

## Intramuscular Hydatid Cyst: A Literature Review on Clinical Findings, Diagnosis, and Treatment

### Kas Yerleşimli Kist Hidatik: Klinik Bulgular, Tanı ve Tedavi Üzerine Literatür Derlemesi

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#### ÖZ

Bu makalenin amacı, Echinococcus granulosus'un neden olduğu zoonotik bir hastalık olan kist hidatik hakkında kapsamlı bir yaklaşım sunmaktır. Parazit, yaşam döngüsünü köpeklerde ve koyunlarda sürdürür; insan enfeksiyonu ise bu hayvanlarla aşırı temas veya onların dışkılarıyla bulaşmış yiyecek veya suyun tüketilmesi sonucunda gerçekleşir. İzole müsküler kist vakalarında negatif serolojik test sonuçları mümkündür ve tanı dikkatli olunması gerekir. Endemik bölgelerdeki kistik lezyonlardan ve tipik görüntüleme özelliklerinden şüphelenilmelidir. İnvaziv prosedürlerden kaçınmak parazit yayılımını önlemek için hayati öneme sahiptir. Klasik görüntüleme bulguları ile endemik bölgeler arasındaki bağlantı, herhangi bir kistik lezyonun bu hastalığın varlığı açısından potansiyel bir tehlike işareti haline gelmesi demektir. Bu makalede biri sakral paravertebral bölgede, diğeri gluteus maximus kasında olmak üzere sporadik durumlar olan iki primer hidatik kist olgusu sunulmaktadır.

**Anahtar Kelimeler:** albendazol; ekinokokkoz; hidatik kist; kas; parazitoloji

#### ABSTRACT

The aim of this article is to provide a comprehensive approach to hydatid cyst, a zoonotic disease caused by Echinococcus granulosus. The parasite maintains its life cycle primarily in dogs and sheep; human infection occurs through excessive contact with these animals or the consumption of food or water contaminated with their feces. In isolated muscular cyst cases, negative serological test results are possible, requiring careful attention during diagnosis. Cystic lesions in endemic regions and typical imaging features should raise suspicion. Avoiding invasive procedures is vital to prevent the spread of the parasite. The association between classical imaging findings and endemic regions suggests that any cystic lesion may be a potential indicator of this disease. In this article, two sporadic cases of primary hydatid cysts are presented—one located in the sacral paravertebral region and the other in the gluteus maximus muscle.

**Keywords:** albendazole; echinococcosis; hydatid cyst; muscle; parasitology

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## INTRODUCTION

Hydatid disease is a parasitic zoonosis primarily observed in regions where animal husbandry is prevalent. The most common causative agent of hydatid cysts is *Echinococcus granulosus*, while *Echinococcus alveolaris* is a less common etiological agent.<sup>1</sup> Hydatid cysts are caused by the parasitic tapeworm *Echinococcus granulosus*. Dogs, sheep, and cattle are the primary hosts; humans are merely incidental intermediate hosts.<sup>2-4</sup> This disease is more common in Africa, Europe, the Middle East, Central and South America, Australia, and Russia.<sup>3-5</sup> Hydatid cyst, endemic in our country, is commonly detected in Central Anatolia, Southeastern Anatolia, and Eastern Anatolia regions.<sup>5</sup>

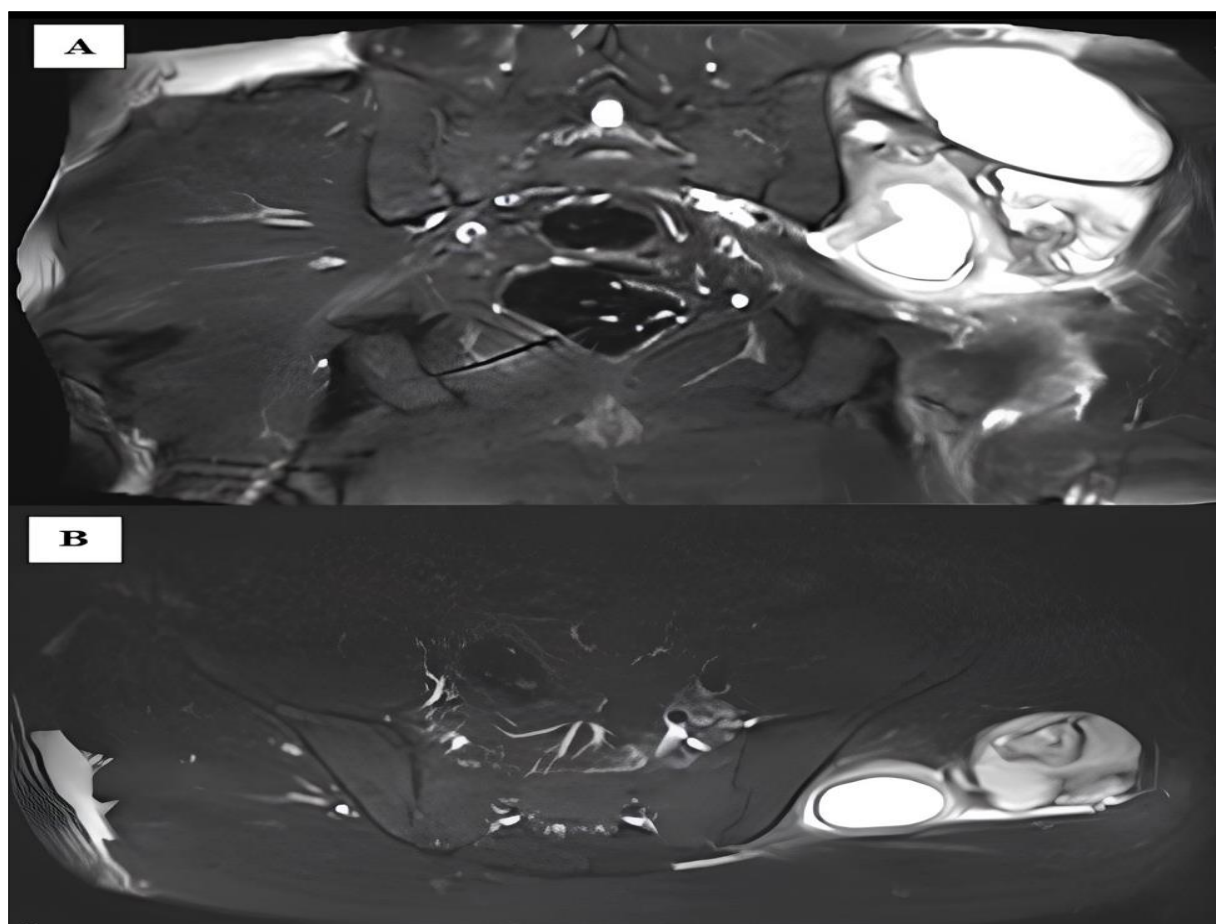
Hydatid cyst mainly affects the liver and lungs but can occur anywhere in the body.<sup>5</sup> While it is most commonly located in the liver (60%) and lungs (30%), hydatid cysts are rarely found in other organs such as the kidneys, spleen, peritoneum, bones, and brain. Approximately 1–5.4% of detected hydatid cyst cases involve the skeletal muscles.<sup>6</sup> Spinal paravertebral hydatid cyst constitutes less than 1% of all hydatid cysts.<sup>7</sup> Sacral hydatid cyst, which includes 20% of spinal hydatid cysts, is much rarer.

The infrequent involvement of the gluteus maximus muscle without signs of hepatic or pulmonary disease is noteworthy. This rare manifestation of primary intramuscular hydatidosis in the gluteus maximus muscle was recorded in 15 patients with bone and muscle echinococcosis by Chiattoni et al. and Toprak et al. Interestingly, only one instance of the gluteus maximus muscle being affected was reported among their cases.<sup>8</sup> This article presents two primary hydatid cysts, one in the sacral paravertebral region and the other in the gluteus maximus muscle. The occurrences in these sporadic areas underscore the importance of maintaining a high index of suspicion when dealing with cystic swellings in muscles, particularly in regions where the disease is endemic.

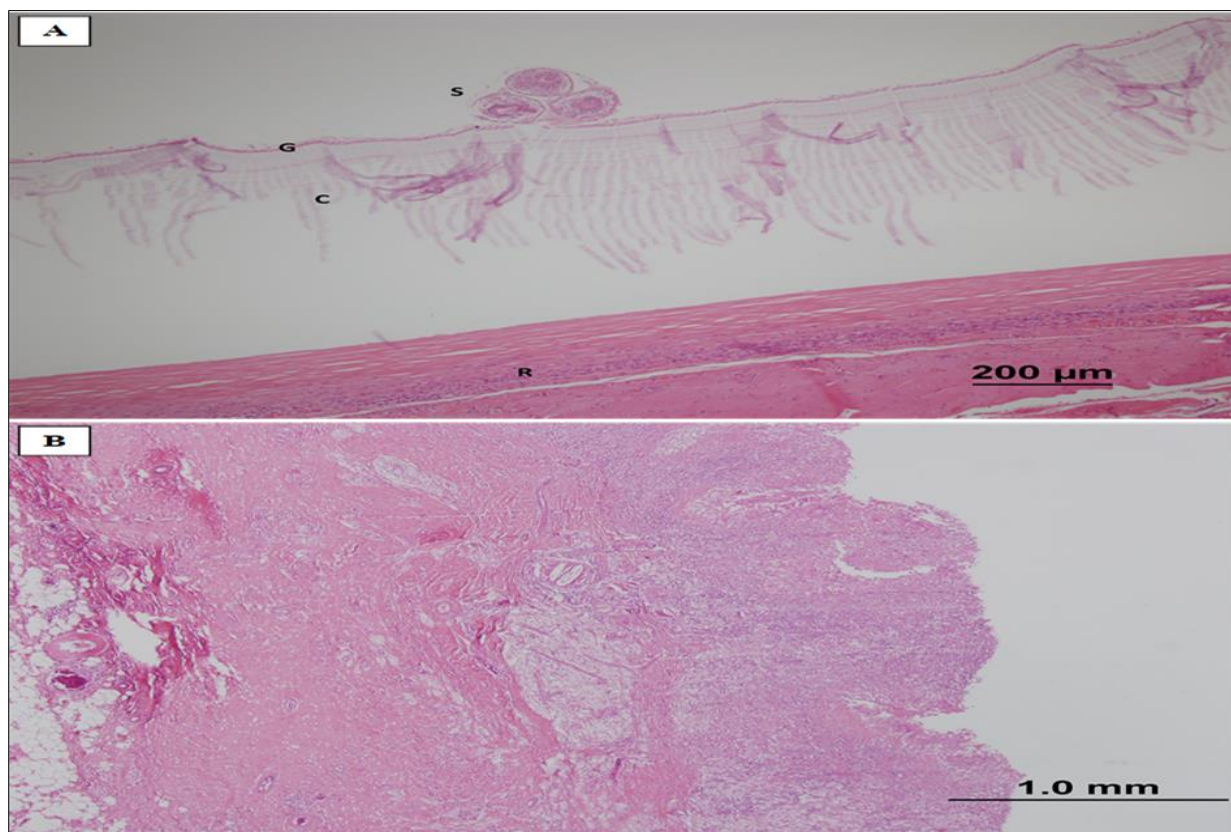
## CASE REPORTS

Patients were enrolled in the study following the acquisition of written consent, allowing for the utilization of their data in academic publications and for further examination and treatment.

CASE-1: A 27-year-old married male patient reported experiencing left gluteal pain for the past three months following a traumatic incident. The intensity of the pain escalated in the week leading up to the visit, resulting in gradual lameness of the left extremity. The patient had no previous medical history. During the physical examination, swelling was observed in the upper lateral quadrant of the left gluteal region without any signs of redness, tenderness, or warmth. Neurological and other examinations yielded the expected results. The left gluteal USG revealed a cystic lesion measuring 46x22x53 mm in the upper lateral quadrant. The pelvic region's magnetic resonance imaging (MRI) identified a cystic lesion measuring 54x32x60 mm within the left deep gluteal muscle. Multiple internal locations suggested a hydatid cyst (Figure 1). The tomography (CT) evaluation detected no fractures or lytic or sclerotic lesions in the bone structures. The patient underwent an *Echinococcus* serological test, which was reported to be positive. To investigate the primary focus, chest radiography, abdominal ultrasonography (USG), and abdominal and thorax tomography were taken, and no focus on hydatid cysts was detected. In response to the persistent and intense gluteal pain, the patient underwent a surgical cystectomy. To prepare for the procedure, albendazole was administered at a dosage of 400 mg twice daily during the week leading up to the surgery. The mass was meticulously excised en bloc under general anesthesia, with careful attention to avoiding puncturing the cyst wall (Figure 2). The postoperative period was uncomplicated. The patient was discharged five days post-surgery and prescribed albendazole at a dosage of 400 mg twice a day for a three-month duration. During follow-up appointments at the first postoperative week, first and second months, and the final third-month follow-up, the patient remained symptom-free with no signs of recurrence.



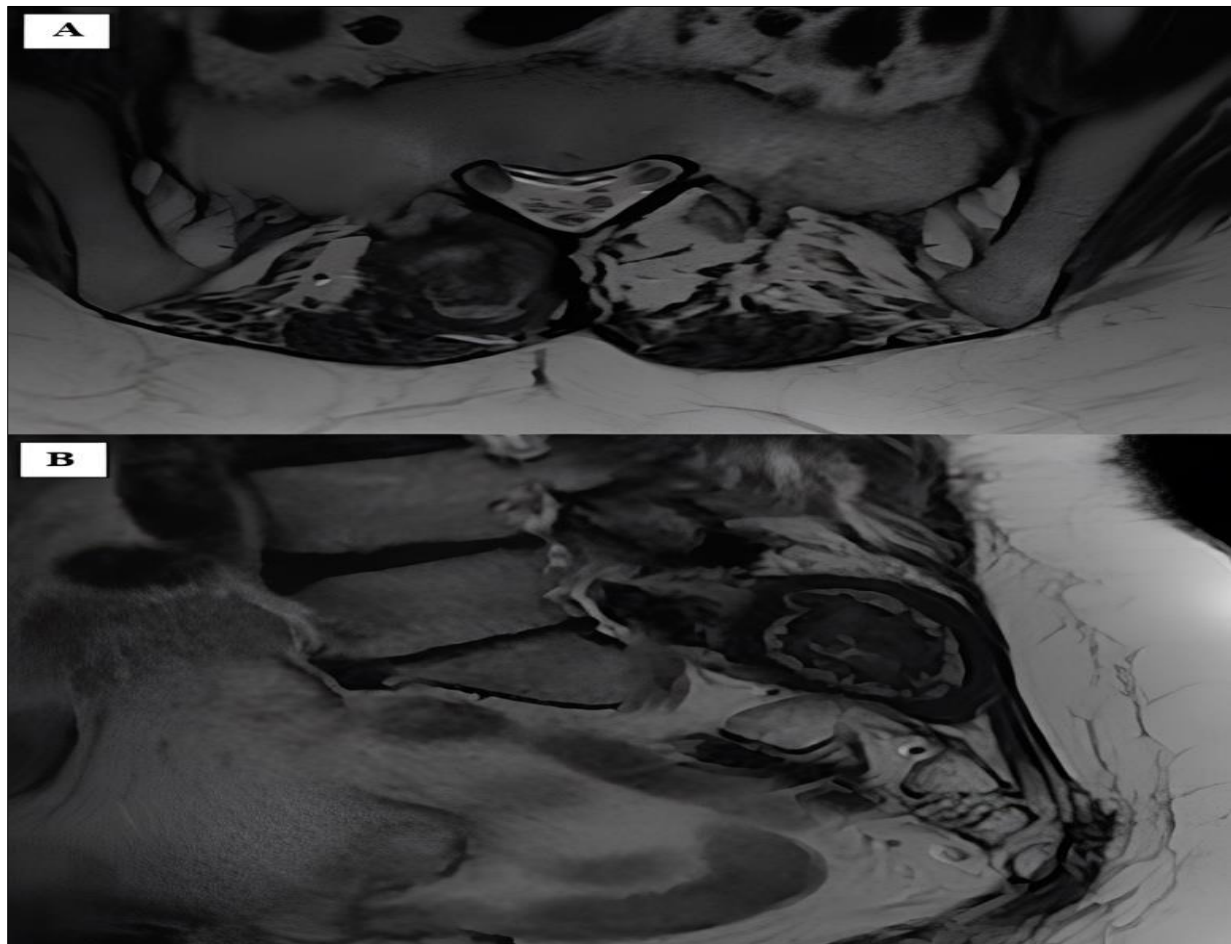
**Figure 1.** A cystic lesion within the left deep gluteal muscle in the coronal plane (A), in the axial plane (B) at T2-weighted images on MRI.



**Figure 2.** A: Image of hydatid cyst wall (H&E stain, x100) (S:Scolex G: Germinative membrane C:Cuticular membrane R:Reactive membrane) B: There are foamy histiocytes, foreign body reaction, and necroinflammatory process around the hydatid cyst (H&E stain, x40).

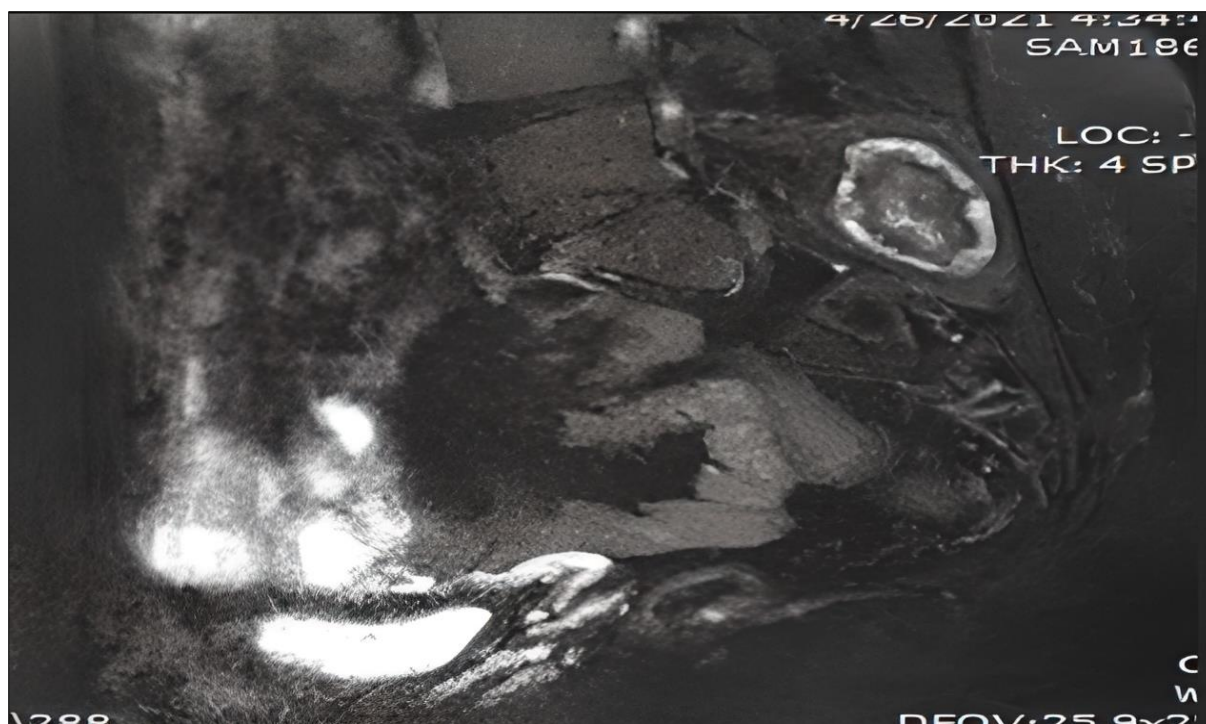
CASE-2: A 57-year-old married female patient applied with complaints of swelling and pain in the sacral region that started two months ago. Her pain had been severe for about one month, and she had difficulty lying down. There was no history of medical illness in her medical history. On physical examination, the joint range of motion was complete, and neurological examination was normal, but there was palpable swelling in the sacral region without erythema, tenderness, or warmth. In the ultrasonographic study of the patient, a 49\*38\*36 mm, oval-shaped, well-circumscribed mass lesion was detected on the right side of the spinous processes in the sacral region, so an MRI examination was performed on the patient. A lobulated contoured lesion was detected, extending in a segment of approximately 42 mm in the right posterior neighborhood at the level of S1 and S2 vertebrae, hypointense in the T1 series (Figure 3), hyperintense in the T2 series, slightly hypointense in the center, and showing peripheral enhancement in contrast-enhanced series (Figure 4). In the evaluation made by tomography, no fractures, lytic, or sclerotic

lesions were detected in the bone structures. The patient underwent an Echinococcus serological test, which was reported to be positive. To investigate the primary focus, chest radiography, abdominal ultrasonography, and abdominal and thorax tomography were taken, and no focus of hydatid cyst was detected. The patient underwent surgical treatment (Figure 5) due to pain and difficulty lying down. One week before the surgery, the patient received albendazole at a dosage of 400 mg twice daily. The mass was then surgically excised en bloc under general anesthesia, with meticulous care to avoid puncturing the cyst Wall, and the postoperative period proceeded without complications. Following a pattern similar to the previous case, the patient was discharged with a prescription for a three-month course of albendazole, 400 mg twice daily. The patient remained symptom-free with no recurrence observed during the first postoperative week, first and second-month follow-ups, and the final follow-up in the third month.

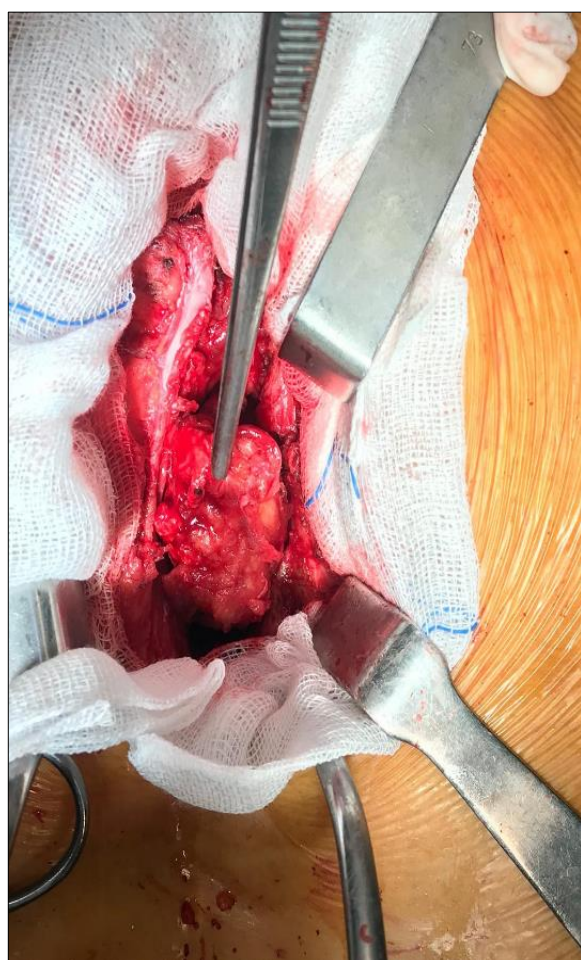


**Figure 3.** A: A lobulated contoured cystic lesion adjacent to S1 and S2 in the axial plane at T1-weighted images. B: A properly limited hypointense cystic lesion in the sagittal plane at T1-weighted images.





**Figure 4.** Hyperintense lesion at T2-weighted images; central mild hypointense, lobulated contoured lesion showing peripheral contrast in contrast-enhanced series on MRI.



**Figure 5.** Image of intraoperative cystic lesion.

## DISCUSSION

CH (cyst hydatid) is a disease caused by the intestinal parasite *Echinococcus*. The most common agent is *Echinococcus granulosus*.<sup>9</sup> Although endemic in many regions, including our country, hydatid disease can be seen worldwide.<sup>10</sup> This pathology is commonly observed in various organs, particularly the lungs and liver. However, there have been rare reports of its occurrence in other locations, including the uterus and gluteal muscle.<sup>2-4</sup> Lactic acid accumulation in the muscles is the most critical reason why muscle involvement is less common because lactic acid negatively affects the growth of hydatid cysts.<sup>11</sup>

Osseous involvement in CH is reported to be 0.5-2.5%. 35% of them are located in the spinal region, 21% in the pelvis, 16% in the femur, and 10% in the tibia. Involvement of ribs, skulls, scapula, humerus, and fibula is less common.<sup>12</sup> As mentioned before, hydatid cysts in muscles are rare and are seen at a rate of 0.5% to 5.4% in the literature.<sup>5</sup> Spinal paravertebral hydatid cyst constitutes less than 1% of all hydatid disease. It is much rarer than sacral hydatid cysts, which include 20% of spinal hydatid cysts.<sup>7</sup> Gluteal region muscle involvement is rare and has been reported in a few studies.<sup>2,13-15</sup> In all documented cases, the cyst was effectively treated through surgical intervention

without complications. Musculoskeletal hydatid cysts, including those in the gluteal region, typically manifest as a chronic painful mass. Additionally, most patients presenting with such cysts have a prior history of hydatid cysts.<sup>14</sup> It's worth noting that this condition may present independently or concurrently with hydatid cysts in other organs. In the initial case, the patient experienced a painful mass, specifically in the gluteal muscle. Our second case presented with swelling and pain in the sacral region; other organs were normal in both cases. Although these areas are rare, hydatid cysts should be considered in the differential diagnosis of any painful mass in endemic regions.

Diagnosing a hydatid cyst is typically established through a combination of factors, including the patient's medical history, physical examination, imaging findings, and serological tests.<sup>2,5,14</sup> ELISA is a serological test that diagnoses CH with 95-97% accuracy.<sup>9</sup> Patients with hydatid cysts frequently have a history of contact with animals, particularly dogs, and reside in rural areas where sheep or cattle farming is prevalent.<sup>15</sup> Ultrasound (USG), CT scan, and MRI are imaging modalities that can reveal both the extent of involvement in adjacent tissues and the features of the cyst. MRI is often considered more sensitive in cases involving muscles, particularly in assessing the depth of the mass. USG is valuable in diagnosis as a first-line imaging method. It can be detected in forms ranging from pure cystic lesions to solid appearance on USG. Still, detecting daughter vesicles in the cystic lesion, separating the endocyst, and observing the membrane forming the water lily landscape are very useful in diagnosis. Observation of daughter vesicles in the cystic lesion on CT, detection of septa formations, and demonstration of plaque-like calcifications on the cyst wall is valuable in diagnosis.<sup>16,17</sup> MRI is the most helpful method among all imaging modalities in planning treatment and determining the extent of the disease.<sup>18</sup> USG, CT, and MRI modalities were used in our patients, and all of them showed the imaging features of the hydatid cyst. Although some authors use needle biopsy for diagnostic purposes, we do not recommend needle biopsy as it may cause cyst rupture and anaphylactic reaction.<sup>19</sup>

The primary treatment method in cases of muscular hydatid disease is total cyst excision. If the cyst cannot be removed entirely, the cyst should be drained, the germinative membrane wholly removed, and the cyst pouch irrigated with scolicidal solutions. A 10% recurrence rate has been reported depending on the distribution of the scolices during the surgical procedure. It has been reported that preoperative antiparasitic treatment may reduce the complications that may occur due to the distribution of the scolices during the operation and the recurrence of the disease.<sup>13</sup> Especially in the treatment of symptomatic and painful hydatid cysts, if the size is more significant than 5 cm, total surgical excision without opening the cyst is the best option.<sup>20</sup> Indeed, alongside surgical intervention, medical treatment with antihelminthic drugs like mebendazole and albendazole should be considered. Administering these drugs before and after surgery helps reduce the risk of local recurrence. Both individuals in our care were prescribed albendazole before their surgeries and continued the medication for three months following the surgical procedures. The cyst was successfully excised in both patients without rupture or complications, so no recurrence was observed during follow-up.

## Conclusion

The lungs and liver are the most common sites for hydatid cysts. Although only a limited number of cases with involvement of the paravertebral region and gluteal muscles—without lung or liver involvement—have been reported in the literature, hydatid cysts should be considered in any patient presenting with a growing mass in any organ, especially in endemic regions. Preoperative needle biopsy or cyst aspiration can lead to parasite dissemination and infection of adjacent anatomical areas. Therefore, when classical imaging findings are observed in endemic areas, any cystic lesion should raise suspicion for this disease. The treatment of choice is total excision of the cyst along with its pericystic contents.

## Conflict of Interests

The authors declare that there is not any conflict of interest regarding the publication of this manuscript.

## Ethics Committee Permission

Informed consent was obtained from the patient.

## Authors' Contributions

Concept/Design: FE, LŞ, ND. Data Collection and Processing: TC, ŞŞ. Data analysis and interpretation: TC, FE, LŞ, YSB. Literature Search: TC, ŞŞ, LŞ. Drafting manuscript: FE, TC, ŞŞ. Critical revision of the manuscript: ND, YSB. Supervision: ND, YSB.

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