

Lesion of the Femoral Nerve Caused by a Hydatid Cyst of the Right Psoas Muscle

Dragoljub Bilanović, Darko Zdravković, Tomislav Randjelović, Borislav Tosković, Jasna Gačić

Department of Surgery, Medical Centre "Bežanijska kosa", Belgrade, Serbia

SUMMARY

Introduction Hydatidosis is a human disease caused by the larval form of *Echinococcus granulosus*. All organs in the human body may be affected by hydatid disease, but excluding liver and lungs, all other organs are considered as uncommon locations. Hydatid disease located in the psoas muscle is uncommon.

Case Outline The authors present a 36-year-old male living in endemic areas of Serbia, admitted due to pain and weakness of the right thigh and weight loss. Duration of symptoms was one year. CT and MRI revealed a big cystic mass (20 cm long) in the right psoas muscle. Neurological investigation showed a loss of function of the right femoral nerve. Serology for Echinococcosis was negative. Surgery was indicated and performed by median laparotomy. Total excision of the cyst was done. Pathohistology confirmed the nature of the cyst. Three years after operation the patient was without any signs of disease relapse.

Conclusion Echinococcal disease of the psoas has been very rarely reported, sometimes associated with paraspinal disease and often with vertebral involvement. Cystic or complex retroperitoneal tumour, pyogenic abscess of the psoas and even tuberculosis should be considered in the differential diagnosis. Treatment of choice is surgery. The greatest danger for the patient is dissemination and anaphylactic reaction. Also, compression of adjacent organs may produce significant morbidity.

Keywords: hydatidosis; femoral nerve; paresis

INTRODUCTION

Hydatid disease is still a serious health problem, especially in many Mediterranean countries, the Middle East, South America, New Zealand and Australia. It is an infection of humans caused by the larval stage of *Echinococcus granulosus* [1]. Humans are accidental hosts that become infected by ingesting its eggs and, just like in the intermediate hosts, allow the development of cysts in various organs. The growth rate of the cysts is about 1 cm per year [2, 3]. Hydatid disease in extrahepatic locations usually remains asymptomatic unless the cyst grows and produces symptoms due to pressure, rupture to the pleural or peritoneal cavity, secondary infection or an allergic reaction [4, 5]. Hydatid disease located in the psoas muscle is very uncommon. In this paper, we describe a rare case of hydatidosis of the psoas muscle.

CASE REPORT

The authors present a 36-year-old male from endemic areas of Serbia. The patient came from rural area where sheep raising and contact with dogs was common. He was admitted to our Surgical Department because of abdominal pain, weakness of the right thigh and weight loss. His symptoms lasted for one year. He was treated at a local Medical Centre by the physiotherapist.

Ultrasound (US), magnetic resonance imaging (MRI) and computed tomography (CT)

demonstrated right heterogeneous solitary lesions in the psoas muscles in close contact with the kidneys and extending from L1 to S2 (Figures 1, 2 and 3). The lesion was multiloculated. Neurological investigation showed a loss of function of the right femoral nerve. Serology investigation for echinococcus was negative.

As there was indication for surgery, median laparotomy was performed and cystic lesion of the right psoas muscle was confirmed. There was infiltration of the right femoral nerve and compression of the right ureter. In our case, total cystectomy was carried out with meticulous dissection of the left ureter, gonadal vessels, hypogastric nerve plexus and the right femoral nerve (Figure 4). *Echinococcus granulosus* was confirmed by pathohistology. Postoperative period was without complications. There was no evidence of disease 3 year after surgery. The function of the right femoral nerve and muscles of the right thigh were improved.

DISCUSSION

Primary cyst may localize anywhere in the body but commonly they are found in the liver (55-70%) and lungs (20-30%) [6]. Other locations of the echinococcus include spleen (0.9-8%), kidneys (1-4%), pancreas (0.25-0.75%), brain, heart, ovaries, bones and abdominal wall [7].

The musculoskeletal involvement has been registered in only 1-4% of the cases [8]. Nevertheless, some cases of primary muscular

Correspondence to:

Dragoljub BILANOVIĆ
Medical Centre "Bežanijska kosa"
Autoput bb, 11070 New Belgrade
Serbia
biledr@bkosa.edu.rs



Figure 1. MRI of the abdomen: a large hydatid cyst of the right psoas muscle cause compression and dislocation of retroperitoneal structures

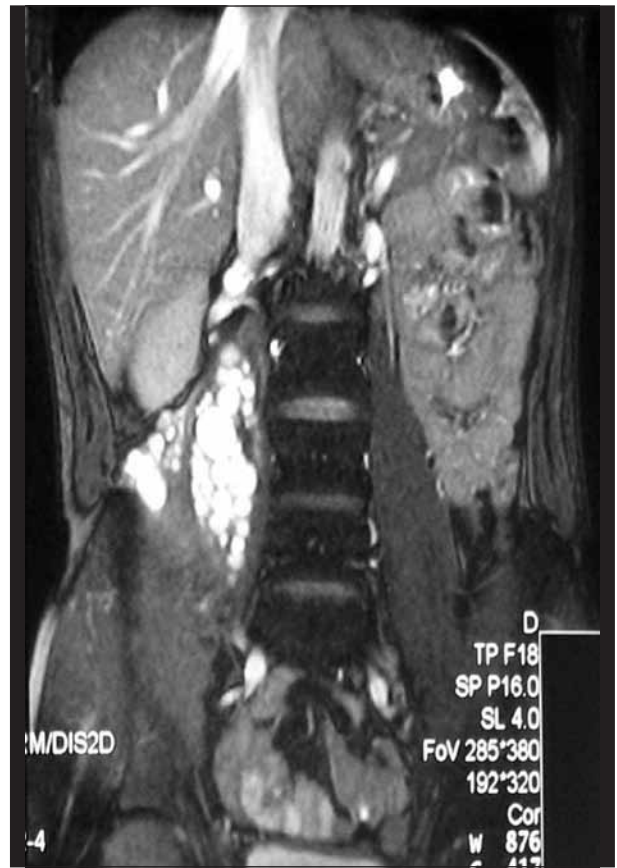


Figure 2. MRI of the abdomen: a large hydatid cyst of the right psoas muscle infiltrating the spine and retroperitoneal nerves

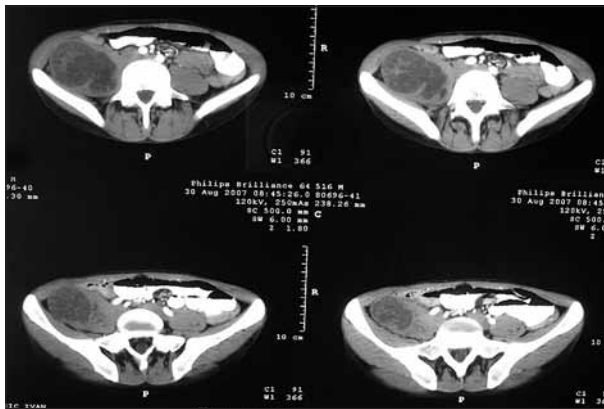


Figure 3. CT image of a large retroperitoneal hydatid cyst infiltrating the abdominal wall, spine and pelvis

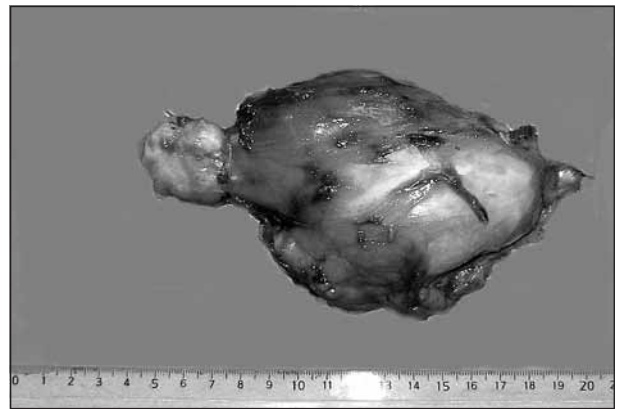


Figure 4. Macroscopic aspect of the hydatid cyst after surgical excision

hydatidosis at various sites have been reported, i.e., sartorius, biceps brachii, supraspinatus, gluteus, pterygoideus and soleus muscles [9-14]. Hydatid disease located in the psoas muscle is very uncommon. Only some thirty-odd such cases have been reported [15]. Symptoms in such cases occur because of pressure or complications including rupture, allergic reaction and secondary infection [16]. The pathogenesis of muscular localization is still not well understood. Most authors believe that embryo can reach the muscles from the systemic circulation after leaving the intestine and passing through the liver and the lungs. Several factors could explain the exceptional nature of muscle localizations of hydatid cysts: efficacy of the hepatic and pulmonary barriers,

and muscle environment unfavourable for growth of hydatid larvae [10]. Muscle function, however, may inhibit, both mechanically and by the lactic acid, implantation of the embryo. Previous trauma and the functional status of the muscle may also have a role in conditioning the implant within the muscle [6]. A review of the literature shows that commonly involved muscles are those of the trunk and proximal limbs as in our case. It could be explained by the volume of the muscle mass and its rich blood supply. In recently published works the left psoas muscle is dominantly affected, which is contrary to our patient [17]. Many authors have a diagnostic problem of making the right preoperative diagnosis of primary hydatid disease of the skeletal

muscle. Diagnosis of hydatid disease may be difficult in the case of muscular localization, especially outside endemic areas. The development of a mass inside a muscle more commonly implies the presence of a haematoma, tumour or septic lesion.

Clinically, the growth of the cyst may produce local symptoms related to compression of adjacent structures, as is our case. Skin and serological tests are helpful in the diagnosis, but often are negative, because the capsule isolates the parasite from the host's immune system [18]. US is the method of choice for the detection of hepatic and extrahepatic echinococcal cysts [19]. CT confirms the diagnosis by revealing the presence of daughter cysts and plaque-like calcifications in the cystic wall. It is important as it provides information regarding the exact location of extrahepatic cysts in relation to neighbouring structures. CT sensitivity ranges from 90% to 97% [20]. Although CT findings are suggestive, a high index of clinical suspicion is needed to establish the diagnosis. MRI was helpful in our case to assess the extent and location of the lesion and evaluate spinal involvement. MRI can also assist in defining treatment strategy. Cystic or complex retroperitoneal tumour, pyogenic abscess of the psoas and even tuberculosis must be considered in the differential diagnosis [21]. Therapy for extrahepatic echinococcal disease is based on considerations regarding the size, location and manifestations of the cysts, and the overall health status of the patient [22]. Surgery is the mainstay of treatment for hydatid cysts. The principle of surgical therapy is total excision of the cyst or cysts whenever possible. The surgeon must be careful to remove the cyst totally avoiding dissem-

ination of cyst contents. It is important that the abdominal cavity is isolated with gauzes soaked in 20% hypertonic saline solution to avoid secondary hydatosis and allergic reaction [23]. The lumbar extraperitoneal approach is the safest access in order to avoid peritoneal dissemination for cysts localized in the psoas muscle. We performed surgical procedure through median laparotomy. Adjuvant chemotherapy with anthelmintics is recommended to reduce the risk of dissemination during surgery and to prevent recurrence. According to the WHO guidelines for the treatment of hydatid disease, chemotherapy is indicated for inoperable patients and those with multiple cysts scattered in many organs where surgery can be ineffective or hazardous [24].

In the case we report, entire cysts were removed, then we did not require adjuvant chemotherapy. The patient remained asymptomatic during the follow-up without recurrence. We found less than 30 cases of hydatid disease of the psoas muscle on Medline. Echinococcal disease of the psoas has been reported very rarely, sometimes associated with paraspinal disease and often with vertebral involvement. Although CT findings are suggestive, a high index of clinical suspicion is needed to establish the diagnosis. MRI was helpful in our case to assess the extent and location of the lesion and evaluate spinal involvement. MRI can also assist in defining treatment strategy. Cystic or complex retroperitoneal tumour, pyogenic abscess of the psoas and even tuberculosis must be considered in the differential diagnosis. The greatest danger for the patient is dissemination and anaphylactic reaction. Also, compression of adjacent organs may produce significant morbidity.

REFERENCES

- Vicidomini S, Cancrini G, Gabrielli S, Naspetti R, Bartoloni A. Muscular cystic hydatidosis: case report. *BMC Infect Dis.* 2007; 30:7-23.
- Eckert J, Deplazes P. Biological, epidemiological and clinical aspects of Echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev.* 2004; 17(1):107-35.
- Garippa G. Updates on cystic echinococcosis (CE) in Italy. *Parassitologia.* 2006; 48:57-9.
- Tepetes K, Christodoulidis G, Spyridakis M, Hatzitheofilou M. Large solitary retroperitoneal echinococcal cyst: a rare case report. *World J Gastroenterol.* 2007; 13(45):6101-3.
- Engin G, Acunaş B, Rozanes I, Acunaş G. Hydatid disease with unusual localization. *Eur Radiol.* 2000; 10(12):1904-12.
- Melis M, Marongiu L, Scintu F, Pisano M, Capra F, Zorcolo L, et al. Primary hydatid cysts of psoas muscle. *ANZ J Surg.* 2002; 72(6):443-5.
- Hamamci EO, Besim H, Korkmaz A. Unusual locations of hydatid disease and surgical approach. *ANZ J Surg.* 2004; 74:356-60.
- Merkle EM, Schulte M, Vogel J, Tomczak R, Rieber A, Kern P, et al. Musculoskeletal involvement in cystic echinococcosis: report of eight cases and review of the literature. *Am J Roentgenol.* 1997; 168:1531-4.
- Rask MR, Lattig GJ. Primary intramuscular hydatidosis of the sartorius. *J Bone Joint Surg Am.* 1970; 52:582-4.
- Duncan GJ, Tooke SM. Echinococcus infestation of the biceps brachii. A case report. *Clin Orthop Relat Res.* 1990; 261:247-50.
- Tatari H, Baran O, Sanlidag T, Gore O, Ak D, Manisali M, et al. Primary intramuscular hydatidosis of supraspinatus muscle. *Arch Orthop Trauma Surg.* 2001; 121:93-4.
- Combalia A, Sastre-Solsona S. Hydatid cyst of gluteus muscle. Two cases. Review of the literature. *Joint Bone Spine.* 2005; 72(5):430-2.
- Turki I, Turki A, Khoctali H, Bakir D, Bakir A. Pterygoidien hydatid cyst. *Rev Stomatol Chir Maxillofac.* 2005; 106:27-9.
- Togrul E, Kalaci A, Sarpel Y, Koltay IS, Ozbarlas S. What's your diagnosis? Solitary hydatid cyst of the soleus muscle. *Ann Saudi Med.* 2004; 24:288, 307-9.
- El Malki HO, El Mejdoubi Y, Bouslami B, Mohsine R, Ifrine L, Belkouchi A. Hydatid cyst of the psoas muscle. *Sante.* 2007; 17(3):177-9.
- Prousalidis J, Tzardinoglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. *World J Surg.* 1998; 22:17-22.
- Bedioui H, Nouira K, Daghfous A, Ammous A, Ayari H, Rebai W, et al. Primary hydatid cyst of the psoas muscle: description of 9 cases in Tunisia and review of the literature. *Med Trop.* 2008; 68(3):261-6.
- Feki W, Ghozzi S, Khiari R, Ghorbel J, Elarbi H, Khouni H, et al. Multiple unusual locations of hydatid cysts including bladder, psoas muscle and liver. *Parasitol Int.* 2008; 57(1):83-6.
- Dahniya MH, Hanna RM, Ashebu S, Muhtaseb SA, el-Beltagi A, Badr S, et al. The imaging appearances of hydatid disease at some unusual sites. *Br J Radiol.* 2001; 74(879):283-9.
- Taori K, Sanyal R, Rathod J, Mahajan S, Jajoo G, Saxena V, et al. CT appearances of hydatid disease at various locations. *Australas Radiol.* 2006; 50(4):298-305.
- Guthrie JA, Lawton JO, Chalmers AG. MR appearances of primary intramuscular hydatid disease. *Clin Radiol.* 1996; 51:377-9.
- Angulo JC, Granell J, Mugerza J, Sanchez-Chapado M. Primary bilateral hydatidosis of the psoas muscle. *J Urol.* 1999; 161(5):1557-8.
- Karavias DD, Vagianos CE, Kakkos SK, Panagopoulos CM, Androulakis JA. Peritoneal echinococcosis. *World J Surg.* 1996; 20:337-40.
- Saimot AG. Medical treatment of liver hydatidosis. *World J Surg.* 2001; 25:15-20.

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Хируршко одељење, Клиничко-болнички центар „Бежанијска коса“, Београд, Србија

КРАТАК САДРЖАЈ

Увод Ехинококна болест је зооноза изазвана псећом пантљичаром (*Echinococcus granulosus*). Ехинококус се најчешће јавља у јетри и плућима, а ретко у другим органима, поготово у попречнопругастим мишићима.

Приказ болесника Мушкарац стар 36 година примљен је због болова и слабости у десној натколеници и губитка телесне тежине. Тегобе су почеле годину дана пре пријема. Након испитивања компјутеризованом томографијом и магнетном резонанцијом установљена је велика цистична туморска промена у десном бедреном мишићу (*m. psoas dex*). Серолошки тестови на ехинококус били су негативни, а неуролошким испитивањем је утврђен губитак функције десног феморалног нерва. Постављена је индикација за хируршко лечење и урађена то-

тална ексцизија цистичне промене. Током операције је уочена инфилтрација десног феморалног нерва, који је сачуван. Макроскопски и патохистолошки потврђена је ехинококна природа цистичне промене. Након хируршког лечења примењена је физикална терапија и три године после операције болесник нема знаке болести и неуролошки дефицит.

Закључак Ехинококна болест бедреног мишића је веома ретко стање. Свака цистична туморска промена локализована у ретроперитонеуму, поред ретроперитонеумских тумора, туберкулозе и цистичних тумора друге природе, диференцијалнодијагностички мора узети у обзир и ехинококус. Радикалан хируршки захват је метода избора, с тим да се посебна пажња мора усмерити на околне захваћене анатомске структуре.

Кључне речи: ехинококус; феморални нерв; пареза

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