

Hydatid Disease of Tibia Reconstructed with Allograft: A Rare Case Report and Literature Review

Abstract

Background: Hydatid disease of bone shows a well-defined, multiloculated lytic lesion with the appearance of a bunch of grapes. The presenting symptoms are pain and swelling with or without pathological fracture. The treatment options include surgery followed by a long duration of albendazole. Removal of the involved bone is required to decrease the chances of recurrences. **Case Report:** In our study, we have included a case of 28-year-old woman presented with complaints of pain and difficulty in weight bearing over her right lower limb for 2.5 months. Radiograph suggested an eccentric lytic lesion in midshaft of tibia and biopsy revealed granulosus cyst wall, nucleate germinal layer, the brood capsule, and protoscolices with visible hooklets. Patient was subjected to surgery with the excision of cyst along with extended curettage of bone creating a bone defect around the lesion and with anterolateral plating with coverage of bone defect by allogenic bone grafting. Patient was kept on above knee slab with non-weight-bearing mobilization for 6 weeks. Postoperative chemotherapy with Albendazole was given for 3 months. Patient was followed up every 6 weeks for 3 months and every month thereafter on outpatient basis. Return to work and patient satisfaction were excellent. **Conclusion:** Definitive Surgical management with Preoperative and postoperative chemotherapy seems to be effective to avoid recurrence. The bone defect caused by the disease or surgery can be managed with a bone graft either of autograft or allograft.

Keywords: Allogenic bone graft, echinococcosis, hydatid cyst, skeletal hydatosis, tibia

Introduction

It is caused by *Echinococcus granulosus* larvae. Dogs are the definitive host whereas cattle are the intermediate hosts. The disease occurs due to accidental infection of humans with eggs of *E. granulosus* followed by the development of larvae.^[1,2]

It can develop in any part of the body, with the most common site being liver involving 70% of cases. Bone involvement is as low as 0.5%–2.5% of all human hydatidosis. It remains asymptomatic for a long duration and thus diagnosis is made at later stages and radiology reveals extensive disease. Clinical feature depends on the anatomic location of the bone involved. The treatment regime is similar to oncologic therapy rather than the simple surgical excision in the case of visceral hydatidosis.^[3,4]

Like in other visceral organs, pericyst formation does not occur in bone. Thus, it

proliferates aggressively along the areas of least resistance likely in bone canals. With time the disease spreads within the bone tissue to reach the cortex and might spread into surrounding tissues.^[5] Hydatid disease of bone shows well-defined, multiloculated lytic lesion with the appearance of a bunch of grapes. The additional features include expansion of bone, thinning of cortex, and spread into the adjacent tissue. The osteoclastic activity is likely due to impairment of blood supply caused due to pressure erosion and local necrosis.^[6]

The presenting symptoms are pain and swelling with or without pathological fracture. As it lacks characteristic clinical features it may mimic tuberculosis, chronic osteomyelitis, aneurysmal bone cyst, giant cell tumor, solitary cyst, chondrosarcoma, or fibrocystic disease.^[5] The treatment options include surgery followed by a long duration of albendazole. Removal of the involved bone is required to decrease the chances of recurrences. The bone defects

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are covered by either bone graft or bone cement due to its added effect of thermal necrosis.^[7,8]

This paper was aimed to present the case of hydatid cyst of midshaft tibia, which was successfully managed with excision of cyst along with extended curettage and allogenic bone grafting and review of literatures available on skeletal hydatidosis.

Case Presentation

A 28-year-old woman presented with complaints of pain and difficulty in weight bearing over her right lower limb for 2.5 months. visual analog scale score was 8/10. There was a mild swelling over her right leg during the period of 2.5 months. The patient had neither a history of any comorbidities (diabetes, hypertension) nor any significant surgical history. There was no significant history of trauma or tuberculosis contact. The patient did not have any complaints of fever, fatigue, malaise, or weight loss.

On clinical examination, there was localized tenderness present on the anterior aspect of the middle 1/3rd of the leg. There was no local rise in temperature. Both knee and ankle range of motion was within normal limits. The rest of clinical evaluation revealed no other significant findings.

On clinical examination, there was localized tenderness present on the anterior aspect of the middle 1/3rd of the leg. There was no local rise in temperature. Both knee and ankle range of motion was within normal limits. The rest of clinical evaluation revealed no other significant findings. Differential diagnosis includes benign bone tumor (simple bone cyst, fibrous dysplasia, and enchondroma), chronic osteomyelitis, and a brown tumor.

After a routine investigation and following biopsy and radiological findings, a definitive diagnosis of skeletal hydatidosis involving shaft of tibia right side was made, and the patient was subjected to surgery with excision of cyst along with extended curettage of bone creating a bone defect around the lesion and with anterolateral plating with coverage of bone defect by allogenic bone grafting. The patient was then kept on non-weight-bearing mobilization with an above knee slab for 6 weeks. For postoperative chemotherapy, tablet albendazole 400mg once daily for 3 months was given to the patient from Post-operative day-1.

The patient was able to bear weight without pain after 6 weeks as shown in Figure 1. The final histopathological slide of intraoperative specimen was positive for hydatid disease as shown in Figure 2. The patient was followed up

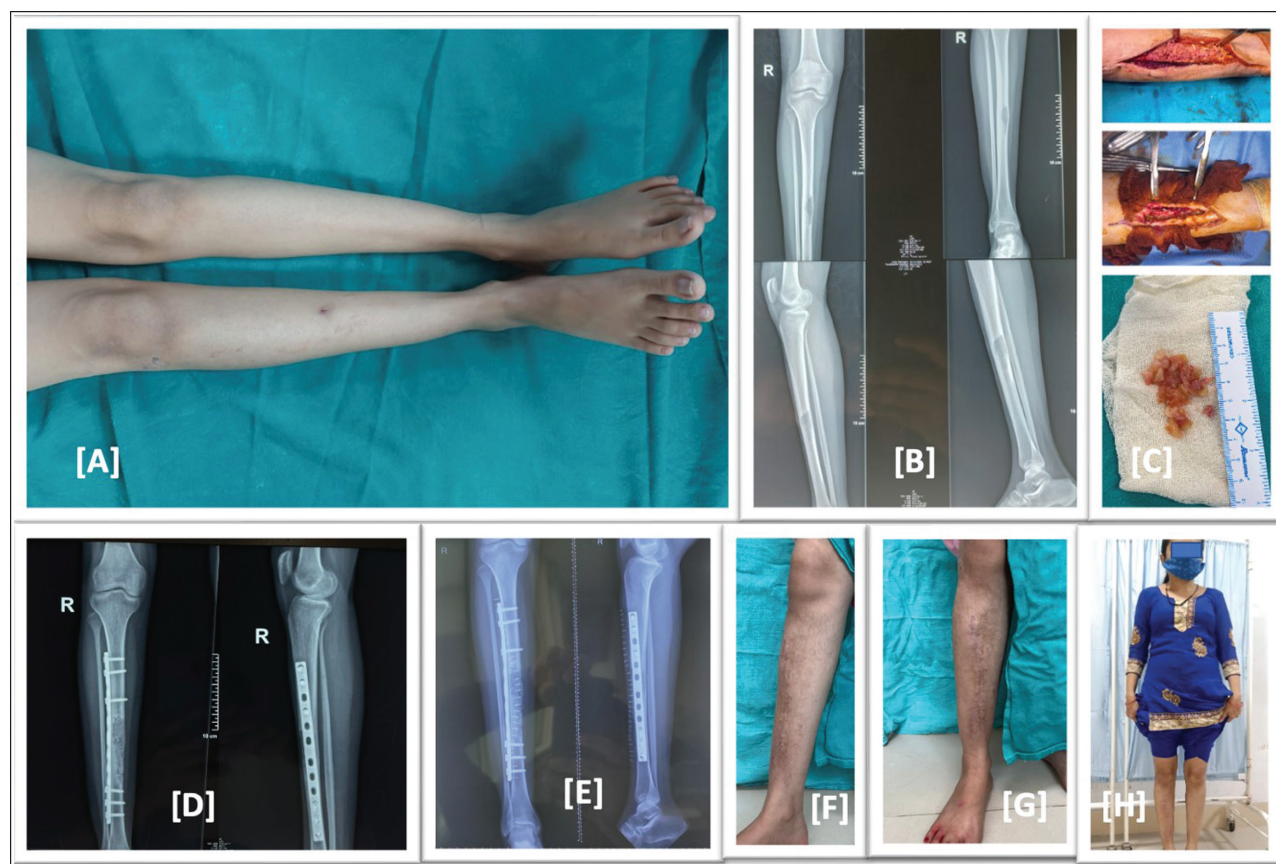


Figure 1: (A) Preoperative image, right leg with scar mark of biopsy and mild swelling. (B) Preoperative X-ray showing expansile lytic lesion in mid shaft tibia. (C) Intraoperative images, excised hydatid cyst mass. (D) Immediate postoperative X-ray showing incorporation of allograft and fixation by anterolateral tibial plate. (E) Post operative X-ray at 6 month follow up showing well incorporation of allograft. (F–H) Clinical images at 6 month follow up showing healed scar mark no swelling or deformity and full weight bearing of the patient with full return to activity of daily living

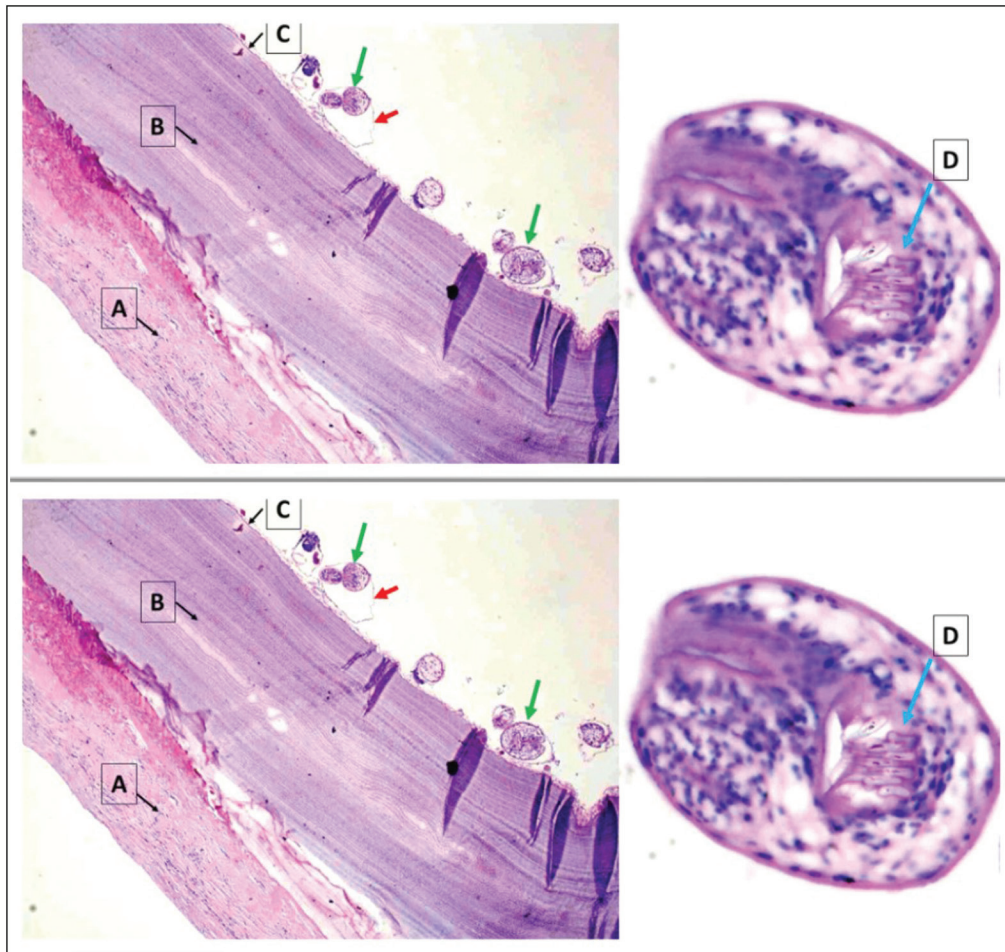


Figure 2: Hematoxylin and eosin (H&E x40) stained section shows *E. Granulosus* cyst wall (A) acellular laminated layer; (B) a nucleate germinal layer; (C) the brood capsule (red arrow), protoscolices (green arrow) with visible hooklets (D)

for a period of 6 months at 6 weeks interval. There were no any radiological signs of recurrence over the 6-month period. There was no any complaints of pain or difficulty in weight bearing. Postoperative visual analog scale at 6-month follow-up was 0/10. The knee and ankle ROM was within normal limits and there was no any localized tenderness. The scar mark of surgery has healed with primary intention. Radiologically, there was adequate sign of uptake of allograft. Serology for hydatid disease was also negative for the disease. Return to work and patient satisfaction were excellent.

Discussion

Hydatid disease is caused by a tapeworm infection, *Echinococcus*, namely *E. granulosus* and *Echinococcus multilocularis*, that infects the human species most commonly.^[9] Man is the accidental host in this disease. Dogs are the definitive hosts whereas cattle are the intermediate hosts. The definitive hosts, dogs, harvest the adult forms in their gut, where the adult form of tapeworm releases its egg into the feces. These eggs are ingested by the intermediate hosts, namely sheep. After the ingestion of the eggs, the embryo is released in the gut and absorbed into the portal

circulation through the duodenal mucosa. The cyst grows in the highly vascular organs such as muscle bed, liver, and lungs of intermediate host. These cysts are ingested by the definitive hosts where the cysts are absorbed in the intestine and grow into protoscolex, scolex, and into an adult form. Humans are infected due to ingestion of eggs of parasite either through food, water or hands contaminated by eggs. After ingestion of the eggs, the embryo is released in the gut and absorbed into the portal circulation through the duodenal mucosa. In the intestinal mucosa, the parasite grows into an adult form and releases egg which are ingested by the intermediate host. A hydatid cyst has three layers, outer pericyst (formed by host cells, limits growth of parasite in visceral organ), middle soft laminated later, and inner germinal layer. In bone infection, the pericyst is absent.^[10-12] In our case the biopsy revealed acellular laminated layer, nucleate germinal layer, protoscolices, and visible hooklets.

The disease mainly involves visceral organ, most commonly the liver (70%). Primary bone hydatidosis is rare and involves 0.5%–2.5%.^[3] It might take 10–20 years for hydatid disease of bone to be clinically noted. Thus, the disease is presented in between 4th and 6th decade and women were twice as commonly infected as men. In our case, the

Table 1: Review of literature of skeletal hydatosis

Author	Age/ sex	Article report	Presenting complaints	Bone involved	Radiograph	Other distant lesion	Biopsy finding	Management	Chemotherapy	Outcome	Follow up	Complication
Babitha <i>et al.</i> ^[9]	52/f	Case report	Dull aching pain	Femur	Fracture with lytic lesion, shaft of femur	Absent	Lamellated linear eosinophilic anucleated membranous structures, granuloma formation and chronic inflammatory reaction, occasional scolices	Interlocking femur nail for pathological shaft of femur fracture	Albendazole 400mg bd for 4 weeks	No non union at fracture site No recurrence	12 months	Non union
Arti	42/m	Case report	Pain, mild swelling	Fibula	Multiple lytic lesion in mid shaft fibula	Absent	Trilamellar cyst and scolices of E. Granulosus	Wide local excision of cyst 10cm above and below lesion	Albendazole 400mg bd for 4 weeks	No recurrence seen	12 months	-
Kalinova <i>et al.</i> ^[5]	45/f	Case report	Pain	Tibia	Oval cystic lesion with diameter of 3.5 cm on diaphysis of tibia, periosteal reaction on cortex	Not mentioned	Osseous tissue with hyaline and germinative membranes, lymphocytes, and monocytes	Povidone iodine injection followed by cystectomy	Albendazole 10mg/kg/day for 12 weeks	No recurrence seen, excellent outcome	24 months	None
Jain	31/f	Case report	Pain	Pelvis (right iliac fossa)	Ill defined lytic lesion with areas of patchy sclerosis, large solid cystic mass of size 10 × 6.3 cm in right iliac fossa	H/O ovarian hydatid cyst	Osseous tissue with laminated membrane of hydatid cyst mixed with lymphocytes and macrophages	Resection of cystic lesion in sacroiliac joint, reconstruction with allograft and autograft (ribs) with lumbosacroiliac fixation	Albendazole for 1 month	No recurrence	6 months	

Table 1: Continued

Author	Age/sex	Article	Presenting complaints	Bone involved	Radiograph	Other distant lesion	Biopsy finding	Management	Chemotherapy	Outcome	Follow up	Complication
Siwach	51/f	Case report	Pain, swelling and deformity of thigh	Femur, Hemipelvis and sacrum with spinal canal left side,	Segmental fracture left femur with honey comb appearance and multiple osteolytic lesion, complete resorption of femoral head and neck, narrow transition zone without reactive bone formation in whole left femur	absent	Trilamellar hydatid cyst wall and scolices of E. Granulosus	Albendazole 10 mg/kg/day, No surgical intervention was done	Albendazole 10 mg/kg/day given as treatment measures	Died due to sepsis and extensive bedsores	1 month	Died due to sepsis and extensive bedsores
Musculo	65/f, 33/f	Case report (2 cases)	Case 1: not mentioned Case 2: pathological fracture	Case 1: right femur Case 2: proximal femur	Case 1: multiloculated osteolytic lesion in diaphysis of femur Case 2: pathological fracture, high signal intensity in femoral head, extensive soft tissue compromise laterally	Case 1: not mentioned Case 2: not mentioned	Case 1: not mentioned Case 2: Not mentioned	Case 1: wide local excision and reconstruction with intercalary allograft, fixation with locked intramedullary nail and four cancellous screws at distal osteotomy site Case 2: wide local excision of proximal femur, proximal femoral prosthesis allograft composite was used to reconstruct the defect, fixation was done with dynamic compression plates and screw	Case 1: preoperative and postoperative chemotherapy was given Case 2: oral albendazole 15 mg/kg/day preoperatively for 1 months and postoperatively for 6 months	Case 1: excellent outcome Case 2: No recurrence, excellent outcome (MSTS score 28/30, no recurrence at follow up) Case 2: No recurrence, excellent outcome (MSTS score 29/30)	Case 1: 108 months Case 2: 60 months	None in both

Table 1: Continued

Author	Age/sex	Article	Presenting complaints	Bone involved	Radiograph	Other distant lesion	Biopsy finding	Management	Chemotherapy	Outcome	Follow up	Complication
Bitar	26/m	Case report	Pathological fracture following blunt trauma after sports injury	Tibia	Pathological fracture of tibia, well defined cystic lesion involving medullary cavity and scalloping of cortex	None	Foreign body granuloma and sheets of lamellated membrane consistent with hydatid cyst	Curettage of lesion followed by reconstruction with bone graft	Albendazole 10 mg/kg/day bd for every 4 weeks out of 6 weeks for 4 months	No relapse, complete healing	44 months	
Alem-daroglu	30/m	Case report	Limping, intermittent pain, swelling	Tibia	Multiloculated mixed lytic and sclerotic lesion in tibia, bunch of grapes appearance, diffusely expanded bone, endosteal thinning, no obvious deformity or fracture	5 × 4 cm cystic lesion in spleen	Cyst of variable sizes with smooth outer membrane, daughter embryos seen in inner layer of cyst	Wide local curettage to create an anterior window on tibia, thermal effect of PMMA applied for 3 min and reconstruction with femoral cortical allograft to cover the anterior defect, splenectomy performed in same session	Albendazole 10 mg/kg/day preoperatively for 1 month	No relapse, complete healing	34 months	Non union of allograft on first procedure
Schnepenheim	54/f	Case report	Pain and swelling	Tibia	Multiple osteolytic lesion with reactive sclerosis	None	Trilamellar cystic wall with scolices of E Granulosus	Wide local curettage and reconstruction of defect with fibular autograft as well as allograft	Postoperative albendazole 10 mg/kg/day and Praziquantal (40 mg/kg/week)	Pain free, asymptomatic and no recurrence, well uptake of allograft	24 months	None

Table 1: Continued

Author	Age/sex	Article	Presenting complaints	Bone involved	Radiograph	Other distant lesion	Biopsy finding	Management	Chemotherapy	Outcome	Follow up	Complication
Gnana-sekaran	25/f	Case report	Discharging sinus on and off	Femur	Cortical thickening and sclerosis with intervening lucencies in diaphysis of femur, mild periosteal reaction with deformity seen	None	Viable and necrotic bones with cyst wall composed of acellular eosinophilic lamellated material surrounded with fibrosis, scolices of E Granulosus with hooklets in germinal layer	Debridement, sequestrectomy and saucerization followed by cotrimoxazole, praziquantel and albendazole, second stage surgery with re debridement with hypertonic saline and hypertonic saline with antibiotic cement spacer for cortical defect	Postoperative praziquantel, albendazole for 6 months	Good wound healing, asymptomatic and no evidence of recurrence	12 months	Recurrence after first debridement

patient was 28-year-old woman, with the complaints of pain and difficulty in bearing weight in her leg. The lesion initially starts in epiphysis or metaphysis in long bones and spreads onto diaphysis at later stages. X-ray and CT scan can aid onto diagnosis and the imaging shows unilocular, bilocular, or multilocular cyst.^[3]

Merkle *et al.*^[13] did a review of literature on 45 patients with 51 skeletal involvements and found the following distribution: spine 35%, pelvis 21%, femur 16%, 10% tibia, 6% in ribs, 4% in scapula and skull, 2% in humerus, and 2% in fibula and concluded that 60% of osseous lesions occur in spine, pelvis, and hip joint. 28% in long bones such as femur, tibia, and humerus and 8% in ribs and scapula. It is often misdiagnosed as tumor due to progressive changes and cystic appearance on radiographs.^[14] In our case, the diaphysis of midshaft tibia was involved where the lesion was expansile, thin-walled, and cystic.

Plain radiograph is considered the investigation of choice. Honeycomb appearance and ill-defined areas of osteolysis are the radiologic features. Periosteal reaction is not seen. CT scan and magnetic resonance imaging are useful in identifying extension of tumor, radiological measurement, and the extent of spread of tumor into the soft tissue.^[15]

The treatment option for hydatid disease of the bone is mainly surgery. But preoperative albendazole followed by surgical removal of cyst followed by postoperative albendazole is considered effective.^[16] In about 25%–30% of cases antihelminthic therapy alone turned out to be ineffective.^[5] We have given preoperative albendazole followed by surgery by wide local curettage around the lesion to create a bone defect that was filled with allograft.

Xie *et al.*^[17] did a retrospective study in 2014 with 40 patients, where 24 patients underwent surgery and 16 patients underwent radiotherapy. Relapse was seen in 14 patients who opted surgery whereas only in three patients postradiotherapy. Pain, bone defects, and limb movement disorder were seen in seven patients with surgery whereas hardening of the irradiated limb was seen in two patients with radiotherapy group. Also, the titers of antibodies of parasites were low among radiotherapy group and patient satisfaction was much better among radiotherapy group.

Gautam *et al.*^[18] reported a case with hydatid disease of femur at the site of nonunion subtrochanteric femur fracture presenting with a lytic lesion. Preoperative albendazole followed by curettage and debridement of lesion with exchange nailing, and cement spacer application was done. Following recurrence of the same case 3 months later, the patient was again operated with excision of lesion and cement spacer application, and the patient was kept on chemotherapy for 6 months. The patient was disease-free as shown by X-ray and magnetic resonance imaging, and a femoral shaft allograft was used to reconstruct the bone defect along with the proud nail which was locked proximally

and distally. At 6-month follow-up, the patient was well, with complete incorporation of allograft and no limb length discrepancy. Another study where femoral allograft was used for the bone defect after wide resection of lesion caused by hydatid disease of bone was done by Muscolo *et al.*^[19] Two cases were included in the study where wide local excision of the lesion in the femur followed by femoral allograft and fixation of the allograft with implant was done. In patient-1, where the distal femoral diaphysis was involved allograft was fixed by locked intramedullar nail and four cancellous screws at distal osteotomy site. The outcome was excellent. In this case, the patient had not received any antihelminthic therapy preoperatively or postoperatively. In Patient-2, the patient presented with pathological fracture of proximal femur due to hydatid disease. She was also operated with wide local resection and reconstruction by proximal femoral allograft, followed by fixation with plating. This patient received both preoperative and postoperative chemotherapy treatments, and the outcome was also excellent in this patient. In our case, we did an extended curettage around the cystic lesion in the mid shaft tibia, coverage of defect by allograft, and fixation and stabilization by 12-hole narrow plate. The review of available literature is summarized in Table 1.

Conclusion

Skeletal involvement is rare, accounting for less than 3% of hydatid disease. They are often diagnosed late or misdiagnosed as usual hematological and serological tests for hydatid disease are absent. Radiological features are usually suggestive of a lytic lesion of the bone. Biopsy is helpful in diagnosing skeletal hydatid disease. Preoperative chemotherapy followed by a surgery followed by postoperative chemotherapy seems to be effective to avoid recurrence. The bone defect caused by the disease or surgery can be managed with a bone graft either of autograft or allograft.

Patient's perspective

Satisfactory with outcome.

Informed consent for publication

Informed consent was obtained from the patient for the publication of this case report. On request, a copy of the written consent is available for review by the Editor-in-Chief of this journal.

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Conflicts of interest

There are no conflicts of interest.

Ethical review committee statement

Not applicable.

Author contributions

B.B.N.—Planning of study, data management, writing, and revising the manuscript.

A.R.—Data management, manuscript preparation.

S.B.—Planning of study, revising the manuscript.

M.D.—Revising the manuscript.

R.H.P.—Data management.

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