Pelvic echinococcosis presenting as a mass in the gluteal region: a case report

Sumit Banerjee, Akshat Gupta, Ramkaran Choudhary, Khushwant Singh Rathore, Sudeep Khera

All India Institute of Medical Sciences, Jodhpur, India

SUMMARY

Hydatid disease of the skeletal system is a rare entity. We present one such case of pelvic echinococcosis in a 62-year-old male who presented with the chief complaints of pain and swelling in the left gluteal region for the past twelve years. The patient was planned for and underwent en-bloc excision of the lesion. Albendazole was used pre- and post-operatively to reduce parasitic load. At the last follow-up, the patient was disease-free and able to carry out his daily activities without much difficulty. We would conclude that, although uncommon, echinococcosis should always be ruled out, especially when dealing with other indolent pathologies of the hip joint.

Keywords: hydatid cyst, pelvis, echinococcus, albendazole.

INTRODUCTION

Hydatid disease is a parasitic zoonosis caused by various species of tapeworms belonging to the genus *Echinococcus* [1-4]. The most common amongst them is *Echinococcus granulosus*, also known as "dog tapeworm" [1-7]. The latter manifests primarily in dogs and other carnivores as definitive hosts with humans, sheep and cattle as intermediate hosts [1-7]. Most common site for manifestation of the disease is the liver (55-75%) followed by lungs (15-35%) [1, 2, 4-10]. Skeletal involvement is rare and is seen in only 0.5-4% cases [1, 2, 4-10]. In addition, bony lesions of hydatidosis are extremely difficult to treat. Even with the best of efforts they are associated with poor functional outcomes in the long run [2, 4, 6, 9. Here, we are presenting a case of hydatid disease of the pelvis. The latter presented as a painful swelling in the gluteal region and was managed by a combination of chemotherapy and surgery.

Corresponding author
Akshat Gupta
E-mail: mirage1.akshat@gmail.com

CASE REPORT

The patient was a 62-year-old farmer who presented to the departmental outpatient clinic with the chief complaints of pain and swelling over the left gluteal region for the past 12 years. He had had multiple episodes of fluid aspiration from the swelling and had also been operated twice at a local hospital (where incision & drainage were done). However, in each case the symptomatic relief was only temporary and the swelling soon recurred. Examination revealed a large, cystic swelling over the left gluteal region which was painful and associated with reduced hip range of motion.

Plain radiographs of the pelvis showed cystic lesions with secondary destruction involving the left ischial tuberosity, acetabulum and femoral head (Figure 1a). Hip joint space was also reduced. Computerized tomography (CT) scan (Figures 1b and 1c) and magnetic resonance imaging (MRI) (Figure 2a-2e) of the pelvis revealed a multiloculated, T_2 hyperintense cystic collection in the left gluteal region (involving the gluteus maximus, medius and the obturator externus muscles). These cysts also showed T_2 hypointense

septations with tiny foci of hypointense calcifications along the periphery. Chest and abdominal-pelvic imaging did not reveal any collection or mass in the liver, lungs or any other viscera.

Laboratory investigations revealed an elevated erythrocyte sedimentation rate (31 mm/ hour by Westergen method), C-reactive protein (16.9 mg/L) as well as eosinophilia (7.5% with an absolute eosinophilic count of 730/ml). However, total leucocyte count was within normal limits and serial blood cultures failed to detect any pathological microorganism. Remaining blood investigations were within normal limits.

Based on these and the patient's clinic-radiological picture, a provisional diagnosis of low-grade infectious arthritis of the hip joint was made. Although pyogenic and tubercular infections were kept in our list of differentials, we decided to rule out parasitic infections also (given the elevated eosinophilic count). Towards this end, serological tests were carried out which demonstrated increased titres of IgG antibody against *Echinococcus granulosus*.

The patient was planned for en-bloc excision of the cyst (Figures 3a-3e). Preoperatively, he was given a course of albendazole (10mg/kg) for 3

Figure 1 - X ray and computerized tomography scan of the pelvis (A-C) showing multiple cystic lesions with secondary destruction of the left acetabulum, ischium and femoral head.

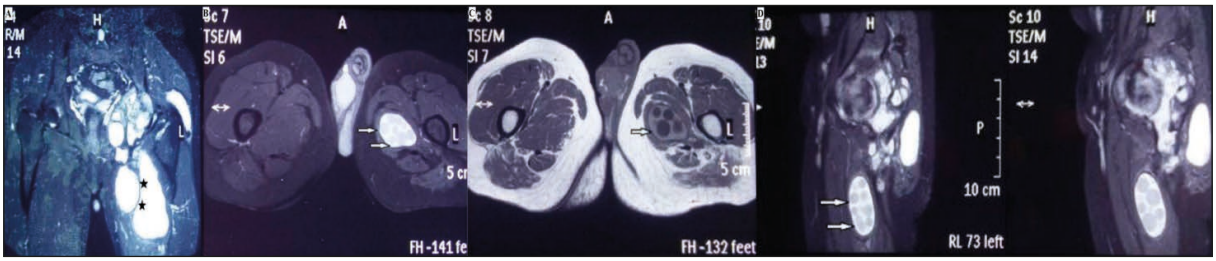

Figure 2 - Magnetic resonance imaging sequences (A-D): Multiloculated T2 hyperintense cystic lesions seen involving the left gluteus and obturator muscles. (NOTE: Hypointense septations [A- black stars] and peripheral daughter cysts [B/C/D- white arrows]).

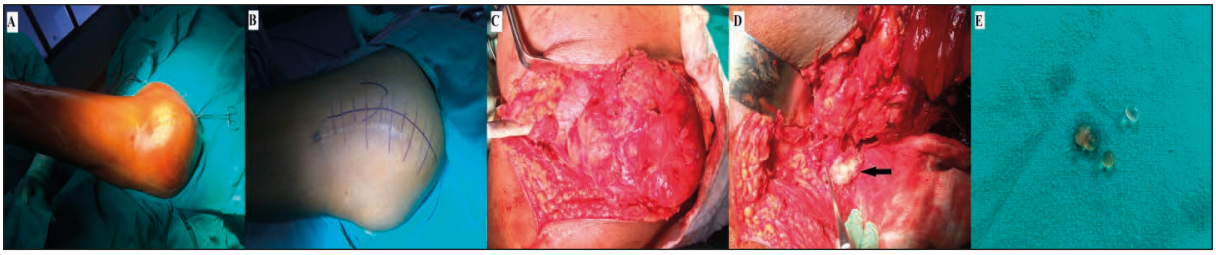

Figure 3 - Intra-operative images (A-D) (NOTE: "Mother" cyst [black arrow] and sciatic nerve [nontoothed-forceps]). (E) Daughter cysts.

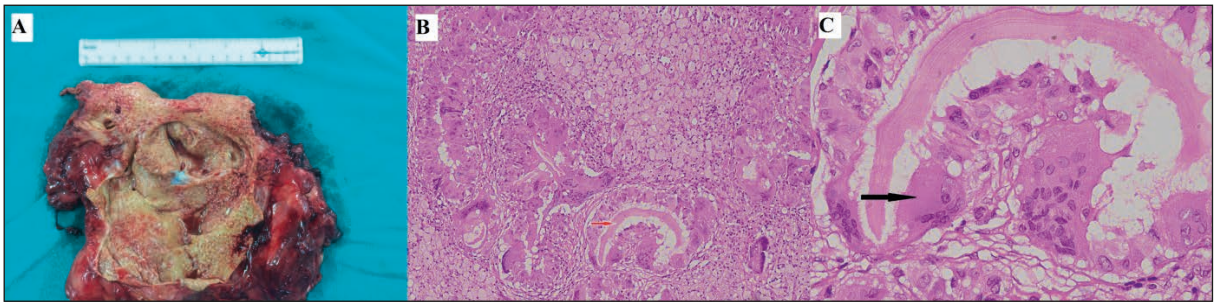

Figure 4 - (A) Gross appearance of the en-bloc resected specimen. (B) and (C) Microscopic examination reveals intense inflammatory response and giant cell reaction (black arrow) against lamellated membranes (red arrow)

weeks to reduce the parasitic load. He was operated in lateral decubitus position and the posterior approach to hip was planned for the excision. Following superficial dissection it was found that the mass infiltrated and occupied most of the gluteus maximus and medius muscles. Hypertonic saline was kept on standby for use as scolicidal agent in case of accidental rupture of cysts. The mass was resected en-bloc with the dissection carried proximally to the exterior iliac crest and distally beyond the insertion of gluteus maximus. The obturator externus muscle also had a cystic mass which was excised. The sciatic nerve was identified with blunt dissection and found to be free of any mass, however a mother cyst was found lying very close to the sciatic nerve and was dissected and excised. No prosthetic reconstruction of the hip joint was attempted to avoid the risk of surgical site infections. The wound was washed thoroughly and closed over a drain. Resected sample was sent for histopathological analysis, Gram and Ziehl-Neelsen staining as well as bacterial and tubercular culture studies. Histopathological findings were consistent with hydatid cyst such as lamellated cystic wall with sub-epithelial granulation tissue, lymphoid aggregates and several foamy histiocytes (Figures 4a-4c). There was no growth on the bacterial and tubercular culture media. Postoperative period was uneventful and patient was subsequently discharged. He was put on albendazole (10 mg/kg) for 3 months. At 6 months follow up, the patient was pain free and walking with the help of crutches. He had abductor weakness and slight lurch but was able to carry out his activities of daily living without much discomfort.

DISCUSSION

Echinococcus granulosus is endemic to the sheep and livestock rearing areas of Northern Africa, Middle East, India and Australia [1, 2, 4-6]. After contamination via faecal-oral route, it lodges primarily in the liver and from here spreads to other areas of the body such as lungs, kidney, spleen, brain and the skeletal system. Liver and lungs are most commonly involved (accounting for up to 90% cases of visceral hydatidosis) [1, 2, 4-10]. In contrast, skeletal involvement has been reported to be between 0.5-4% only [1-7].

Echinococcus granulosus infestation in the skeletal system is primarily via hematogenous seeding although direct spread from adjacently located bones/soft tissues can also be seen [1]. Usually, spongy vascularised bones like vertebral bodies, pelvic bones and epiphyseal/metaphyseal ends of long bones are commonly implicated [1, 11]. Zlitni et al., Papanikolaou, Tuzun et al. have reported the incidence of skeletal hydatidosis in the spine to be between 35-50% followed by pelvis (21%) and long bones (femur 16% and tibia 10%) respectively [1, 7, 11]. In the spine, the disease most commonly involves the dorsal region (50%) followed by lumbar (20%), sacral (20%) and cervical regions (10%) [12].

Unlike other viscera, there is no adventitia formation in skeletal hydatidosis (due to the rigid structure of the bone). This allows for microvesicular invasion of the daughter cysts into the bony tissue [1, 11]. As a result, complete eradication of the parasite is often unsuccessful despite extensive surgery. Zlitni et al. proposed three mechanisms to account for the bony destruction: a mechanical process whereby the expanding cyst compresses

the surrounding soft tissues; an ischemic process due to obstruction of the nutrient blood vessels; and a cellular process which is mediated by the proliferating osteoclasts around the compressed bony tissue [1-3, 11]. According to Markakis et al. average age of presentation is 52 years and that the disease is more commonly seen in men [13]. Usually it is secondary to some complication such as pathological fracture (in case of long bones) or radiculopathy (in case of vertebral involvement), that the disease gets detected. In case of pelvic involvement, the disease usually forms a cold abscess which communicates both inside and outside the pelvis (via the sciatic notch). The chief complaint of the patient is usually swelling in the gluteal region accompanied by reduction in the hip range of motion. Compressive neuropathies of the femoral & sciatic nerves and tumefaction - when a cold abscess without fistula becomes hot due to secondary bacterial infection - may also be seen [1, 4, 11].

The most common radiographic picture is osteolysis with grouping of lacunar lesions without clear-cut boundaries giving a "waffle" like appearance. Other features include absence of periosteal reaction and preservation of joint space for a long period of time. Although, ilium is the most commonly involved site in the pelvis, disease may also spread to involve the hip and the sacro-iliac articulations [1, 11].

Eosinophilia is a diagnostic marker of several parasitic infections, chiefly tissue helminthiases [14]. However, elevated eosinophilic counts may also be seen in a host of allergic conditions and also as an expression of various paraneoplastic syndromes [14]. Serological tests are usually needed to make a more accurate assessment of the disease. The Casoni intradermal test as well as the complement fixation test have both become quite obsolete and are rarely performed nowadays. The chief serological tests carried out include the enzyme linked immunosorbent assay (ELISA), indirect haemagglutination test (IHA) and the immunoblot test (IB) to detect specific antibodies as well as circulating antigens (15). While ELISA is more sensitive, the IB techniques are more specific [1, 15]. Therefore, many have suggested using a combination of both during the serological diagnosis of cystic hydatidosis [1]. However, despite recent advances, these tests are of limited value and therefore, final diagnosis should be made intraoperatively or on histopathological analysis only [2, 11, 16].

The chief differentials of the disease include tuberculosis of the hip/sacroiliac joint; chronic osteomyelitis of the ilium and tumours with pelvic localisation such as chondrosarcomas and osteosarcomas [1, 2, 4, 11].

Management of osseous hydatidosis is similar to a malignant neoplasm. It involves a combination of chemotherapeutic agents (albendazole being the drug of choice) and surgery to attain a satisfacto-

Table 1 - A summary of the incidence of pelvic hydatid disease.

Author	*Age/gender*	*Location*	*No of cases*	*Treatment*
Wirbel[10] [1995]	49/M	Lt ilium, acetabulum & proximal, femur	1	5 year mebendazole + partial pelvic resection & custom made THR
Masse[8] [2004]	25/M	Lt ischium with hip	1	Excision with curettage
Siwach[2] [2009]	51/F	Entire Lt hemipelvis with femur head/ neck & shaft pathological fracture	1	Hindquarter amputation Albendazole
El- Ibrahim[3] [2009]	35/F	Lt Ilium, acetabulum & proximal femur	1	Albendazole
Nath[9] [2009]	35/M	Lt Ilium, ischial tuberosity,	1	Wide excision
Krishnan[16] [2012]	38/M	Entire Rt hemipelvis, proximal femur with B/L hydroureteronephrosis	1	Albendazole for 6 months + cystopericystectomy -
Haghighi[6] [2015]	51/M	Lt Ilium	1	Mebendazole
Bhatnagar[4] [2017]	35/M	Lt Ilium, acetabulum & proximal femur	1	3 month albendazole + girdlestone arthroplasty + endopelvic resection
Wahid[5] [2017]	61/F	Rt Ilium	1	Excision

ry outcome. Since curettage is associated with increased risk of anaphylaxis and local recurrence, an en-bloc excision of the lesion along with wide resection of the involved portion of the bone is the surgical procedure of choice [1, 2, 11].

Various authors have used different techniques to minimise the risk of recurrence. Herrera-Martinez recommended excision of an additional 1-2 cm of healthy bone [17]. Bhatnagar et al. advocated irrigation with various scolicidal solutions such as 10% povidione iodine [4]. We used a combination of chemotherapy and surgery. Cyst was resected en-bloc and hypertonic saline was used as scolicidal agent. A summary of studies describing the occurrence of pelvic hydatid disease and the treatment strategy adopted is given in Table 1.

Prognosis of pelvic hydatidosis is not good. There are two main reasons for this. One, due to the extensive bony invasion by the micro-vesicles and lack of clear-cut demarcation between normal and pathological tissues, complete eradication of the parasite is almost impossible. Secondly any reconstruction with prosthesis would be inadvisable due to increased chances of secondary bacterial infection [1, 11].

CONCLUSION

Although rare, hydatid disease of the pelvis must always be kept in mind when dealing with other indolent pathologies of the hip such as tuberculosis. This is especially important in people who are from a rural background and come into regular contact with animals.

Conflicts of interest: none.

REFERENCES

[1] Papanikolaou A. Osseous hydatid disease. *Trans. R. Soc. Trop. Med. Hyg*. 102, 3, 233-238, 2008.
[2] Siwach R., Singh R., Kadian V.K., et al. Extensive hydatidosis of the femur and pelvis with pathological fracture: A case report. *Int. J. Infect. Dis*. 13, 6, e480-482, 2009.
[3] El Ibrahimi A., Ankouz A., Daoudi A., Elmrini A. Pelvic bone and hip joint hydatid disease revealing a retroperitoneal location. *Orthop. Rev (Pavia)*. 1, 1, e8, 2009.
[4] Bhatnagar N., Kishan H., Sura S., Lingaiah P., Jaikumar K. Pelvic hydatid disease: A case report and review of literature. *J. Orthop. Case. Rep*. 7, 4, 25-28, 2017.
[5] Wahid M.A., Ahmed S.Z., Gowher H.A., Butoo A.J., Ahmed S.A., Nabi M.G. Musculoskeletal hydatid masquerading as malignancy: A diagnostic dilemma. *Afr. J. Med. Health. Sci*. 16, 1, 75-77, 2017.
[6] Haghighi M.S., Bagherifard A., Qoreishy M., Mokhtari T. Primary iliac bone hydatid disease: A case report. *Joint Bone Science. Journal* 2, 2, 129-134, 2015.
[7] Tüzün M., Hekimoğlu B. CT findings in skeletal cystic echinococcosis. *Acta Radiol*. 43, 5, 533-538, 2002.
[8] Massè A., Parola P.G., Brach del Prever E.M., Gallinaro P. Hydatidosis of the pelvis: a case report with a 25-year follow-up. *Arch. Orthop. Trauma. Surg*. 124, 3, 203-205, 2004.
[9] Nath P., Bhattacharya S., Dutta V., Joshi G.R., Patel M. Primary iliac bone hydatid disease: An unusual presentation. *Med. J. Armed. Forces. India* 65, 2, 180-181, 2009.
[10] Wirbel R.J., Mues P.E., Mutschler W.E., Salomon-Looijen M. Hydatid disease of the pelvis and the femur. A case report. *Acta. Orthop. Scand*. 66, 5, 440-442, 1995.
[11] Zlitni M., Ezzaouia K., Lebib H., Karray M., Kooli M., Mestiri M. Hydatid cyst of bone: diagnosis and treatment. *World. J. Surg*. 25, 1, 75-82, 2001.
[12] Scarlata F., Giordano S., Saporito L., et al. Cystic hydatidosis: a rare case of spine localization. *Infez. Med*. 19, 1, 39-41, 2011.
[13] Markakis P., Markaki S., Prevedorou D., Bouropoulou V. Echinococcosis of bone: clinico-laboratory findings and differential diagnostic problems. *Arch. Anat. Cytol. Pathol*. 38, 3, 92-94, 1990.
[14] Scarlata F. Eosinophils and eosinophilia. *Infez. Med*. 23, 4, 307-312, 2015.
[15] Hijjawi N.S., Al-Radaideh A.M., Rababah E.M., Al-Qaoud K.M., Bani-Hani K.E. Cystic echinococcosis in Jordan: A review of causative species, previous studies, serological and radiological diagnosis. *Acta. Trop*. 179, 10-16, 2018.
[16] Krishnan A., Easwaran S., Jeswanth S., Surendran R. An unusual presentation of primary pelvic hydatid cyst. *Hellen. J. Surg*. 84, 6, 378-380, 2012.
[17] Herrera A., Martinez A.A. Extraspinal bony hydatidosis. *J. Bone. Joint. Surg. Am*. 85, 9, 1790-1794, 2003.