



Hydatid cyst of biceps brachii associated with peripheral neuropathy



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ABSTRACT

INTRODUCTION: Hydatidosis represents the most significant parasitic disorder in the Mediterranean countries and leads to major problems through unfavorable effects on the public health and national economy. Localization of the primary cyst hydatid infection in the extremity is rare and biceps brachii localization is also rarely reported in the literature.

PRESENTATION OF CASE: A 43-year-old woman, who presented with the complaints of mass and pain in the left arm and numbness of the hand. Laboratory investigations, X-ray and magnetic resonance (MRI) findings revealed hydatid cyst of the biceps brachii muscle. The mass was totally excised and the diagnosis was confirmed by the macroscopic images of the mass and the pathologic results. After the surgery, the patient had an improvement in the nerve compression findings including numbness of the hand and the upper extremity and pain.

DISCUSSION: Localization of a primary cyst hydatid infection in the upper extremity is rare and there are no reports of peripheral neuropathy secondary to mass effect. Even if the pre-surgical electromyography performed for the nerve conduction study reveals a normal result, the potential for the hydatid cysts to cause nerve compression should be taken into consideration in such patients.

CONCLUSION: Cases of concomitant neurologic findings and complaints secondary to peripheral nerve compression are very rare. The clinical findings should not be ruled out even if the EMG result is negative.

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1. Introduction

Hydatidosis is a parasitic and zoonotic disease of the humans and animals. It represents the most significant parasitic disorder in the Mediterranean countries and leads to major problems through unfavorable effects on the public health and national economy [1]. Located in the Mediterranean region, Turkey is among the endemic countries [2].

Hydatid cyst most commonly occurs in the young population and the herds of sheep-goats raised by the countries and the relation between the humans and dogs are effective on the increased prevalence [3]. Particularly, the eggs swallowed as a result of the human-animal contact under rural conditions crack in the intestinal system and reach the liver via portal venous route. The liver (60%) and the lungs (30%) are the most commonly affected organs with the brain, the heart, the kidneys, the ureter, the spleen,

the uterus, the pancreas, the diaphragm and the extremity muscles being rarely involved [4]. Localization of the primary cyst hydatid infection in the extremity is rare and biceps brachii localization is also rarely reported in the literature [5,6]. In this report, a 43-year-old woman, who presented with the complaints of mass and pain in the left arm, and numbness of the hand, and diagnosed as cyst hydatid of the biceps brachii muscle, was presented.

2. Presentation of case

A 43-year-old woman presented with the complaints of mass and pain in the left arm, and numbness of the hand. The mass with an indolent course for 7 years had started exhibiting progressive enlargement and pain, resulting in a limitation in daily activities. The physical examination of the patient, who had a history of living in the village and sheep raising, revealed a soft, mobile fluctuating mass of 150 * 100 mm size that extended toward the medial site from the upper-middle part of the left arm. No findings of an increase in heat or redness on the mass were detected. The laboratory findings were as follows: leukocytes, CRP, sedimentation, eosinophilia and hemoglobin are normal. The cyst hydatid indirect hemagglutination test was strongly positive. X-ray showed soft tissue swelling. Magnetic resonance (MR) images of the left

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Fig. 1. (A) Pre-operative X-ray, (B) coronal, (C) sagittal MR, and (D) Post-operative X-ray.

arm revealed a cystic lobulated mass of 150 * 90 * 55 mm size containing septa and minimal soft tissue density and fluid levels, which showed peripheral contrast uptake following IVCM and was located under the skin in anterior adjacency to the biceps muscle, extending from the 1/3 upper–middle section anterior toward the medial section (Fig. 1). In addition, axial and coronal sections showed close adjacency of the mass to the neurovascular bundle (Fig. 2). Whether there were any concomitant hydatid cysts in the other organs was investigated via whole abdominal, thoracic and cerebral computed tomography and no pathological findings were detected. The patient who had numbness in the left hand underwent electromyography (EMG) and was considered to be normal.

The patient was operated in supine position under general anesthesia. Longitudinal incision was performed from the left arm anterior section and over the mass. We reached the mass proceeding beyond the subcutaneous section. During dissection, we observed that the mass was on the neurovascular bundle and caused compression, but it was not attached to it. The cyst was carefully scraped from the adjacent structures and totally excised without causing any rupture. After excision, the median nerve was seen to be relieved of the pressure it had been exposed to with the effect of the mass. Following irrigation of the cystic cavity with hypertonic sodium chloride solution, the cyst wall was excised. A large number of viscous, gelatinous-appearing cysts with an irregular ragged inner surface were observed

macroscopically inside the mass transferred into the investigation container (Fig. 3). The histopathological examination of the cyst wall revealed a characteristic eosinophilic ectocyst with a chitin membrane. No intraoperative or postoperative complications were seen. As postoperative chemotherapy, high dose albendazole was initiated for 6 weeks, and a favorable response was obtained. The patient had complaints of numbness in the early period, which spontaneously recovered and no recurrence was observed on early or long-term radiographs or USG examinations.

The patient was informed that the data concerning his case would be submitted for publication.

3. Discussion

Cyst hydatid is commonly observed in the Mediterranean, Baltic states, South America, Australia, and the Middle East, primarily in sheep-raising countries [7]. The liver and the lungs being the most commonly involved organs, this condition can occur in all organs and tissues [8]. The skeletal system is rarely involved [9,10]. Because the echinococcus entering the body via intestinal route would pass into the venous circulation system and respectively pass the liver and the lungs, which have a filtering, function.

Primary musculoskeletal cyst hydatid generally affects the proximal muscle groups and localizes more commonly in the lower extremity compared to the upper extremity [11,12]. Biceps brachii

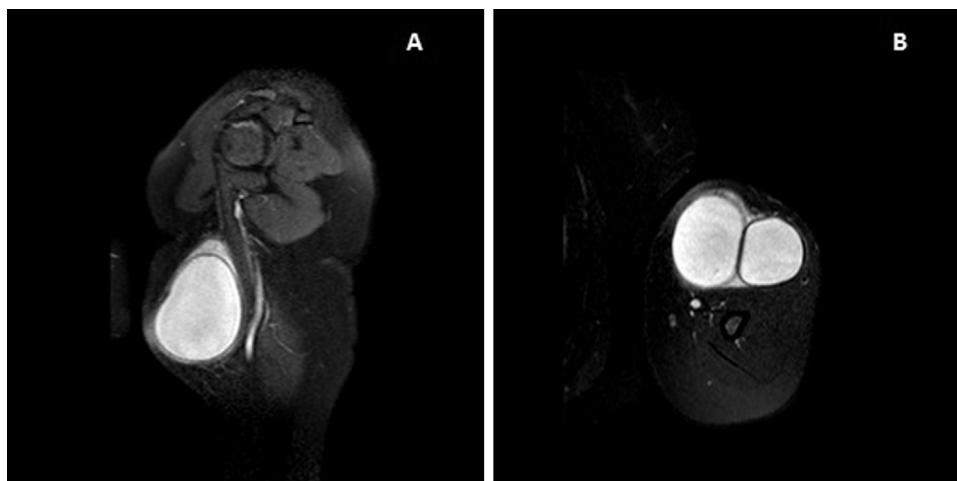


Fig. 2. The MR sections showing the association between the mass and the neurovascular bundle. (A) sagittal, (B) axial.

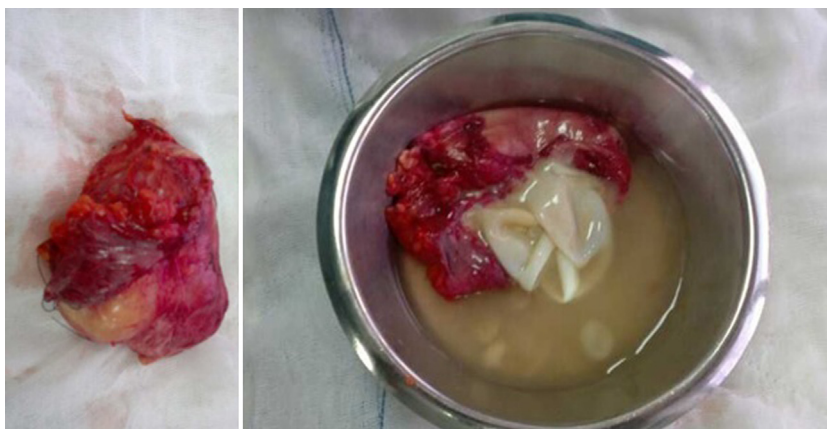


Fig. 3. Intraoperative macroscopic assessment; a large number of viscous, gelatinous-appearing cysts with an irregular ragged inner surface inside the mass.

localization has been rarely reported in the literature [5,6]. The patient in our case report presented with the complaints of mass and pain in the left arm, and numbness of the hand, and was diagnosed with cyst hydatid in the biceps brachii. In such cases, peripheral nerve involvement is very rare and there are no reports of peripheral neuropathy secondary to the impact of cyst hydatid mass located in the upper extremity. In our patient, the neuropathic findings were limited to the complaints in the median nerve sensory conduction sites without concomitant motor deficit. Several authors have reported occurrence of motor deficit, sciatic pain and foot drop as well as sensory neuropathies as a result of the effect of compression from the lower extremity-localization hydatid cysts on the sciatic nerve or lumbar plexus [13,14]. Alimehmeti et al. reported that a large-diameter hydatid cyst located in the adductor muscle only resulted in hypoesthesia in the lower extremity saphenous nerve distribution without concomitant motor deficit. In addition, they showed the effect of compression from the mass on the neurovascular bundle on MR sections [15]. In our case, we detected a compression of the cystic mass on the neurovascular bundle and their adjacency via MRI. EMG was performed to evaluate the nerve conduction functions. However, while the preoperative EMG study was considered normal, the potential for positive sensorial clinical findings should also be considered. Sensorial potentials on EMG may sometimes show as normal even in abnormal clinical situations. This is because if there is no impairment in the axonal structure of the peripheral nerves, the axonal signal from the cell body is carried to the peripheral axons without impairment [16]. Our patient was observed to have improvement in pain and numbness on short-term follow-up following excision.

The laboratory findings may be supportive for diagnosis. In the presence of liver and lung diseases, serologic tests are commonly negative [11]. However, serologic tests including indirect immunofluorescent antibody test, ELISA, immunoelectrophoresis and immunoblot are commonly used [17]. Our patient was detected to have strongly positive indirect hemagglutination antibody test.

Due to the fact that it leads to a mass formation, hydatid cysts may be mistaken for soft tissue tumors [18]. This should be considered in differential diagnosis. Due to the high risk of spreading of infection and the potential for resulting in an anaphylactic shock, needle biopsy should be avoided [11]. In general, radiologic imaging is very valuable in the diagnosis and differential diagnosis of cyst hydatidosis. Magnetic resonance, ultrasonography (USG) or computed tomography may be performed. USG may yield beneficial data on the dimensions, localization of the mass and the type of the cyst with a 95% sensitivity. BT may be performed to determine the structure of the adjacent tissues and

the surgical technique [19]. However, MR is the golden standard of diagnosis and detection of multilocular polycystic lesions on the cross-sections and the presence of a two-layer membrane of the lesion, which consists of a pericyst vascularized by collagen tissue is also supportive in differential diagnosis [20].

The surgery site should be rinsed with hypertonic sodium chloride and the cyst removed completely without any punctures. In case the cyst content spreads during surgery, anaphylaxis or secondary echinococcus may occur [21]. In this case, the cyst was surgically excised without any complications. Consulting with the Infectious Diseases Clinic, antihelminthic chemotherapy was administered for preoperatif 3 weeks and postoperatif 2 months (albendazol 800 mg/d). While there are literature reports of rare extremity involvements such as supraspinatus, gracilis, adductor magnus, sartorius and biceps brachii, cases of concomitant neurologic findings and complaints secondary to peripheral nerve compression are very rare [22–25].

4. Conclusion

Cases of concomitant neurologic findings and complaints secondary to peripheral nerve compression are very rare. The main treatment for musculoskeletal cyst hydatidosis is surgical excision and the clinical findings should not be ruled out even if the EMG result is negative.

Conflict of interest

No conflict of interest was declared by the authors.

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Author contribution

ST, TD, SM, and HY were involved in the conception, design, and interpretation.

ST, TD, and SM wrote the manuscript. ST, MD, and SE collected data, reviewed relevant published reports and provided the images.

All authors read and approved the final manuscript.

Consent

Written informed consent was obtained from patient who participated in this case.

References

- [1] A. Merdivenci, Hydatid cyst disease in Turkey; Ist, 93, Faculty of Medicine Publishing, Istanbul, 1976.
- [2] R. Dogan, M. Yuksel, G. Cetin, K. Suzer, M. Alp, S. Kaya, M. Unlu, B. Moldibi, Surgical treatment of hydatid cysts of the lung: report on 1055 patients, *Thorax* 44 (3) (1989) 192–199.
- [3] A.G.T.W. Finnes, D.G.T. Thomas, Combined medical and surgical treatment of spinal hydatid disease. A case report, *J. Neurol. Neurosurg. Psychiatry* 45 (10) (1982) 927–930.
- [4] C. White Jr, P.F. Weller Cestodes, in: S. Anthony Fauci (Ed.), *Harrison's Principles of Internal Medicine*, Seventeenth ed., McGraw-Hill, New York, 2008, p. 1339.
- [5] M. Ates, M. Karakaplan, Hydatid cyst in the biceps and gluteus muscles: case report, *Surg. Infect. (Larchmt)* 8 (4) (2007) 475–478.
- [6] G.J. Duncan, S.M. Tooke, Echinococcus infestation of the biceps brachii. A case report, *Clin. Orthop. Relat. Res.* 261 (1990) 247–250.
- [7] J.E. Doty, R.K. Tompkins, Management of cystic disease of the liver, *Surg. Clin. North Am.* 69 (1989) 285–295.
- [8] P. Moro, P.M. Schantz, Echinococcosis a review, *Int. J. Infect. Dis.* 13 (2009) 125–133.
- [9] A.A. Uraiqat, A. Al-Awamleh, Case report: hydatid cyst in the muscles, *JRMS* 17 (Supplement 1) (2010) 72–74.
- [10] M. Mseddi, M. Mtaoumi, J. Dahmene, R. Ben Hamida, A. Siala, T. Moula, M.L. Ben Ayeche, Hydatid cysts in muscles: eleven cases, *Rev. Chir. Orthop. Reparatrice Appar. Mot.* 91 (3) (2005) 267–271.
- [11] H.F. Loyarte, J.L. Curutchet, Quistes hydatidos musculares: contribution à la casuística nacional, *Pren. Med. Argent.* 48 (1961) 259–269.
- [12] J. Eckert, M.A. Gemmell, F.X. Meslin, Z.S. Pawłowski, Echinococcosis in Humans: Clinical Aspects, Diagnosis and Treatment. In WHO/OIE Manual on Echinococcosis in Humans and Animals: A Public Health Problem of Global Concern, World Organisation for Animal Health 12, rue de Prony, 75017, Paris, France, 2001, pp. 20–25, Chapter 2 <http://www.oie.int/ISBN92-9044-522-X>
- [13] C.J. Kazakos, V.G. Galanis, D.A. Verettas, A. Polychronidis, C. Simopoulos, Primary hydatid disease in femoral muscles, *J. Int. Med. Res.* 33 (6) (2005) 703–706.
- [14] A. Kalaci, T.T. Sevinç, A.N. Yanat, Sciatica of nondisc origin: hydatid cyst of the sciatic nerve. Case report, *J. Neurosurg. Spine* 8 (4) (2008) 394–397.
- [15] R. Alimehmeti, A. Seferi, A. Rroji, M. Alimehmeti, Saphenous neuropathy due to large hydatid cyst within long adductor muscle: case report and literature review, *J. Infect. Dev. Ctries.* 6 (6) (2012) 531–535.
- [16] M.J. Aminoff, in: *Aminoff's electrodiagnosis in clinical neurology*, 6th ed., Saunders, Philadelphia, 2012.
- [17] W. Zhang, J. Li, D.P. McManus, Concepts in immunology and diagnosis of hydatid disease, *Clin. Microbiol. Rev.* 1 (2003) 18–36.
- [18] A. Gupta, A. Kakkar, M. Chadha, C.B. Sathaye, A primary intrapelvic hydatid cyst presenting with foot drop and a gluteal swelling: a case report, *J. Bone Joint Surg. Br.* 80 (6) (1998) 1037–1039.
- [19] T. Fikry, A. Harfaoui, H. Sibai, B.L. Zryouil, Echinococcosse musculaire primitive, *J. Chir.* 7-8 (1997) 325–328.
- [20] A. Memis, R. Arkun, I. Bilgen, E.E. Ustun, Primary soft tissue hydatid disease: report of two cases with MRI characteristics, *Eur. Radiol.* 9 (1999) 1101–1103.
- [21] M.S. Khuroo, Hydatid disease: current status and recent advances, *Ann. Saudi Med.* 22 (2002) 56–64.
- [22] H. Tatari, O. Baran, T. Sanlidag, O. Gore, D. Ak, M. Manisali, H. Havitioglu, Primary intramuscular hydatidosis of supraspinatus muscle, *Arch. Orthop. Trauma. Surg.* 121 (2001) 93–94.
- [23] M.O. Durakbasa, O. Kose, N.C. Islam, G. Kilicoglu, A primary hydatid cyst of the gracilis: a case report, *J. Orthop. Surg. (Hong Kong)* 1 (2007) 118–120.
- [24] G. Ozkoç, S. Akpınar, M.A. Hersekli, M. Ozalay, R. Tandoğan, Primary hydatid disease of the quadriceps muscle: a rare localization, *Arch. Orthop. Trauma Surg.* 123 (2003) 314–364.
- [25] F. Duygulu, S. Karaoğlu, N. Erdoğan, O. Yildiz, Primary hydatid cyst of the thigh: a case report of an unusual localization, *Turk. J. Pediatr.* 48 (2006) 256–259.

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