Case Report

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20240234

Primary hydatid cyst of erector spinae muscle of upper back

Mir Fazil Illahi¹, Mubashir Gani²*, Ajaz Ahmad Malik¹

¹Department of General and Minimal Invasive Surgery, Sher-I- Kashmir Institute of Medical Sciences, Soura, Srinagar, Jammu and Kashmir, India

²Department of General Surgery, SKIMS Medical College Bemina, Srinagar, Jammu and Kashmir, India

Received: 24 December 2023 Accepted: 12 January 2024

*Correspondence:

Dr. Mubashir Gani, E-mail: mubashir.gani72@gmail.com

Copyright: [©] the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Hydatid disease is a zoonosis caused by an infection with the larvae of the tapeworm *Echinococcus granulosus*, it mostly involves liver and lungs but it may exceptionally affect muscle. We report the case of a 29 years old man who was admitted to our surgical clinic with a primary hydatid cyst in erector spinae muscle of upper back diagnosed by clinic examination, imaging and serological testing. The treatment was a complete pericystectomy and chemotherapy by antiparasitic drugs. Hydatid disease is an endemic disease in sheep-producing regions. In human, this disease involves usually the liver and the lungs and exceptionally the muscle. The diagnosis may be challenging, as it should consider arguments such as history, physical examination, imaging and serological testing. The best treatment is pericystectomy with perioperative chemotherapy to reduce risk of occurrence. The hydatid cyst of erector spinae muscle is exceptional even in endemic area, the diagnosis may be challenging and the surgery is the gold standard of treatment.

Keywords: Hydatid cyst, Erector spinae muscle, Pericystectomy

INTRODUCTION

Hydatid disease or Echinococcosis is a zoonosis that occurs in sheep-grazing countries of the world but is common worldwide because dog is a definitive host. Echinococcosis is endemic in Mediterranean Countries, Middle East, Asia, South America, Australia, New Zealand and East Africa. Liver is the most commonly affected organ, followed by lung. The parasite can colonize virtually any organ of the body. The current case report demonstrates primary hydatid disease of erector spinae muscle with a successful surgical cure.

CASE REPORT

A 29-year-old male with no underlying comorbidities presented to our surgical clinic at Sher-e-Kashmir Institute of Medical Sciences, with the complaints of pain lower neck and left upper back for 6 months and swelling same area for 2 months. Pain was gradual in onset, progressive in nature, initially mild to moderate then became severe in intensity, non-radiating, no referral. There was no history of trauma to the area. Patient received 16 sessions of physiotherapy and analgesia for the symptoms. On examination there was a soft, cystic, non-tender swelling with regular appearing margins approximately $5 \times 6 \times 6$ cm in size, present on upper back at medial border of left scapula. There was no sensory or motor deficit noted. Patient was subjected to ultrasonography which suggested a well-defined multiseptated cystic area along the superior aspect of left back in paramedian position 70×40 mm in size. Serum for hydatid serology Echinococcus IgG tested positive. Contrast enhanced computed tomography (CECT) chest/abdomen/pelvis was done which was suggestive of thick walled multiseptated cystic lesion noted in left paraspinal muscle planes measuring 38×94×54 mm overlying the transverse process of C7 to lower border of D4 with no foramina extension or pleural extension seen (Figure 1). It did not show cyst at any other location in the body. Patient was admitted in department of general and minimal invasive surgery SKIMS SOURA with the diagnosis of hydatid cyst of left paraspinal muscle

of upper back based on clinical and radiological findings and underwent surgery. Intraoperative cyst was approx. 6×7 cm in size. There were multiple adhesions between deep fascia of muscle and cyst cavity (Figure 2a and b). Outcomes were uneventful and patient was discharged 3 days after surgery. Histological examination confirmed the diagnosis of hydatid cyst.



Figure 1: CECT of chest showing the location of hydatid cyst in relation to spine and chest wall.



Figure 2: (a) and (b) Intraoperative picture of intramuscular hydatid.

DISCUSSION

Hydatid disease is caused by *Echinococcus granulosus*, commonly known as dog tapeworm. The disease is globally distributed, though common in tropics, it is much less common in other countries, e.g., in UK. The dog is a definitive host while the intermediate hosts are humans, sheep and cattle. In humans, liver is the most common site of cyst development, followed by lungs, and much less commonly kidneys, spleen, brain and soft tissue.¹ Muscular involvement is usually seen as secondary location of liver or lung's hydatid cyst. However, it may be rarely seen as primary location even in endemic

countries (2-3%).¹⁻³ This atypical location may affect musculature of the chest wall, pectoralis major, sartorius, quadriceps, gluteus muscle and exceptionally paraspinal muscles.⁴⁻⁸ Musculo-skeletal system is reported to be the fifth common site of cystic echinococcosis (CE) after lung, liver, central nervous system (CNS) and orbit. Paraspinal involvement is even more uncommon.9-11 The exceptional muscular involvement can be explained by high lactic acid level in muscle tissue, unfavorable for parasite survival and the muscular contraction that prevent fixation of larvae to the tissue.¹² The proposed theory for translocation of parasite to paraspinal muscle is through the portal system to IVC from where the parasite gets access to lumbar plexus during Valsalva maneuver occurring during daily activities.^{13,14} Clinically, the muscular hydatid cyst may manifest as asymptomatic slow-growing mass with normal overlying skin to painful progressive mass growing between muscle planes causing stretch on the inter muscular septa. These nonspecific symptoms can simulate lipoma, hematoma, abscess or tumor or even the spine disorders which make the diagnosis of hydatid cyst in muscle very difficult, especially in patient without history of liver or lung location. The diagnosis of hydatid cyst in muscle may be challenging, as it should consider arguments such as history, physical examination, imaging and serological testing.^{15,16} Different serologic tests such anti-echinococcal as ELISA for antibody, hemagglutination test, immunoelectrophoresis (IEP) have been used with variable success.¹⁷

Although serologic tests are reported to have an acceptable sensitivity for intra-abdominal disease, they suffer low sensitivity for other involved sites.¹⁸ ELISA has been reported to have a sensitivity and specificity rate of 95% and 94% respectively.¹⁹ Serologic tests are reported to be falsely negative in half of isolated intra- muscular hydatidosis because of capsulate nature of the disease.^{9,15,16}

In our case who presented with swelling at upper back ultrasound and CECT chest/abdomen and pelvis were helpful supplements to physical examination in narrowing the differential diagnosis and the final diagnosis of hydatid cyst in paraspinal muscles was confirmed after successful and complete excision of the lesion by histopathologic assessment. Surgical removal of the cyst and chemotherapy by antiparasitic drugs are the treatment of choice for muscle hydatidosis.^{20,21} The best technique is the pericystectomy without opening the cyst and protection of surrounding tissue by hypertonic saline to reduce the risk of occurrence.²²

CONCLUSION

The hydatid cyst of erector spinae muscle is exceptional even in endemic area, it must be considered in front of any cystic mass especially when associated with other location. Diagnosis is very difficult in isolated forms. Pericystectomy with perioperative chemotherapy is the best treatment to reduce risk of recurrence. *Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required*

REFERENCES

- 1. Lorenzetti L. Contribution to the knowledge of primary echinococcosis of the muscles. Gazz Int Med e Chir. 1962;67:2775-95.
- 2. Sakka SA. Primary hydatic cyst of the thigh has painful manifestation at the hip about a case and review of the literature. Rev Chir Orthop Reparatrice Appar Word. 1993;79:226-8.
- Alouini Mekki R, Mhiri Souei M, Allani M, Bahri M, Arifa N, Jemni Gharbi H, et al. Hydatid cyst of soft tissues: MR imaging findings (Report of three cases). J Radiol. 2005;86(4):421-5.
- 4. Alvarez-Sala R, Terreros FJGD, Caballero P. Echinococcus cyst as a cause of chest wall tumor. Ann Thoracic Surg. 1987;43:689-90.
- 5. Abdel-Khaliq RA, Othman Y. Hydatid cyst of pectoralis major muscle. Case report and note on surgical management of muscle echinococcosis. Acta Chir Scand. 1986;152:469-71.
- 6. Rask MR, Lattig GJ. Primary intramuscular hydatidosis of the sartorius. Report of a case. J Bone Joint Surg. 1970;52:582-4.
- 7. Ozkoc G, Akpinar S, Hersekli MA, Ozalay M, Tandogan R. Primary hydatid disease of the quadriceps muscle: a rare localization. Arch Orthop Trauma Surg. 2003;123:314-6.
- Khorshidi H, Taghipour M, Moslemi M, Farsangi LE. Hydatid Cyst of Para-Spinal Muscles as a Rare Cause of Back Pain in a Woman: A Case Report and Review of Literature. Case Rep Clin Med. 2020;9:184-90.
- 9. Geramizadeh B. Unusual Locations of the Hydatid Cyst: A Review from Iran. Iran J Med Sci. 2013;38:2-14.
- 10. Atalay F, Orug T, Arda K, Tosun O, Atalay F. An unusual case of hydatid disease located in the erector spinae muscle. JBR-BTR. 2003;86(6):329-31.
- 11. Sadeghian H, Motiei-Langroudi R. Hydatid Cyst in the Lumbar Para- vertebral Muscle: A Case Report. Cureus. 2019;11:e5353.
- 12. Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary subcutaneous cyst hydatic disease in

proximal thing: an unusual localisation: a case report. BMC Musculoskelet Disord. 2003;4:25.

- Sener RN, Calli C, Kitis O, Yalman O. Multiple, primary spinal-paraspinal hydatid cysts. Eur Radiol. 2001;11(11):2314-6.
- Schnepper GD, Johnson WD. Recurrent Spinal Hydatidosis in North America. Neurosurgical Focus. 2004;17:1.
- 15. Bilgic S, Kose O, Sehirlioglu A, Safaz I, Ozkan H. Primary paraspinal hydatid cyst treated with puncture, aspiration, injection and re-aspiration (PAIR) technique: a case report. Eur Spine J. 2009;18(2):165-7.
- 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.
- 17. García-Díez AI, Ros Mendoza LH, Villacampa VM, Cózar M, Fuertes MI. MRI evaluation of soft tissue hydatid disease. Eur Radiol. 2000;10(3):462-6.
- Engin G, Acunas B, Rozanes I, Acunas G. Hydatid Disease with Un- usual Localization. Eur Radiol. 2000;10:1904-12.
- Shambesh MA, Craig PS, Macpherson CN, Rogan MT, Gusbi AM, Echtuish EF. An extensive ultrasound and serologic study to investigate the prevalence of human cystic echinococcosis in northern Libya. Am J Trop Med Hyg. 1999;60(3):462-8.
- 20. Ormeci N, Idilman R, Akyar S, Palabiyikoğlu M, Coban S, Erdem H, et al. Hydatid cysts in muscle: a modified percutaneous treatment approach. Int J Infect Dis. 2007;11:204-8.
- Ferrandez HD, Gomez-Castresana F, Lopez-Duran L, Mata P, Brandau D, Sanchez-Barba A. Osseous hydatidosis. The Journal of bone and joint surgery. American volume. 1978; 60: 685-690.
- Kazakos CJ, Galanis VG, Verettas DAJ, Polychronidis A, Simopoulos C. Primary Hydatid Disease in Femoral Muscles. J Int Med Res. 2005;33:703-6.

Cite this article as: Illahi MF, Gani M, Malik AA. Primary hydatid cyst of erector spinae muscle of upper back. Int J Res Med Sci 2024;12:594-6.