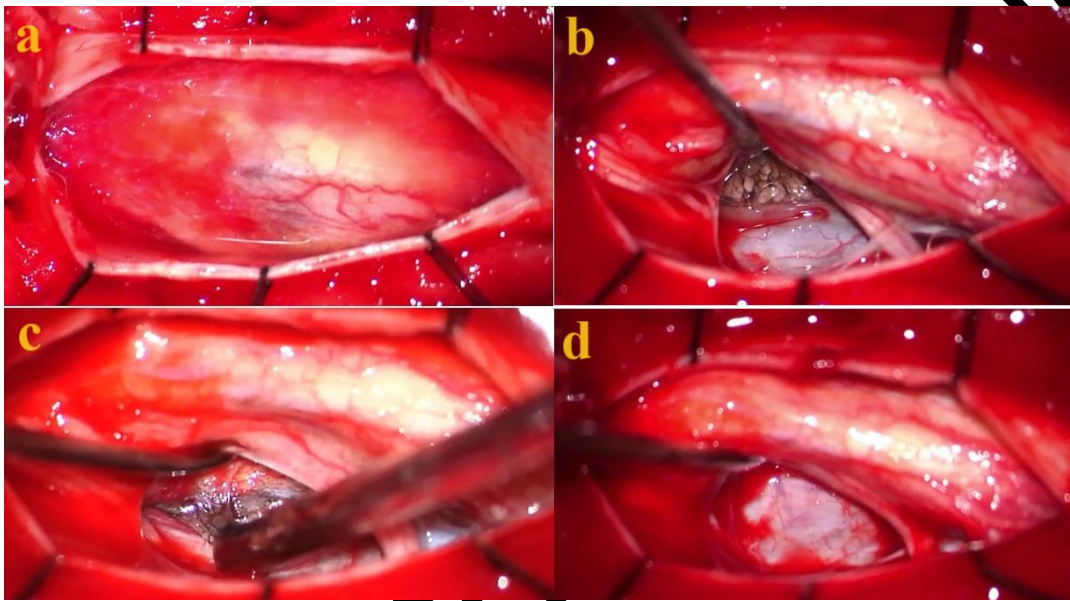
133 Figure 2: Contrast MRI scan of the spine.

134 Note: A sagittal (a) and coronal (b) scan showing faint contrast enhancement at the junction
135 between the cyst and the adjacent spinal cord at the C7-D1 level. Enhancement is not observed
136 in the anterior part of the cyst, where the spinal cord is absent. (c) In the axial scan, the lesion
137 is seen almost completely obliterating the spinal canal, with the spinal cord displaced
138 posteriorly and thinned out.

139 Abbreviations: C: Cervical, D: Dorsal, MRI: Magnetic Resonance Imaging.

140 The patient underwent surgery in a prone position under general anesthesia. A C7-D1
141 laminectomy was done, and the dura was opened. The spinal cord was noted to be bulging
142 posteriorly due to the anteriorly placed lesion (Figure 3a). The arachnoid was opened, and
143 cerebrospinal fluid (CSF) was drained. The lesion in the anterior part of the spinal cord was
144 accessed from the left side of the spinal canal. The spinal cord was mobilized with care. A cyst
145 with a greyish-white thin wall was observed in the spinal canal, displacing the spinal cord
146 posteriorly (Figure 3b). It was identified as an intradural-extramedullary lesion. The cyst wall
147 was opened, and clear fluid was drained. The posterior part of the cyst wall was adhering to the
148 spinal cord (Figure 3c), but it was not adhering to the dura mater. Partial excision of the cyst
149 wall was done, leaving a small portion adhering to the spinal cord. The C7-D1 spinal canal area

150 was found to be free and lax after the excision of the cyst (Figure 3d). The dura mater was
151 sutured, and the wound was closed in layers. At the 2-week follow-up, her power had improved
152 to MRC grade 3 in both lower limbs. The patient was advised to continue physiotherapy.
153 Histopathological examination using hematoxylin and eosin stain confirmed the diagnosis as a
154 neurenteric cyst (NC), resembling the “Type A” pattern of NC proposed by Wilkins and Odum
155 (Figure 4).

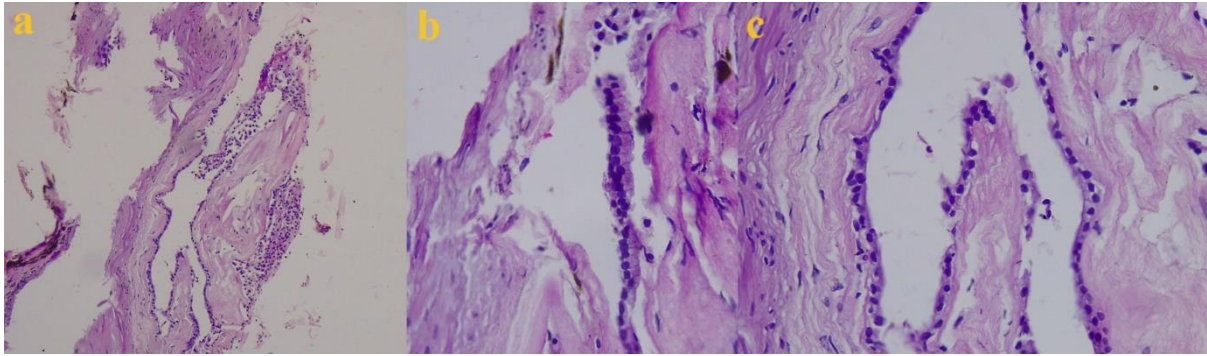


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157 Figure 3: Intraoperative findings of neurenteric cyst excision.

158 (a) The spinal cord was observed bulging posteriorly due to the anteriorly placed cyst, with the
159 arachnoid covering the spinal cord. (b) A cyst with a greyish-white thin wall was seen anterior
160 to the spinal cord. (c) The posterior part of the cyst wall was adhering to the spinal cord. (d)
161 The anterior part of the spinal canal area was free after the excision of the cyst.

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164 Figure 4: Histopathological slides of neurenteric cyst.

165 Note: Haematoxylin and eosin stain (a-10x, b-40x, c-40x) showing a cyst lined by monolayered
166 cuboidal to columnar mucinous cells resting on a collagen layer. No lymphatic, glial, or smooth
167 muscle components were noted.

168 Discussion

169 Holcomb and Matson coined the term NC.⁴ They are also known as intraspinal enterogenous
170 cysts.⁵ At the 3rd week of fetal development, NCs are derived from endodermal remnants
171 during the period of notochordal formation. In about 50% of cases, NCs are associated with
172 vertebral abnormalities, like spina bifida, cleft vertebra, hemivertebra, absent or fused vertebra,
173 or diastematomyelia, and Klippel-Feil anomaly, as well as anomalies that may be fatal.¹

174 These cysts are observed in males twice as often as in females. The mean age of presentation
175 is 22 years (range 1–59 years). It is very rare to see patients presenting after the age of 50.⁵
176 More than half of cases are located in the cervical region. In the spinal canal, the intradural
177 extramedullary compartment is the most common location.¹

178 They commonly present with spine pain and radicular or myelopathic symptoms, such as neck
179 pain, sphincter disturbances, quadriplegia, and paraplegia. It is a slowly progressive disease,
180 and symptoms of acute onset are rare. A thoracic neurenteric cyst with a ten-day history of
181 progressive weakness of the lower limbs was reported by Liu et al. in a 39-year-old female

182 patient ⁵ The case had a rare presentation of acute onset of paraparesis, which occurred within
183 24 hours of admission due to spinal cord compression by an NC.

184 The oldest reported case of an NC was an 81-year-old male patient presenting with myelopathic
185 symptoms in the upper limbs that had lasted for one year. The patient underwent partial excision
186 of the NC at the C1-3 level.² Before this, Kim et al. reported the oldest patient with spinal NC,
187 who was 72 years old.¹ The case involved a septuagenarian (aged 71 to 80 years) who presented
188 with symptoms at the age of 77.

189 MRI is the investigation of choice. It shows a ventrally located T2 hyperintense, non-contrast-
190 enhancing lesion. The T1-weighted image shows isointensity, hypointensity, or hyperintensity
191 of the lesion, depending on the density of the content.⁶ Frequent variations in the MRI findings
192 are observed. An NC showing T1 hypointensity and T2 hyperintensity was reported by Miyagi
193 et al. and Nagi et al.³ One rare case of an NC mimicking an abscess with intense peripheral
194 contrast enhancement were reported.⁷ Preece et al. analyzed five cases of intracranial NCs with
195 posterior rim enhancement at the junction with the brain parenchyma; however, this
196 enhancement did not show any pathological correlation ⁸. In this case, the cyst had a grayish-
197 white thin wall and contained clear fluid. It showed hypointensity on the T1-weighted image
198 and hyperintensity on the T2-weighted image. Contrast MRI revealed faint enhancement at the
199 interface between the cyst and the adjacent spinal cord; however, this contrast rim enhancement
200 of the cyst did not correlate with histological findings. An NC with vertebral anomalies can be
201 diagnosed using a CT scan. The close differential diagnosis of an NC is an arachnoid cyst,
202 which is located dorsal to the cord and shows T1 hypointensity and T2 hyperintensity .⁶

203 Symptomatic patients are managed with surgical excision. Most symptoms disappear following
204 surgery, except in a few cases where they recur or exacerbate.⁵ The posterior approach is the
205 most widely used technique due to fewer intraoperative complications. In this approach, the

206 obscuring cord is manipulated, and the cyst is aspirated before excision. The anterior surgical
207 approach reduces the risk of cyst rupture during surgery, thereby preventing leakage of
208 contents; however, it increases surgical complexity, necessitates instrumented fusion, and raises
209 the risk of complications. No postoperative improvement was seen in 18% of cases, and
210 worsening of symptoms was documented in 11% of cases.³ In the author's case, the NC was
211 located anteriorly at the C7-D1 level and was found to be displacing the spinal cord posteriorly.
212 It was operated on using the posterior approach, and partial excision of the cyst wall was
213 performed. The patient improved following the surgery. Total excision carries an excellent
214 prognosis, but in older patients, it is safer to perform partial excision due to the adherence of
215 the cyst to the spinal cord. The recurrence rates have ranged between 0% and 37%.⁵

216 Hematoxylin and eosin staining of the NC showed a collection of mucin-producing simple
217 columnar or cuboidal ciliated and non-ciliated goblet cells surrounding a central cystic cavity.
218 Wilkins and Odom classified three histopathological presentations of NC. The author's case
219 exhibited the 'Type A' pattern proposed by Wilkins and Odom. Type A cysts contain either
220 columnar or cuboidal cells, with ciliated and non-ciliated components atop a basal membrane
221 composed of type IV collagen.³

222 Our patient was a 77-year-old woman with a recent history of myocardial infarction (MI). She
223 developed acute onset paraparesis due to spinal cord compression caused by an NC. This
224 presented a difficult situation for the authors, as the cardiac risk was higher for this elderly
225 patient with a recent MI if she underwent surgery in the prone position. Since surgery was
226 unavoidable, the patient underwent surgery after receiving clearance from the cardiologist. She
227 recovered well in the postoperative period. This is a very rare case of an NC presenting in a
228 septuagenarian with acute onset paraparesis.

229 **Conclusion**

230 An anteriorly placed intradural cystic lesion at the C7-D1 level should raise the suspicion of a
231 neurenteric cyst, even if the patient presents in the seventh decade of life. An MRI scan of an
232 NC may show uncommon findings, such as T1 hypointensity and faint contrast enhancement
233 of the cyst wall. In elderly patients, part of the cyst wall may be left behind if it is adhering to
234 the spinal cord to avoid the risk of neurological deterioration.

235

236 **Ethical Considerations**

237 **Compliance with ethical guidelines:** Informed consent was obtained from the patient.

238 **Funding:** This case report did not receive any grant from funding agencies in the public,
239 commercial, or non-profit sectors.

240 **Conflicts of interests**

241 The authors declared no conflicts of interest.

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