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Spinal Hydatid Cyst - A rare case report

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I. Introduction:

Hydatid disease is a zoonosis caused by Echinococcus tapeworm. Parasite usually infects the host in the larval stage. The two main species causing hydatid disease in humans are E.granulosus and E.multilocularis. E.granulosus is seen more commonly in the Mediterranean region, Africa, South America, Australian subcontinent especially in rural areas.E.multilocularis is relatively less common. Liver & lungs arethe most commonly encountered type of hydatid disease in humans.(1)The classical radiological findings in hydatid disease are commonly known; however findings when disease complications occur or if disease occurs at unusual anatomic locations are less commonly described in the literature.(2)

Hydatidosis of bone accounts for only 0.5% to 2.5% of hydatid disease in humans. It is one of the severest forms of the disease.Itis asymptomatic for a long time, so usually detected at a later advanced stage, when radiologically the lesions are already extensive. Most frequent involved bones are vertebrae and pelvic bones , where disease is usually very severe. (3)Adequate radiological suspicion is important as imaging features may closely resemble infective etiology.

II. Case Report:

Presented is the case of 36 year old female who came with chief complaints of insidious onset back pain, progressive lower limb weakness and sensineural disturbances since one year. Patient has fecal and urinary incontinence for the last one month.ForPatient is non-hypertensive , non-diabetic and non-vegetarian in diet. Patient had no history of any previous similar episode. Chest X ray of patient was normal. Keeping in view of insidious onset and progressive nature of symptoms , MRI of whole spine was advised.

Contrast MRI brain and spine was performed. MRI spine revealed multilevel involvement of dorso-lumbar spine. At L2 vertebral level, The lesion predominantly involves right half of vertebral body and right posterior elements and extending into adjacent extradural space and bilateral neural foramen (L2-L3 level) resulting in partial vertebral body collapse and compression of cauda equina nerve roots (fig.1a,1b,1c). The lesion also infiltrates right psoas muscle. The lesion appears hypointense on T1WI and shows mixed signal intensity on T2WI.Multiple small round T2 hyperintense cystic lesions (daughter cysts) are noted within the lesion giving multivesicular appearance. On post contrast sequences, the lesion shows peripheral enhancement.

Another large illdefined lesion of similar signal intensity was seen involving right posterior elements of D10,D11 and D12 vertebrae and extending into epidural space of spinal canal causing severe narrowing of thecal sac, cord compression resulting in altered cord signals at these levels(fig.2a,2b,2c). The lesion was seen extending into right paravertebral space and infiltrates right paraspinal muscles (fig.2c,2d,2e,2f).Posteriorlythe lesion is also seen extending into subcutaneous plane indorso-lumbar region (fig.2e).

A large well defined cystic lesion was seen in spleen (size, 10.0x7.2 cm) showing multiple T2 hypointense folded membranes, characteristic of hydatid cyst (fig.3a,3b).

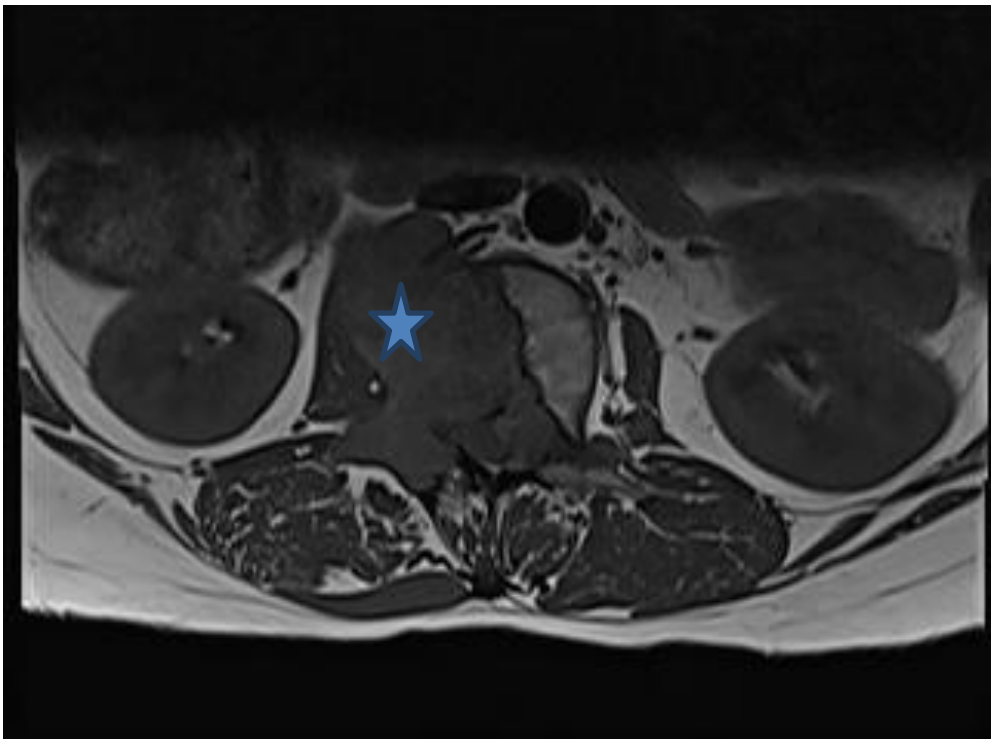


Fig 1a

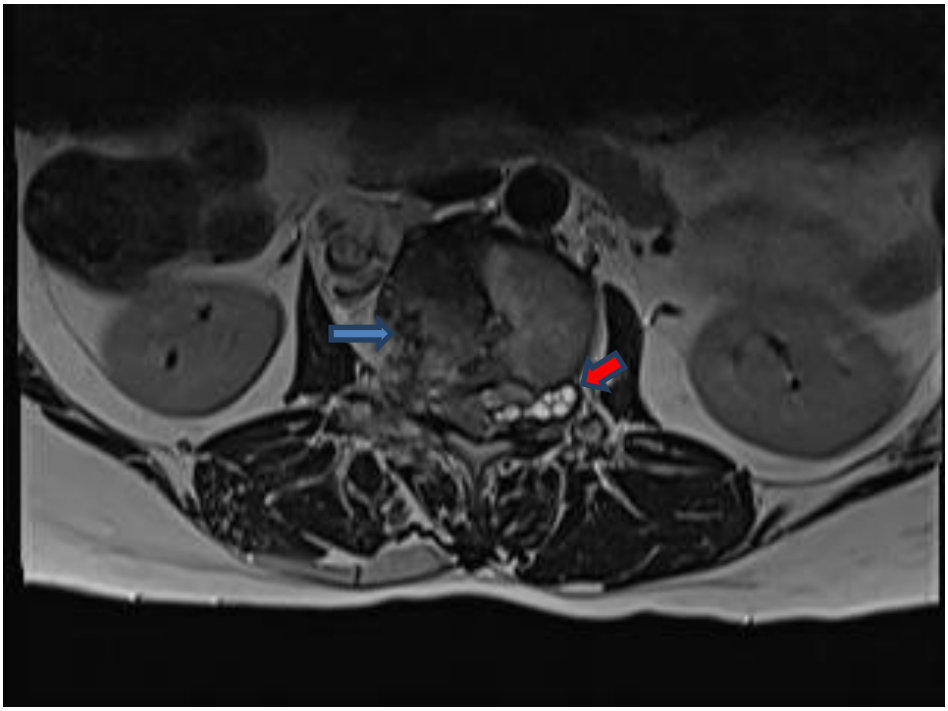


Fig 1b

1c

Fig.1: A 36 years old woman with spinal hydatid disease: (a) Axial T1WI shows a hypointense lesion predominantly involves right half of L2 vertebral body and right posterior elements (star), and extending into adjacent extradural space (arrow) and bilateral neural foramen. (b) Axial T2WI shows, mixed signal intensity lesion in involved vertebral body and posterior elements(arrow), extradural intraspinal space with multiple small rounded hyperintense daughter cysts (multivesicular appearance) in left neural foramen level(red arrow). Infiltration of right psoas muscle is also noted(star). (c) Sagittal T2WI shows, partial collapse of involved segment of L2 vertebral body (star).

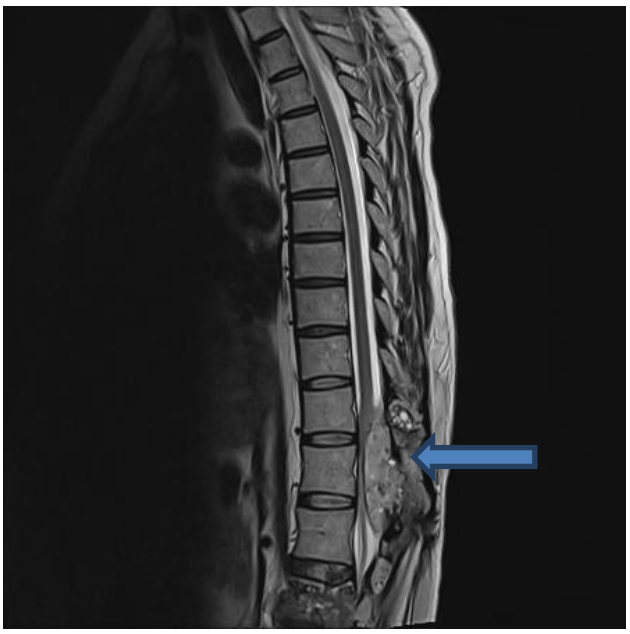


Fig 2a

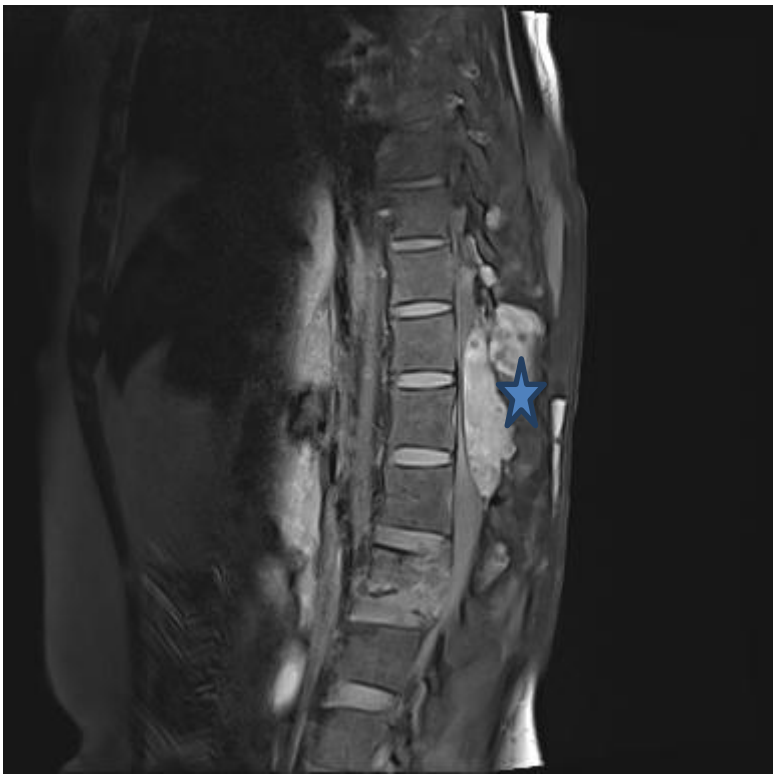


Fig 2b

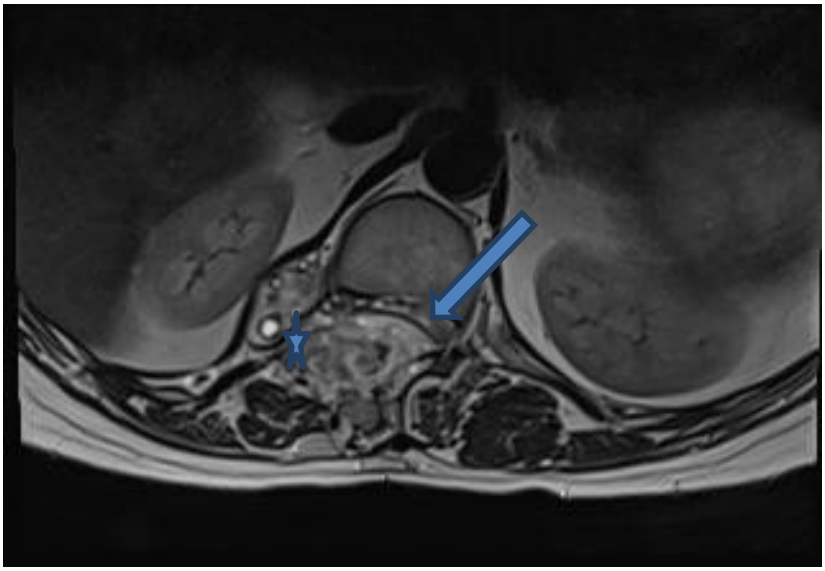
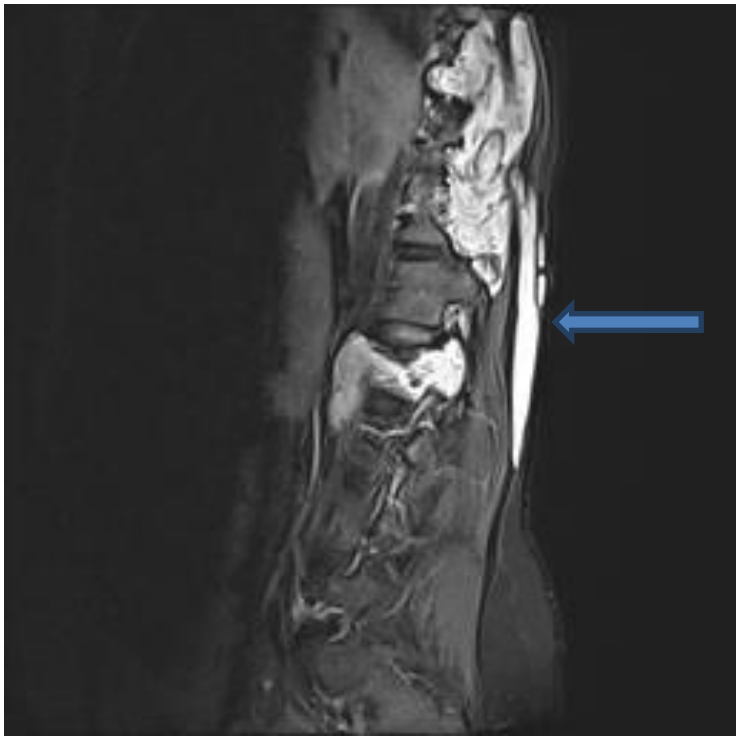
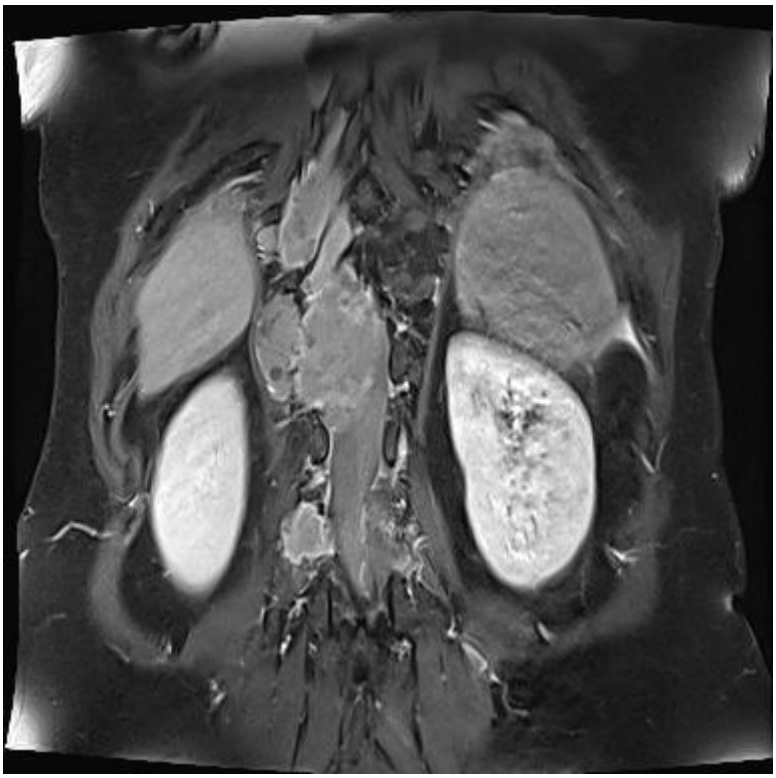


Fig 2c

2d

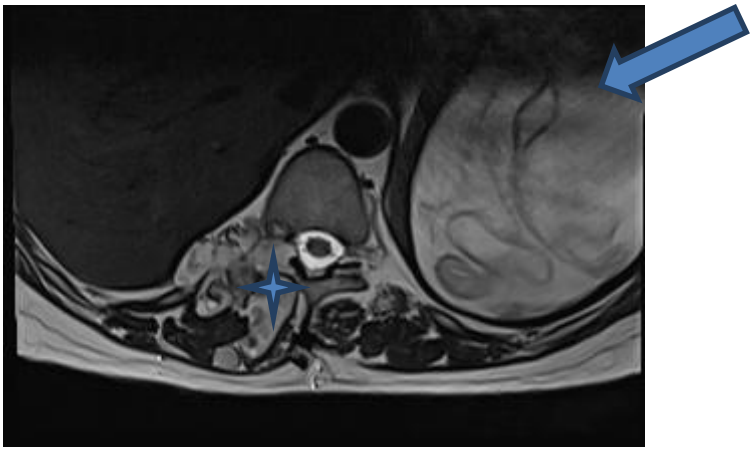


2e

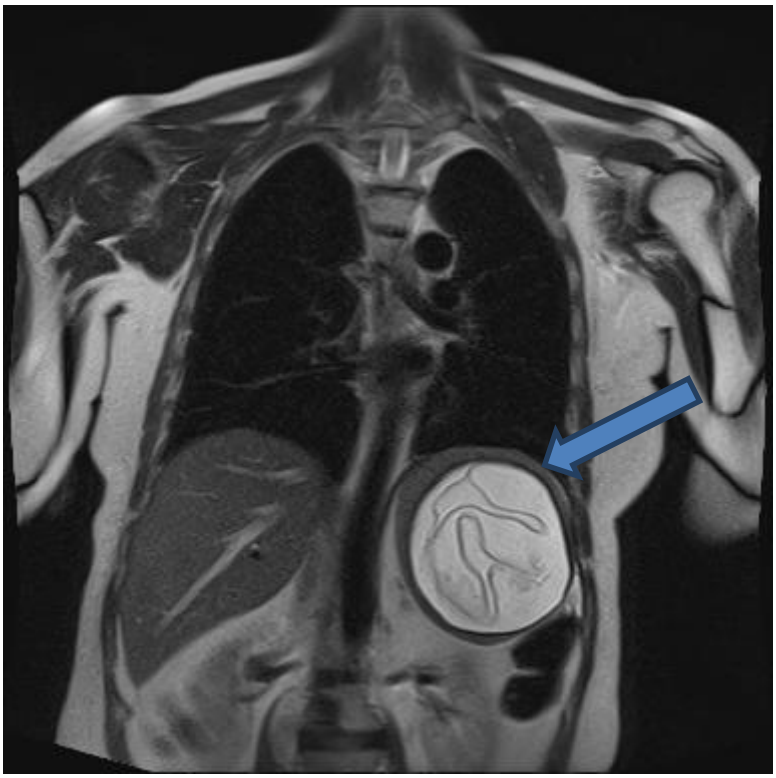


2f

Fig.2:A 36 years old woman with spinal hydatid disease: (a) Sagittal T2WI shows intermediate signal intensity lesion in posterior epidural space at D10-D12 vertebral levels (arrow). (b) Sagittal STIR image shows hyperintense lesion in posterior epidural space at D10-D12 vertebral levels and L2 vertebral body (asterisk). (c) Axial T2WI shows mixed signal intensity lesion in epidural space of spinal canal causing severe narrowing of thecal sac and cord compression resulting in altered cord signals at D11 level (arrow). Extension of the lesion is also seen into right paravertebral space and right paraspinal muscle (asterisk). (d) Sagittal T2WI image shows mixed signal intensity lesion in right neural foramen and posterior elements at D10-D12 vertebral levels with matrix (asterisk), daughter cysts (triangle)and membranes (thin arrow). (e) Sagittal STIR image shows hyperintense lesion in right paravertebral space and right paraspinal muscle extending into subcutaneous plane in dorso-lumbar region (arrow). (f) Post contrast coronal image shows peripheral enhancement around the lesion.



3a



3 b

Fig.3:A 36 years old woman with spinal hydatid disease: (a) Axial T2WI shows a well defined hyperintense lesion with internal folded membranes in spleen (arrow), characteristic of hydatid cyst. Spinal lesion is also visualized (star). (b) Cor T2WI shows a well defined hyperintense lesion with internal folded membranes in spleen (arrow), characteristic of hydatid cyst.

III. Discussion:

Liver (60–70%) and lungs (10–15%) are the most commonly affected visceral organs in hydatid disease. Bone involvement isobserved only in just 0.5–2% of all cases. Most common involved bone is vertebrae.[4] . Thoracic vertebrae are most commonly involved (60%), followed by lumbosacral vertebrae (35%) and cervical vertebrae (5%).[5].

Pathophysiologically, the larva spreads to the paraspinal tissues or epidural space by perforating the cortex, thenextending from the anterior vertebral body , to the extradural space or paraspinal region laterally, or to the spinal canal posteriorly, compressing the spinal cord.[5]The cyst involving multiple vertebrae is rare. Intervertebral discs act as a strong barrier to extension of hydatid cyst.[6]Fibrous pericyst is not formed in bones and, thus, peripheral capsular calcifications are not seen. The cyst starts growing along least resistance path and thus acquires an irregular & branching pattern, so typical spherical masses are not seen.[7]

Clinical manifestations are variable, depending upon the level of the vertebrae affected and the stage of the disease. "Braithwaite and Lees classified the spinal disease in five types: 1) primary intramedullary hydatid cyst, 2) intradural extramedullary hydatid cyst, 3) extradural intraspinal hydatid cyst, 4) spinal hydatid cyst disease of the spine and 5) paravertebral hydatid cyst disease". [8]Usually there are no diagnostic signs or symptoms of spinal hydatid disease other than caused by spinal cord compression.[9] Cord or nerve root compression can be caused by vertebral fracture or due to direct pressure effect of hydatid cyst, then the patient will present with continuous back pain or radicular pain.[10]. Symptoms may progress to paraplegia in 25–84% of patients. The symptoms in our patient, diagnosed one year after the onset, were back pain, paraplegia and urinary and fecal incontinence.

In our case, the hydatid cyst was located in the vertebral and paravertebral regions of the thorax and extradural intraspinal regions of the lumbar spine.

Radiological work up is mainstay for further management of the patient. Plain radiography, Ultrasound, CT and MRI all can play role for adequate diagnosis.[11] CT is useful for screening of the damaged vertebraand MRI is the imaging modality of choice.[12]

Plain radiographs will show a well-defined multiloculated osteolytic lesion with associated bone expansion, thinning of cortex and extension into adjacent soft tissues. There may or may not be calcification of

extraosseous part of cyst , but the intraosseous part will not. There is sometimes no clear boundary between the normal and pathologic portions of the bone[13].

NCCT will show lytic lesions, usually multiple and are irregular in shape. Endosteal scalloping can be noted when the cyst erodes the cortical bone, no periosteal reaction is seen.[14]. this finding can be useful in the differential diagnosis of other lytic lesions.

MR imaging is required to look for the pathognomonic daughter cysts and for preoperative planning. On MR images, the cystic lesions have thin wall and usually follow CSF intensity i.e on T2-weighted images have high signal intensity (due to high water content) and on T1-weighted images show low signal intensity in majority of cases.The lesion shows no internal gadolinium uptake.[6] Our CT and MRI results conformed to those in the literature. If there is superimposed infection or debris present , it can affect the typical appearance of a hydatid cyst, making it look like a complex cystic or solid lesion that can be confused with a tumor.[15]Upon contrast administration the cyst shows peripheral enhancement with no central enhancement. This helps in differentiating it form possible infectious or neoplastic conditions. [16].The presence of the visceral hydatid cyst should alert the physician against the spinal hydatid cyst disease.[17]Usually, wide surgical excision like a neoplastic lesion , in combination with adjuvant antihelminthic therapy, is the most preferred treatment. Most commonly used adjuvant antihelminthics are (3%, 10%, 20%) hypertonic saline, 0.5% betadine, 0.5% silver nitrate and 2% formalin.[10]In conclusion, the spinal hydatid cyst is a severe disease with significant morbidity. The disease may not be fully resolved via surgical or medical treatment. Medical treatment is recommended by almost all authors although it is not absolutely effective in preventing the relapse.

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