Primary Gluteal Intramuscular Hydatid Cyst : A Case Report

ABSTRACT:

INTRODUCTION: Hydatid disease is a parasitic infection caused by the tapeworm, Echinococcus granulosus. It commonly occurs in the liver and lungs. Primary intramuscular hydatidosis is a rare presentation.
CASE REPORT: We report a 68-year-old lady diagnosed with primary gluteal intramuscular hydatid disease with no involvement of any other viscera. Perioperative Albendazole therapy with surgical excision was done. Follow-up till 3rd month after the surgery was maintained. No signs of local recurrence were seen.
DISCUSSION: The primary intramuscular hydatid cyst is an uncommon location because the lactic acid in muscles and contraction of muscles hinders the development of parasitic cysts. Treatment of choice is complete surgical excision of the cyst along with thorough irrigation of the wound with hypertonic saline (3%) to prevent local recurrence and anaphylaxis. Systemic antihelminthic therapy with Albendazole before and after the surgery also plays an important role.
CONCLUSION: Diagnosis of primary gluteal hydatid cyst in an isolated form is difficult. However, it must be kept in mind by the clinicians in patients coming from endemic areas with musculoskeletal swelling involved in agricultural work.

KEYWORDS: Hydatid cyst; Intramuscular; Gluteal swelling; Soft tissue swelling; Echinococcus; Albendazole; Recurrence.

INTRODUCTION:

Hydatid disease is a zoonotic larval infection caused by tapeworm, Echinococcus granulosus. It is characterised by the growth of parasitic cysts in the organs of intermediate hosts like human beings, who accidentally ingest eggs which are excreted in the faeces of definitive hosts like dogs contaminating food and drinking water [1,2]. Geographically, it has a higher prevalence in the Mediterranean, Russia, China, North and East Africa, Australia and South America [1,2]. In India, the southern states of Andhra Pradesh and Tamil Nadu are commonly associated with hydatid disease [7]. The liver and Lungs are the most frequently affected organs. Although less frequently, it can also affect the heart, brain, vertebral column, spleen, ovaries and pancreas. Primary soft tissue involvement such as thigh muscles is extremely rare, having an incidence of 0.5-5.3% of all hydatid cases [1,3, 7]. The patient can present with a painless, gradually progressing mass in any of the extremities which may mimic the appearance of soft tissue tumours like sarcoma, lipoma or liposarcoma. Preoperative clinical examination and appropriate radiological investigations like ultrasonography and magnetic resonance imaging are very important in order to avoid misdiagnosis and prevent unnecessary fatal complications [7,8].
In this case report, we discuss a 68-year-old lady diagnosed with a primary gluteal intramuscular hydatid cyst, successfully managed with surgical intervention and antihelminthic therapy.

CASE REPORT:

A 68-year-old lady, a resident of the rural Kutch region of India, visited the surgical outpatient department of G.K. General Hospital, Bhuj, in October 2023, with complaints of right gluteal swelling for 30 years. The swelling was insidious in onset and spontaneous in origin. It had gradually increased in size over the years to a size of 10x10cm on presentation. It was associated with discomfort while walking and prolonged sitting for the past 3 months. There was no history of a rapid increase in size or regression in the size of the swelling. There is no history of changes in the overlying skin. There was no history of pain over the swelling. There is no history of fever or trauma at the local site. There are no known comorbidities. The general examination was within normal limits. Local physical examination revealed a firm, non-tender mass of approximately 10x10cm in the lower inner quadrant of the right gluteal region, which extended to the posteromedial aspect of the right thigh. The overlying skin was normal. There were no similar swellings present elsewhere in the body. Routine blood investigations were within normal limits. Ultrasonography of the right gluteal region was suggestive of a well-defined multiloculated anechoic cystic lesion measuring approximately 9.1x6.3cm with internal septations, and few internal echoes were seen at the local site of swelling in the right gluteal region, indicating the possibility of infective aetiology such as hydatid cyst. Magnetic resonance imaging (MRI) revealed the presence of a 90x84x108 mm well-defined multiloculated altered signal intensity lesion seen involving the skin, subcutaneous plane, and intramuscular plane of the posteromedial aspect of the right thigh region. Lesion abuts and displaces the adductor longus, adductor brevis, adductor magnus, semimembranosus, semitendinosus, and gluteus maximus muscles. The femoral vessels and sciatic nerve appeared normal. Possibility of infective aetiology, likely to be a hydatid cyst. Chest radiographs and ultrasonography of the abdomen and pelvis did not reveal any other organ involvement. Based on history, clinical examination, and radiological investigations, a diagnosis of intramuscular gluteal hydatid cyst was made. The patient was planned for preoperative and postoperative anti-helminthic therapy with Albendazole along with surgical excision. The patient was started on Albendazole 400 mg twice daily 5 days before the surgery and was planned for surgery after taking well-informed consent. A single oblique incision of 12cm was made over the swelling, and dissection was deepened in layers. A large single tense cyst having an intramuscular extent in the posteromedial compartment of the thigh with superior extension into the lower inner quadrant of the right gluteal region was present. After securing the wound with hypertonic saline (3%) irrigation, the cyst wall was opened. It contained multiple daughter cysts along with approximately 750 ml of dirty, sanguineous fluid. Deroofing of the cyst with the evacuation of multiple daughter cysts was done. Intraoperative irrigation of the wound with hypertonic saline (3%) was done regularly to prevent the risk of recurrence and anaphylaxis. The wound cavity was thoroughly washed with 10% povidone-iodine after a complete pericystectomy. A negative suction drain was kept in the wound cavity, following which primary closure of the wound was done. The patient was continued on Albendazole 400 mg twice daily after the surgery for 3 months to prevent the risk of local recurrence. She was discharged in hemodynamically stable condition on postoperative day 7, and the drain was removed on postoperative day 5. Follow-up on days 15, 1st month, and 3rd month was done, during which clinical examination of the primary incision site revealed no signs of recurrence and was confirmed by ultrasonography of the local part.

DISCUSSION:

Hydatid disease is an endemic zoonotic larval infection caused by the tapeworm “Echinococcus Granulosus”. It is significant in countries where sheep rearing is common; but

due to enhanced immigration throughout the world, it is supposed to be found everywhere now, even in developed nations. In India, the southern states of Andhra Pradesh and Tamil Nadu are commonly associated with Hydatid disease [7].
It can be classified as primary or secondary. Primary disease occurs due to accidental ingestion of ova by human beings which are excreted in the faeces of definitive hosts[2]. As a result, it is more frequently seen in people involved in farming and animal breeding[3]. The ingested egg then can spread to almost any organ. Liver and Lungs are the most commonly involved organs[1,3,7,8]. In the secondary form, there is a proliferation spread of larval tissue to a site distant from its primary location like the liver, lungs or spleen[7].
Primary intramuscular hydatidosis is an extremely rare presentation having an incidence of 0.5 to 5.4%[4,7,10]. It is uncommon because the lactic acid in the muscle and muscular contraction create an environment unsuitable for the survival of parasites in muscles[5,7]. Parasites have a propensity to grow around muscles which are more vascular and have limited activity. The cysts grow slowly and over time progress to form space-occupying lesions which cause pressure effects on surrounding vital tissues[7]. Pathogenesis behind the muscular location is still a controversy. Direct implantation through the wound, as seen in dog bites and muscular spread via systemic circulation after passing through liver sinusoids and the capillary bed of the lungs are two commonly proposed explanations by some authors [7,10].
Important differentials of hydatidosis include soft tissue tumours and traumatic lesions like hematoma and abscess. Incisional biopsy and marginal excision are contraindicated in hydatidosis due to the increased risk of disease spread and anaphylaxis[7-10]. It is also important to rule out other probable primary locations of the disease by performing appropriate clinical and radiological examinations of the patient[9].
Muscular hydatid disease commonly presents as a gradually progressive painless mass with normal overlying skin[5,10].
Casoni skin tests and various serological tests are convenient in diagnosing hydatidosis. They are useful in distinguishing hydatid cysts from other non-parasitic cysts and abscesses. However, false negative results can occur in encapsulated lesions. Therefore, a negative test does not rule out the possibility of hydatidosis[7].
Radiological investigations like ultrasound, computed tomography and magnetic resonance imaging help describe the exact location and characteristic features of the cyst-like cystic membranes, septa, hydatid sand, floating membranes, daughter cyst, vesicles, cyst wall calcification and cyst infection. However, ultrasonography and computed tomography are not ideal for identifying endo vesicular daughter cysts in cases of skeletal muscle cysts.
Magnetic resonance imaging (MRI) is the investigation of choice in terms of intramuscular hydatid disease. MRI images help in identifying different patterns of intramuscular hydatidosis like peripheral rim (known as the rim sign), membranes in the cyst and peripheral enhancement with contrast occurring due to vascularity of the cyst wall[7,9].

Lamghari et al reported a 22 year old Moroccan man diagnosed with left gluteal hydatid cyst which showed gluteus medius muscle involvement along with cortical destruction of iliac bone on magnetic resonance imaging[5].

V. Lakhanpal et. al also reported a 52 year old gentleman, cattle rearer in rural India diagnosed with left thigh intramuscular hydatid cyst. Swelling was causing compression of the anterior neurovascular bundle of the left thigh. Surgical excision with postoperative

Albendazole therapy for one month was followed. Regular follow-up till 3 months after surgery revealed no signs of recurrence[7].

Soltany et al. conducted a case series study on musculoskeletal hydatid cyst from 2009 to 2014 where they diagnosed 15 cases of musculoskeletal hydatidosis out of 208 patients with hydatid cysts. Out of those 15 patients, only 5 patients had recurrence and 3 patients had simultaneous other viscera involvement. 14 patients were diagnosed before the surgery and only 1 patient was operated with a misdiagnosis of lipoma[10].

It is important to suspect the diagnosis of a hydatid cyst before the surgery, as there is a risk of spilling of contents of the cyst into the systemic circulation during the surgery. This can lead to a fatal anaphylactic reaction. Ideal treatment of choice includes complete surgical excision of the cyst along with thorough irrigation of surrounding tissues with hypertonic saline (3%) to reduce the risk of local recurrence. This is combined with antihelminthic therapy using Albendazole after the surgery[7-9]. A regular follow-up in the long term is advised after the surgery to keep a check on recurrence[7-10].

CONCLUSION:

Diagnosis of primary gluteal hydatid cysts in an isolated form is difficult. However, it must be considered as one of the differentials when associated with other primary locations and endemic area residents involved in agricultural work.
Surgical removal of the cyst with perioperative antihelminthic therapy is the best treatment to reduce the risk of local recurrence.

Ethical Approval:
As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

Consent
As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

AUTHOR'S CONTRIBUTIONS:

All the authors have worked in collaboration while preparing the manuscript. Final version of the manuscript was read and approved by all the authors.

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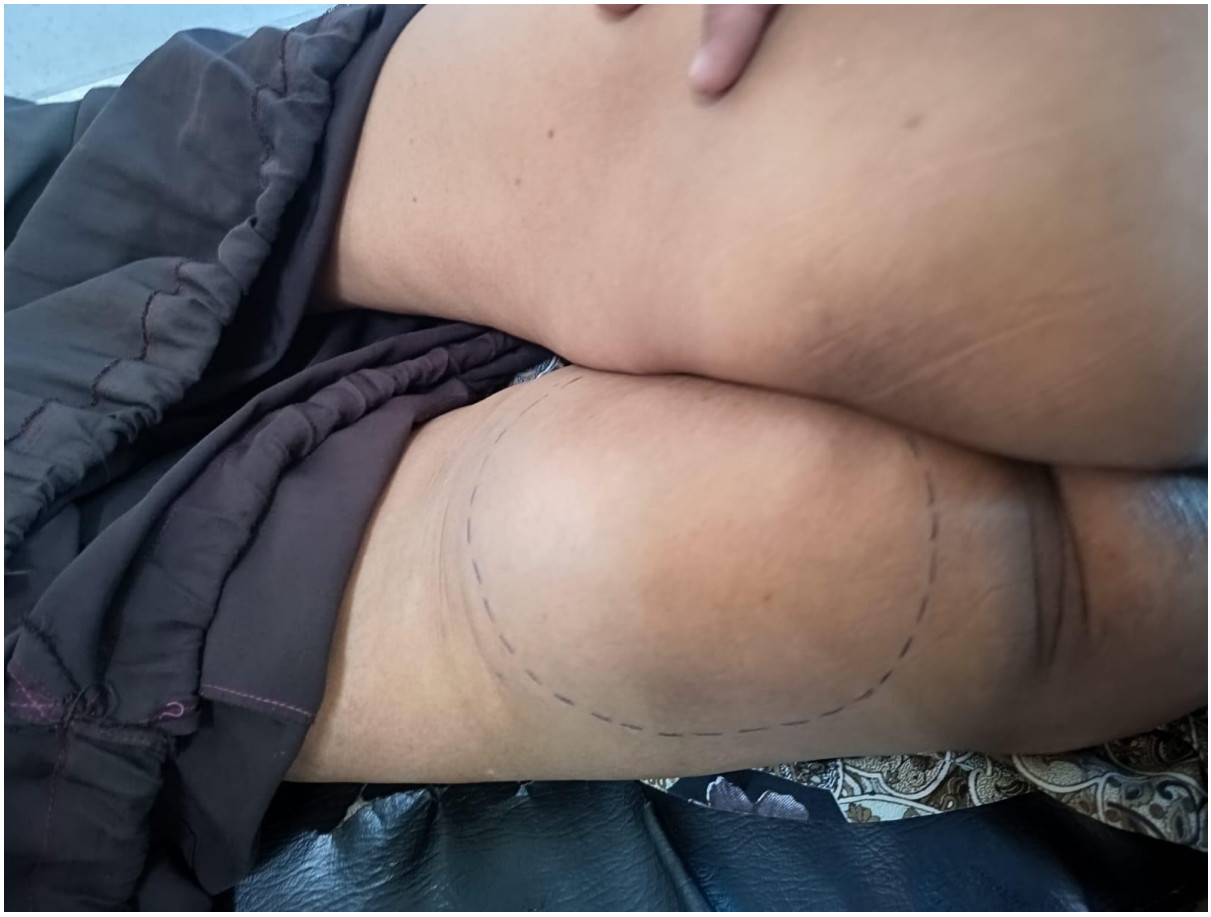
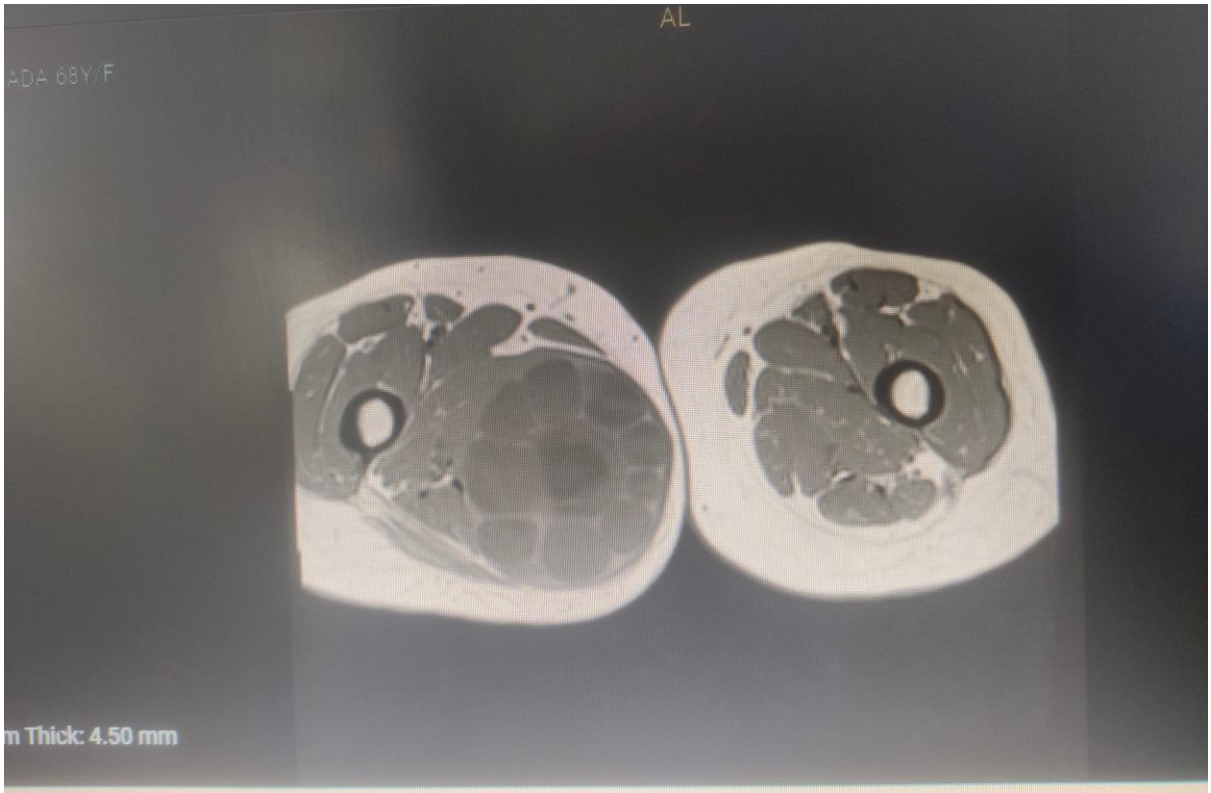


Fig 1. 68 year old lady with right gluteal swelling (10x10cm, boundaries marked during clinical examination).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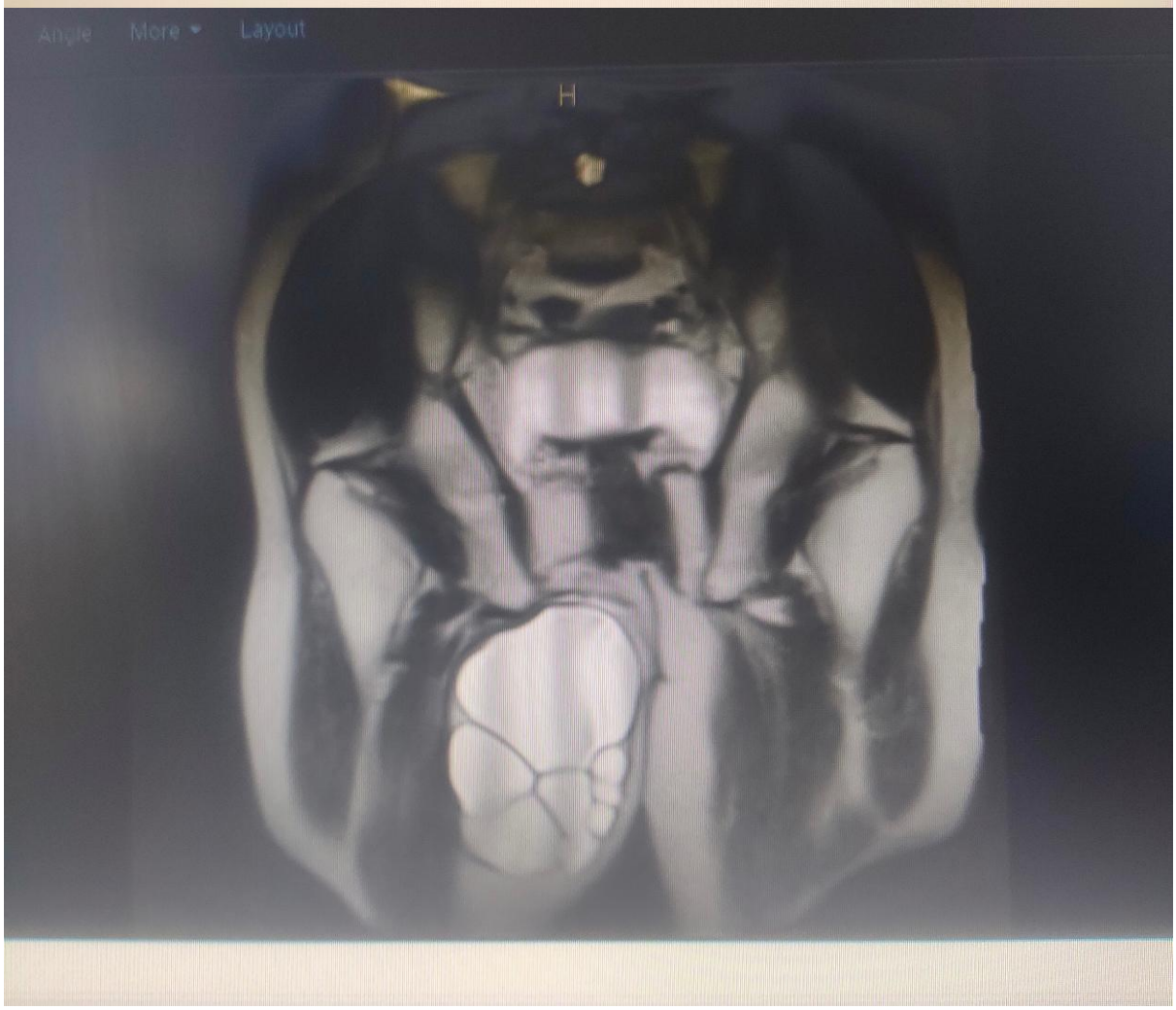
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Fig 2 a and b. Magnetic resonance imaging images showing a multiloculated mass, having intramuscular extension in the right gluteal and thigh region.

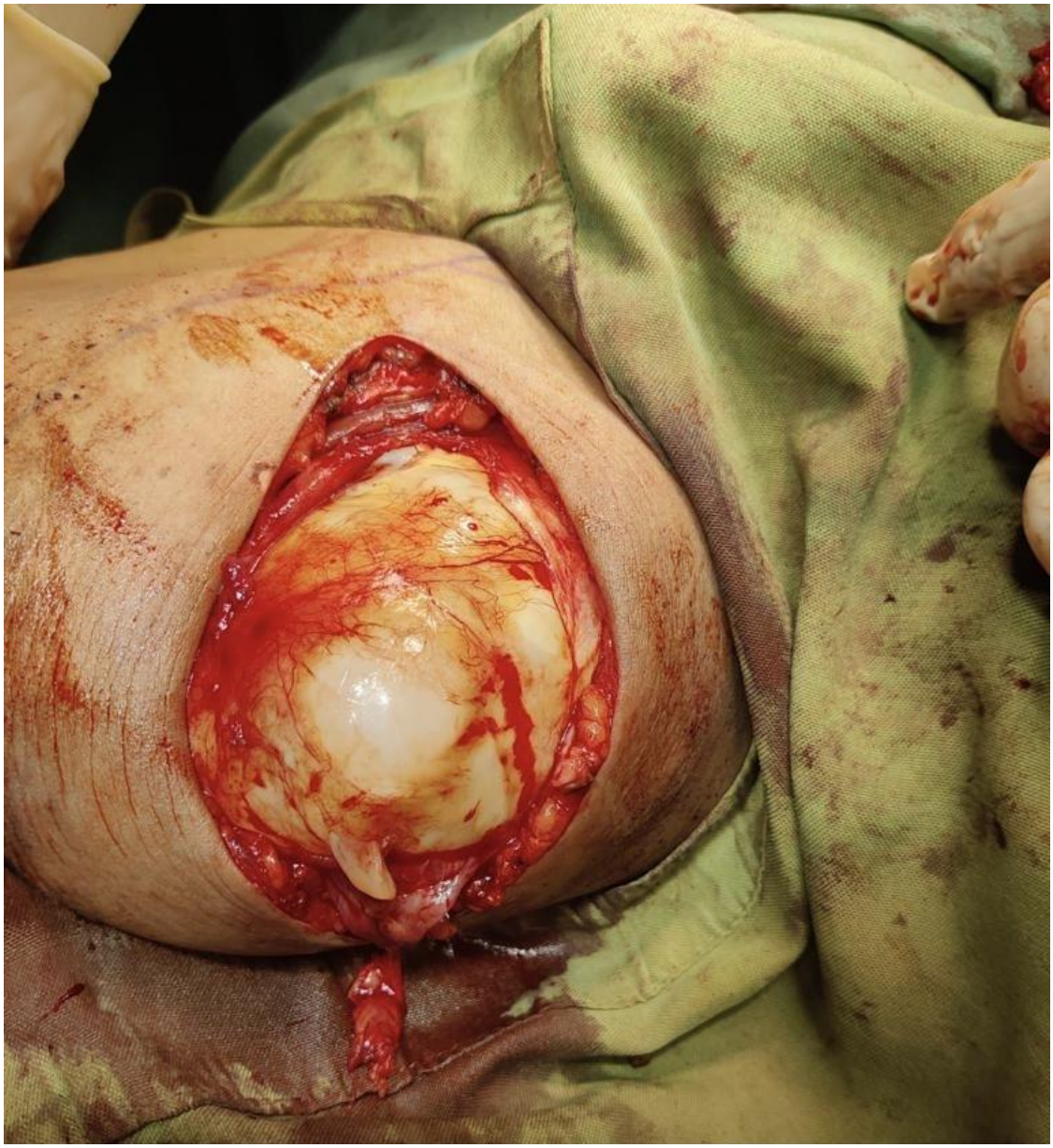

Fig 3. Intraoperative image of the hydatid cyst having intramuscular extension (a single 12cm oblique incision was given over the swelling)

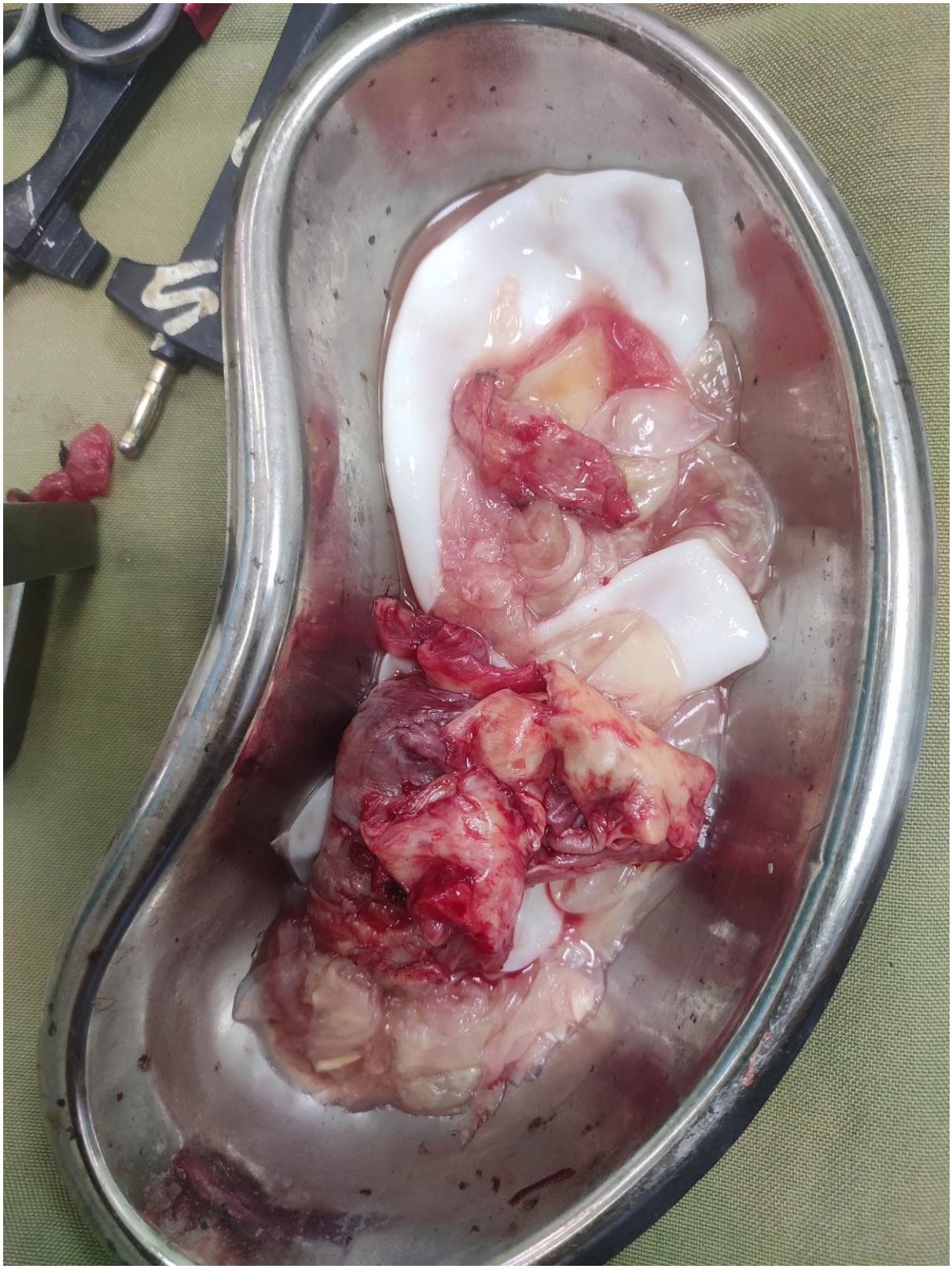

Fig 4a and b. Showing multiple daughter cysts and complete cyst wall after en-bloc excision of the cyst.