Case Report

A Rare Primary intramuscular hydatid cyst misdiagnosed as malignant spindle cell tumor: A Case Report

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<u>Abstract</u>

Primary hydatid disease of musculoskeletal system is rare. A 46 year old man presented with soft swelling in anterior aspect of thigh of two years duration which was gradually increasing in size. He was initially diagnosed as malignant spindle cell tumor of thigh, but ultrasonography and Commuted Tomography Scan revealed to be a cystic swelling suggestive of hydatid disease. Our patient had not been operated for hydatid disease previously and investigations did not reveal any hydatid cyst in liver, lung or spleen. So our patient was diagnosed having primary hydatid disease of musculoskeletal system. Serologic test (ELISA) was negative. Patient was given albendazole preoperatively. A careful management is required to prevent systemic dissemination and anaphylactic shock. The swelling was removed en bloc without causing damage to cyst wall and advised for adjunctive albendazole chemotherapy (15 mg/kg/day) for three months.

Keywords: Hydatid cyst; Primary; Thigh; Spindle cell tumor

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Introduction

Hydatid cysts predominantly involve the liver and lung with muscle tissue being the less preferred site for primary hydatidosis. Primary intramuscular hydatid cyst presents a diagnostic problem not only because of the unusual location and low prevalence, but also because complicated cysts may imitate solid or complex lesions.¹ The differential diagnosis in these cases must include malignant soft-tissue tumors such as myxoidliposarcoma, soft tissue abscesses, and chronic hematoma.²

<u>Case report</u>

A 46 year old male presented with large soft tissue swelling in the anterior aspect of right thigh. The swelling was present from last 2 years and gradually increasing in size. There was no pain in the swelling. On local examination, swelling was approximately 20 cm \times 7 cm, non tender and soft. Initial diagnosis of malignant spindle cell tumor was made on FNAC out side our institute and patient was referred to our institute. There were no symptoms or signs of an inflammatory process. Routine laboratory tests

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were normal, except peripheral smear was showing eosinophilia. Plain radiographs showed only softtissue swelling with no bony destruction. CECT scan right thigh demonstrated a well-defined multiseptated intramuscular cystic lesion measuring 22 cm x 7 cm x 5 cm (Fig, 1). musculoskeletal system. Before we plan for surgical excision or biopsy or FNAC, diagnosis of hydatid disease should be excluded, so as to avoid leakage of cyst contents and accompanying risk of anaphylaxis and secondary hydatidosis. Serologic tests and ultrasonography should be performed before any



The differential diagnosis of soft-tissue tumor and hydatid cyst were offered by the radiologist. Plain chest X-rays and abdominal ultrasonography did not reveal any organ involvement. Serologic test, ELISA was negative. Cyst was removed en bloc, which was 21 cm \times 7 cm in dimensions (Fig 2). Cut section showed multiple daughter cysts. Cyst cavity was thoroughly irrigated with 10% betadine and wound closed after putting closed suction drain. Post operative period was uneventful. Histopathological diagnosis confirmed the diagnosis of hydatid disease.

Discussion

The hydatid disease is caused by *Echinococcus granulosus* and multilocularis. Musculo-skeletal hydatid disease may either be secondary or .Primary hydatid disease of the skeletal muscle is rare, as the parasite has to cross pulmonary and hepatic barriers to reach the muscles³. The high lactic acid level in muscle tissue is considered unfavorable for the survival of parasite⁴. In secondary disease, there is primary location of hydatid cyst in liver, lung or spleen that has been operated or not operated. This patient had not been operated for hydatid disease previously and investigations did not reveal any hydatid cyst in liver, lung or spleen. So, this patient was diagnosed having primary hydatid disease of

invasive procedure. ELISA is 80-100% sensitive and 88-96% specific for hydatid liver disease but less sensitive for lung (50-56%) or other organ involvement $(25-26\%)^5$. Hydatid serology is only valuable when it is positive, negative serology does not exclude the diagnosis⁶. Serologic test (ELISA) was negative in this case. Ultrasonography still remains the major non-invasive screening tool to discover the primary site of the disease and may confirm the diagnosis of hydatid disease by demonstrating the pathognomonic daughter cysts7. The CT appearance of the hydatid cyst is not diagnostic as it may mimic malignant

and benign conditions such as congenital cyst, pseudocyst, or hematomas. However, the presence of daughter cysts, germinal epithelium detachment, and calcification may confirm the diagnosis. Similarly, an MRI can reveal a cystic mass containing daughter cysts, with rim sign and "Water Lilly sign."8. Peripheral smear may reveal eosinophilia in only 50% of the patients, which was seen in our case. The best way to establish the diagnosis is the direct visualization of parasitic elements in the surgically resected pathological specimen. En bloc resection alone is curative for intramuscular hydatid disease. In this patient we were able to remove whole of the cyst in toto. Adjunctive chemotherapy was given to eliminate any possible larvae dissemination and to take care of possible hydatid disease at other site.

Diagnosis of echinococcosis should be considered when slowly growing soft tissue tumour is present and cautions should be taken not to be misdiagnosed as a malignant tumour.

Ethical approval: This case report was ethically approved prior the submission for publication. Conclusion

Conflict of interest: None declared

References

- Von Sinner W, te Strake L, Clark D, Sharif H. MR imaging in hydatid disease. *Am J Roentgenol* 1991; **157**: 741-5.
- 2. Said M, Babba H, Golli M, Ganouni A. Can We Really Have Coexistence of Hydatid Cyst and Tapeworm in Humans? *Am J Roentgenol* 2001; **176**: 252-3.
- Garcia-Alvarez F, Torcal J, Salinas JC, Navarro A, Garcia-Alvarez I, et al. Musculoskletalhydatid disease: a report of 13 cases. *ActaOrthop Scand*. 2002; 73: 227– 231. doi: 10.1080/000164702753671858.
- 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**: 93–94. doi: 10.1007/PL00013775.
- 5. Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary subcutaneous cyst hydatid disease in proximal

high: an unusual localisation; a case report. *BMC MusculoskeletDisord*. 2003;**4**:25. doi: 10.1186/1471-2474-4-25.

- Durakbasa CU, Tireli GA, Sehiralti V, Sander S, Tosyali AN, et al. An audit on pediatric hydatid disease of uncommon localization: incidence, diagnosis, surgical approach, and outcome. *J Pediatr Surg.* 2006; **41:** 1457– 1463. doi: 10.1016/j.jpedsurg.2006.04.024.
- Diez G, Mendoza LHR, Villacampa VM, Cozar M, Fuertes MI. MRI evaluation of soft tissue hyaditid disease. EurRadiol. 2000; 10: 462–466. doi: 10.1007/ s003300050077.
- Arazi M, Erikoglu M, Odev K, Memik R, Ozdemir M. Primary echinococcus infestation of the bone and muscle. *ClinOrthopRelat Res*. 2005; **432**: 234–241. doi: 10.1097/01.blo.0000149816.86222.2d.