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# Rare anatomical localizations of primary hydatid cysts: A single-center retrospective observational study

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## ABSTRACT

**Objective:** To describe the clinical, radiological, and surgical characteristics of primary hydatid cysts arising in uncommon anatomical locations other than the liver and lungs.

**Material and Methods:** This single-center retrospective observational study included 523 patients who underwent surgical treatment for hydatid disease at Servergazi State Hospital, Denizli, Türkiye, between January 2009 and June 2024. Patients with hepatic or pulmonary involvement, a history of previous hydatid surgery, or multi-organ disease were excluded. Demographic characteristics, clinical presentations, cyst localizations, diagnostic modalities, and treatment approaches were systematically recorded. Diagnostic evaluation was based on serological assays and imaging techniques, including ultrasonography, computed tomography, and magnetic resonance imaging.

**Results:** Of the 523 patients evaluated, 29 (5.5%) were identified as having primary hydatid cysts in atypical anatomical locations. The spleen was the most frequently involved site, followed by musculoskeletal structures, the central nervous system, and various soft tissue localizations. Serological tests were positive in 82.7% of cases. All patients underwent surgical management tailored to cyst location, and postoperative albendazole therapy was administered. Postoperative complications were limited, and no procedure-related mortality was observed.

**Conclusion:** Primary hydatid cysts may present in a wide spectrum of uncommon anatomical locations and often manifest with non-specific clinical features. Awareness of these atypical presentations is essential for accurate diagnosis and appropriate surgical management, particularly in endemic regions.

**Keywords:** Hydatid disease, cystic echinococcosis, atypical localization, extrahepatic hydatid cyst, retrospective study

## INTRODUCTION

Cystic echinococcosis (CE), caused by *Echinococcus granulosus*, remains a persistent global health concern due to its chronicity and potential for severe morbidity (1,2). Despite considerable public health efforts, the disease continues to be endemic in many regions—particularly in areas with extensive livestock farming such as South America, the Middle East, East Africa, Central Asia, and the Mediterranean basin (1,3). Türkiye is among the Mediterranean countries where CE remains a significant problem, owing to close human–animal contact and rural agricultural practices (4).

Humans become accidental intermediate hosts after ingestion of parasite eggs, with larvae typically reaching the liver via the portal circulation, making it the most commonly affected organ (75-80%) (2,5). The lungs represent the second most frequent site (5-15%) (5). When larvae bypass these primary "filter organs," hydatid cysts can arise in almost any tissue through hematogenous or lymphatic dissemination (6).

Although hepatic and pulmonary hydatid cysts account for the majority of cases, hydatid disease may occasionally present in atypical or uncommon sites, constituting approximately 3-10% of reported cases in the literature (7-9). Such locations include the spleen, kidneys, peritoneum, musculoskeletal system, heart, brain, thyroid, breast, ovaries, and bone. These unusual localizations frequently mimic other pathological entities and may therefore delay diagnosis (7-11).

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The aim of this study was to describe primary hydatid cysts identified in organs and tissues other than the liver and lungs in patients treated at a state hospital in Western Anatolia. By presenting these rare cases, we aim to contribute to the growing literature on atypical hydatid disease and emphasize the importance of considering CE in the differential diagnosis of cystic lesions in endemic regions.

## MATERIAL and METHODS

This study was designed as a single-center, retrospective observational analysis. All patients who underwent surgical treatment for hydatid disease in the surgical departments of Servergazi State Hospital, Denizli, Türkiye, between January 2009 and June 2024 were identified through the hospital's electronic medical record system and archived clinical files.

Primary hydatid cysts were defined as cystic lesions located in isolated organs or tissues without any evidence of hepatic or pulmonary involvement. Patients with concomitant liver or lung hydatid disease, a history of previous hydatid cyst surgery, or multi-organ involvement were excluded to ensure a homogeneous study population and to avoid confounding clinical characteristics.

For each eligible patient, demographic characteristics, presenting symptoms, cyst number and anatomical localization, serological test results, imaging modalities used for diagnosis, and treatment strategies were systematically recorded. Diagnostic confirmation was based on a combination of serological assays and radiological imaging, including ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI), as clinically indicated.

All available surgical specimens previously diagnosed as hydatid cysts were re-evaluated by two independent pathologists using hematoxylin-eosin-stained sections to confirm the diagnosis and ensure histopathological accuracy.

The study obtained ethical permission from the Pamukkale University Non-Interventional Clinical Research Ethics Committee (number: E-60116787-020-453576, date: 31.10.2023). This retrospective study is based on anonymized medical records and does not involve any direct patient contact or intervention. Therefore, as per the institutional ethics committee's guidance, obtaining informed consent was not necessary. The manuscript underwent linguistic editing with the assistance of artificial intelligence tools and was subsequently reviewed and approved by the authors.

### Statistical Analysis

Statistical analyses were performed using IBM SPSS Statistics for Windows, version 26.0 (IBM Corp., Armonk, NY, USA). Given the retrospective and purely descriptive nature of the study, only descriptive statistical methods were applied. Continuous variables were summarized as mean, standard deviation, and minimum-maximum values, while categorical variables were presented as frequencies and percentages. No comparative or inferential statistical analyses were performed.

## RESULTS

A total of 588 patients diagnosed with hydatid disease were initially identified through the hospital information system. Twenty-two patients with incomplete clinical data and 43 patients who were diagnosed at our institution but operated on elsewhere were excluded. The final study cohort consisted of 523 patients, including 286 females (54.7%) and 237 males (45.3%), with a mean age of 42.6 years (range: 18-89 years). More than half of the patients (55.6%) resided in rural areas.

At the time of presentation, 421 patients (80.5%) were symptomatic, whereas the remaining patients were diagnosed incidentally during radiological examinations performed for unrelated clinical indications. Presenting symptoms were categorized into broader clinical groups to improve clarity. Abdominal symptoms included abdominal pain, dyspepsia, nausea, and vomiting. Respiratory symptoms consisted of cough, dyspnea, chest pain, and hemoptysis. Neurological symptoms, such as headache and fainting, were observed in a small number of patients. Mass-related complaints, including palpable swelling in superficial or deep soft tissues, were also frequently reported.

Single-organ involvement was detected in 367 patients (70.1%), while 156 patients (29.9%) had multi-organ involvement. The distribution of organ involvement among patients with single and multiple organ disease is summarized in Table 1.

Among the entire cohort, 29 patients (5.5%) were identified as having primary hydatid cysts located outside the liver and lungs. The spleen was the most frequently involved atypical site. Other uncommon localizations included musculoskeletal tissues, the central nervous system, posterior cervical region, paraspinal area, breast, ovary, uterus, mesentery, omentum, inguinal region, and axilla. Several lesions—particularly those involving the breast, ovary, mesentery, and omentum—were detected incidentally during imaging studies performed for unrelated reasons.

Serological evaluation using the indirect hemagglutination assay yielded positive results in 82.7% of patients with atypical localizations, interpreted according to the manufacturer's recommendations. All patients underwent diagnostic imaging with US, CT, magnetic resonance imaging, or various combinations of these modalities, depending on cyst location and clinical presentation. Representative radiological findings are presented in Figures 1 and 2.

Surgical management was performed in all patients and was tailored according to cyst localization and adjacent organ

involvement. Surgical procedures included total cystectomy, pericystectomy, and organ resection when deemed necessary. Representative intraoperative images of primary hydatid cysts at atypical anatomical sites are shown in Figure 3. Histopathological examination confirmed the diagnosis in all cases, demonstrating the characteristic laminated acellular cuticular membrane and germinal layer. Representative microscopic findings from different anatomical sites are presented in Figure 4. Postoperative complications included surgical site infection, incisional hernia, hematoma, and local recurrence. One case of recurrence was observed in a patient with paraspinal involvement. No procedure-related mortality was recorded. Detailed demographic characteristics, clinical presentations, diagnostic methods, surgical approaches, and postoperative outcomes of patients with atypical primary hydatid cysts are summarized in Table 2.

All patients received postoperative albendazole therapy, with treatment duration adjusted according to cyst characteristics, surgical findings, and clinical course.

## DISCUSSION

CE remains a significant public health problem in endemic regions due to its chronic course, diagnostic challenges, and potential for serious morbidity. Although the liver and lungs function as the primary biological filters for *Echinococcus granulosus* larvae, bypass of these organs may result in cyst development in almost any tissue. Consequently, primary hydatid cysts in atypical anatomical locations, while uncommon, represent a well-recognized but diagnostically challenging clinical entity (1,2,7,10-21).

The proportion of patients with primary extrahepatic and extrapulmonary hydatid cysts in the present series is consistent with rates reported in the literature. Previous studies from endemic regions have demonstrated that such atypical localizations account for a small but clinically relevant subset of cases (1,13-15,22). These lesions often mimic neoplastic, inflammatory, or congenital cystic conditions, leading to delayed diagnosis or inappropriate initial management, particularly when hydatid disease is not considered in the differential diagnosis (9,10,16-25).

Several mechanisms have been proposed to explain the occurrence of hydatid cysts in unusual anatomical locations. These include hematogenous dissemination after bypassing the hepatic and pulmonary filters, lymphatic spread, and, in selected cases, transdiaphragmatic migration (6,25-39). These pathways may account for the wide spectrum of anatomical involvement observed in primary atypical hydatid disease, including musculoskeletal, central nervous system, and pelvic localizations (16-18).

Splenic involvement represented the most frequent atypical localization in our cohort, in line with previous reports (16,18,21). Despite being among the more frequently reported extrahepatic sites, isolated splenic hydatid cysts remain rare

**Table 1. Distribution of single and multiple organ involvement in hydatid disease**

| **Single organ involvement (n=367)** | | **Multi-organ involvement (n=156)** | |
|---|---|---|---|
| **% (n)** | **Organs** | **% (n)** | **Organs** |
| 68.6 (252) | Liver | 50.6 (79) | |
| 23.4 (86) | Lung | 3.2 (5) | Liver and kidney |
| 2.4 (9) | Spleen | 4.4 (7) | Liver and mesentery |
| 0.5 (2) | Neck | 5.1 (8) | Liver and spleen |
| 0.2 (1) | Brain | 1.9 (3) | Liver, lung, and spleen |
| 0.5 (2) | Inguinal area | 3.2 (5) | Liver, mesentery, and spleen |
| 0.8 (3) | Gluteal region | 7 (11) | Liver and omentum |
| 0.2 (1) | Uterus | 5.1 (8) | Liver, spleen, and omentum |
| 0.2 (1) | Paraspinal region | 1.9 (3) | Lung, spleen, and omentum |
| 0.2 (1) | Ovary | 17.3 (27) | Others |
| 0.2 (1) | Omentum | | |
| 0.8 (3) | Thigh | | |
| 0.2 (1) | Axilla | | |
| 0.2 (1) | Mesentery | | |
| 0.2 (1) | Cervical area | | |
| 0.5 (2) | Breast | | |

(19,23). Importantly, there is no universally accepted standard treatment algorithm for splenic hydatid disease. Management strategies vary depending on cyst size, location, presence of complications, and institutional or surgeon experience (19,23). While spleen-preserving techniques and percutaneous approaches such as puncture-aspiration-injection-reaspiration have been described in selected cases, total splenectomy remains a commonly employed option, particularly for large, centrally located, or complicated cysts, owing to concerns regarding rupture, recurrence, and intraoperative dissemination (19,24,25).

Musculoskeletal and subcutaneous hydatid cysts constitute another diagnostically challenging group, largely due to their rarity and non-specific clinical presentation. Reported incidence remains low even in endemic regions (11,30-32). When present, these cysts frequently manifest as slowly enlarging, painless masses and may be misdiagnosed as soft tissue tumors (31-35). Imaging—particularly MRI—plays a crucial role in preoperative assessment, while complete surgical excision remains the treatment of choice to minimize recurrence (30,32,34,36).

Central nervous system involvement is among the rarest manifestations of CE but carries substantial clinical importance due to the potential for severe neurological sequelae (26-29). Radiological findings are often characteristic, facilitating preoperative diagnosis (26-29). Surgical excision using techniques that minimize cyst rupture remains the cornerstone of management, supplemented by postoperative anthelmintic therapy (26-28).

No cases of orbital or intraocular hydatid cysts were identified during the study period, which may be partly attributable to the fact that such cases are more commonly managed and followed

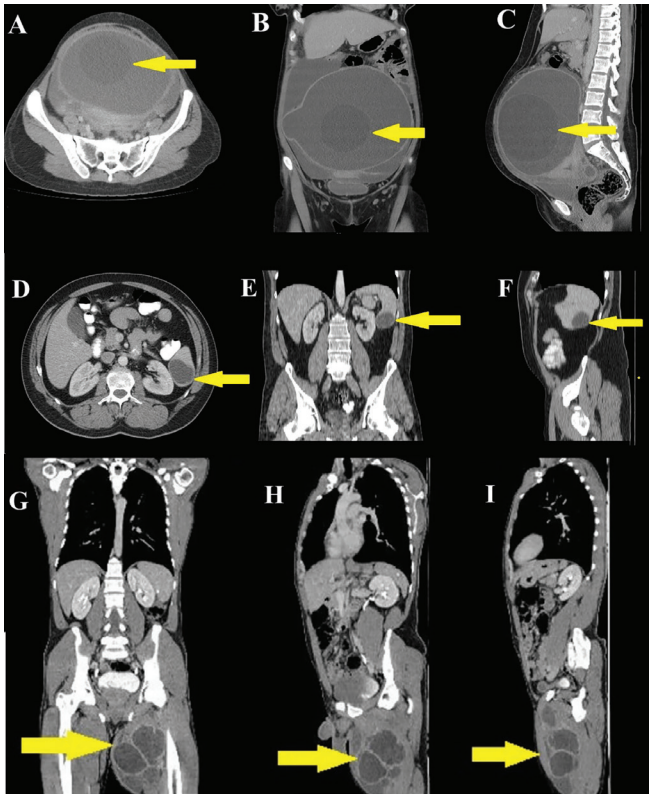


**Figure 1.** Computed tomography images of primary hydatid cysts in atypical anatomical locations.

(A-C) Large cystic lesion in the uterus shown in axial, coronal, and sagittal planes.

(D-F) Cystic lesion of the spleen demonstrated in axial, coronal, and sagittal planes.

(G-I) Multiloculated cystic lesion in the left thigh shown in coronal, oblique, and sagittal planes.

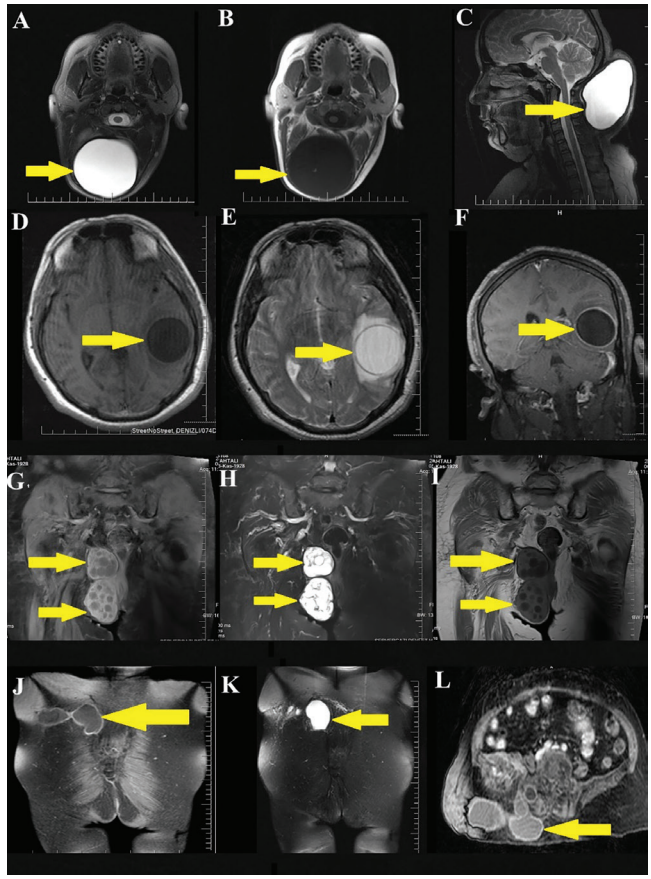


**Figure 2.** Magnetic resonance imaging findings of primary hydatid cysts in atypical localizations.

(A-C) Cystic lesion in the posterior cervical region on T2-weighted axial, T1-weighted axial, and T2-weighted sagittal images.

(D-F) Cystic lesion adjacent to the central sulcus in the right parietal region, hypointense on T1-weighted and hyperintense on T2-weighted images.

(G-I) Multiloculated cystic lesions in the pelvic region and right lateral rectum on T1- and T2-weighted coronal images.

(J-L) Cystic lesion in the right lateral sacrum demonstrated on T1- and T2-weighted coronal and T1-weighted axial images.

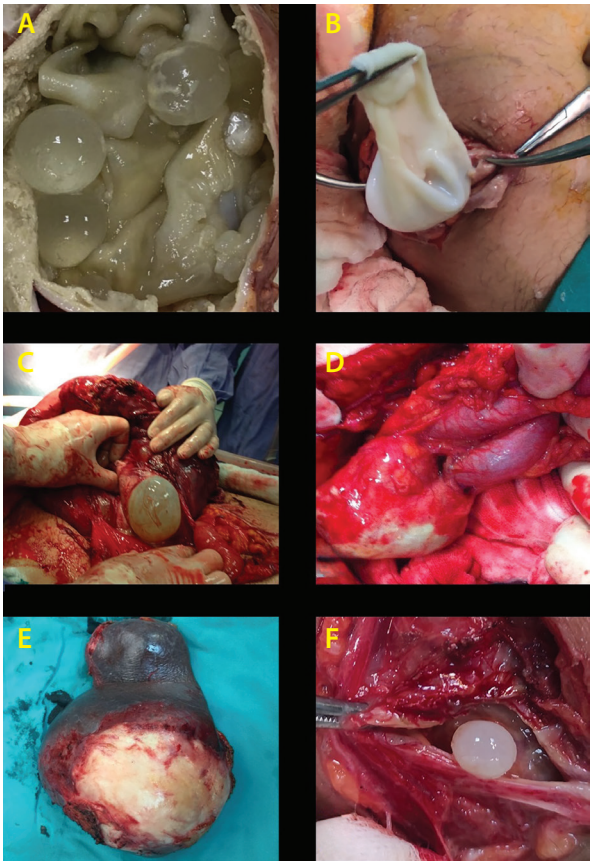


**Figure 3.** Intraoperative views of primary hydatid cysts in atypical anatomical sites.

(A) Multiple cysts in the gluteal region, (B) Hydatid cyst of the thigh, (C) Intrauterine hydatid cyst, (D) Omental hydatid cyst, (E) Splenic hydatid cyst, (F) Inguinal hydatid cyst.

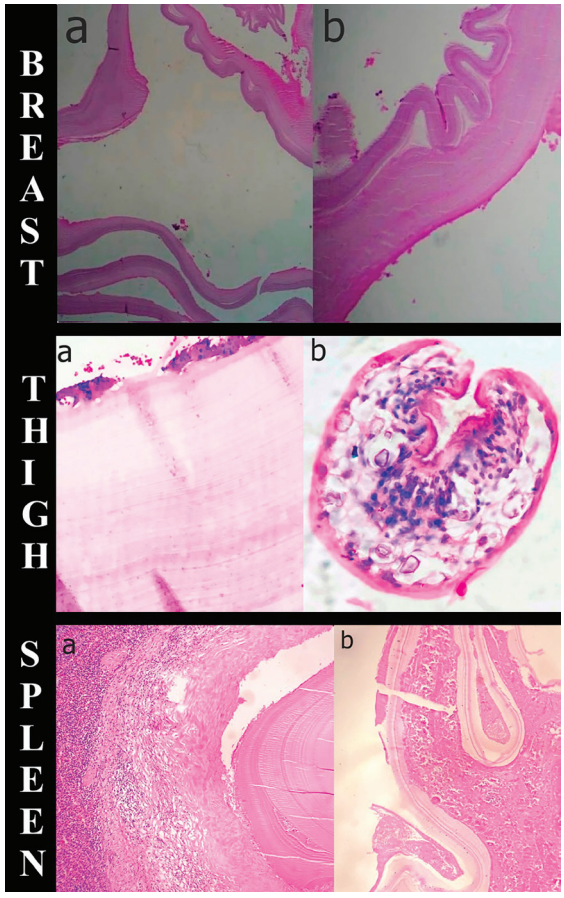


**Figure 4.** Histopathological features of hydatid cysts.

Laminated acellular cuticular membrane and germinal layer in a breast hydatid cyst (H&E ×20, ×40). Laminated membrane and scolices in a thigh hydatid cyst (H&E ×40, ×100). Laminated acellular cuticular membrane adjacent to normal splenic tissue in a splenic hydatid cyst (H&E ×40).

H&E: Hematoxylin&eosin.

by ophthalmology departments rather than general surgery.

Primary peritoneal, omental, and mesenteric hydatid cysts—as well as pelvic involvement—are exceptionally rare and are often discovered incidentally (37-39). These lesions are thought to arise predominantly through hematogenous or lymphatic dissemination (37-39). Given their non-specific presentation and radiological resemblance to intra-abdominal neoplasms, a high index of suspicion is required, particularly in endemic regions (37,38-44). Female genital tract involvement is also rare and may mimic gynecological cystic tumors; surgical excision remains the preferred approach when feasible (45-50). Breast involvement is uncommon even in endemic settings and may resemble benign breast lesions; diagnosis relies on imaging and clinical suspicion, with surgery as standard treatment (40-43).

### Study Limitations

The retrospective design and single-center nature of this study represent inherent limitations. However, the strengths of the present study include a relatively large overall cohort, a long study period, and histopathological confirmation of all surgically treated cases, providing a comprehensive overview of primary hydatid cysts in atypical anatomical locations within an endemic region.

## CONCLUSION

Hydatid disease continues to pose a notable health problem in endemic regions, and its presentation beyond the liver and lungs should not be overlooked. Primary hydatid cysts in atypical anatomical sites may mimic a wide range of clinical conditions, often leading to diagnostic delay. Our findings underscore the need for heightened clinical suspicion, particularly in patients presenting with unexplained cystic lesions. Surgical excision, when performed meticulously to avoid intraoperative rupture, remains the most effective treatment strategy, and the addition of perioperative albendazole contributes to reducing recurrence risk. Ultimately, recognizing the diverse clinical spectrum of CE is essential for timely diagnosis, optimal management, and improved patient outcomes.

**Table 2. Demographic characteristics, clinical presentation, diagnostic methods, management strategies, and outcomes of patients with primary hydatid cysts in atypical locations**

| Case no | Age | Gender | Location | Clinical features | Serology (IHA) | Imaging methods | Surgical management | Complications | Chemotherapy |
|---|---|---|---|---|---|---|---|---|---|
| 1 | 71 | Female | Spleen | Abdominal pain | + | US-CT | Splenectomy | Hematoma | Three months adjuvant therapy |
| 2 | 42 | Male | Spleen | Left hypochondriac pain | + | US-CT | Splenectomy | - | Three months adjuvant therapy |
| 3 | 54 | Female | Spleen | Abdominal pain | + | US | Splenectomy | Surgical field infection | Three months adjuvant therapy |
| 4 | 26 | Male | Spleen | Incidental finding during US | + | US | Splenectomy | - | Three months adjuvant therapy |
| 5 | 59 | Female | Spleen | Left hypochondriac pain | + | US-CT | Splenectomy | - | Three months adjuvant therapy |
| 6 | 57 | Male | Spleen | Abdominal pain | + | US | Splenectomy | - | Three months adjuvant therapy |
| 7 | 61 | Female | Spleen | Abdominal pain | - | US-CT | Splenectomy | - | Three months adjuvant therapy |
| 8 | 66 | Male | Spleen | Incidental finding during US | + | US-CT | Splenectomy | Incisional hernia | Three months adjuvant therapy |
| 9 | 39 | Female | Spleen | Abdominal pain | - | US-CT | Splenectomy | - | Three months adjuvant therapy |
| 10 | 30 | Male | Neck | Palpable swelling on the right side of the neck | + | US | Total cystectomy | - | Six weeks adjuvant therapy |
| 11 | 54 | Female | Neck | Palpable swelling on the left side of the neck | + | US | Total cystectomy | - | Six weeks adjuvant therapy |
| 12 | 63 | Male | Inguinal | Tender mass in the right inguinal region | - | US | Total cystectomy | surgical field infection | Six weeks adjuvant therapy |
| 13 | 84 | Female | Inguinal | A swelling in the right inguinal region | + | US | Total cystectomy | - | Six weeks adjuvant therapy |
| 14 | 60 | Female | Gluteal | Right hip pain | + | US-MRI | Total cystectomy with muscle excision | - | Two months adjuvant therapy |
| 15 | 89 | Male | Gluteal | Right hip pain and a swelling | + | US-MRI | Total cystectomy with muscle excision | - | Two months adjuvant therapy |
| 16 | 59 | Female | Gluteal | Left hip pain and a swelling | + | MRI | Total cystectomy | - | Three months adjuvant therapy |
| 17 | 82 | Female | Paraspinal area | Lumbalji | + | CT-MRI | Total pericystectomy | recurrence | Six months adjuvant therapy |
| 18 | 39 | Female | Ovary | Pelvic pain | + | US-CT-MRI | Pericystectomy | - | Three months adjuvant therapy |
| 19 | 52 | Female | Mesenter | Incidental finding during US | - | US-CT | Pericystectomy | incisional hernia | Four months adjuvant therapy |
| 20 | 28 | Female | Uterus | Pelvic pain | + | US-CT | Total hysterectomy | - | Six months adjuvant therapy |
| 21 | 47 | Male | Omentum | Incidental finding during US | + | US-CT | Total cystectomy | Surgical field infection | Three months adjuvant therapy |
| 22 | 35 | Female | Breast | Incidental finding during US | + | US | Total cystectomy | - | Six weeks adjuvant therapy |

**Table 2. Continued**

| Case no | Age | Gender | Location | Clinical features | Serology (IHA) | Imaging methods | Surgical management | Complications | Chemotherapy |
|---|---|---|---|---|---|---|---|---|---|
| 23 | 31 | Female | Breast | Incidental finding during US | + | US | Total cystectomy | - | Six weeks adjuvant therapy |
| 24 | 32 | Female | Thigh | A swelling in right thigh | + | US-CT | Total cystectomy | - | Two months adjuvant therapy |
| 25 | 23 | Female | Thigh | Left thigh pain | - | US-MRI | Total cystectomy | Surgical field infection | Two months adjuvant therapy |
| 26 | 60 | Female | Thigh | A swelling in right thigh | + | US-MRI | Total cystectomy | - | Two months adjuvant therapy |
| 27 | 82 | Female | Cervical area | Neck pain | + | US-MRI | Pericystectomy | - | Six weeks adjuvant therapy |
| 28 | 42 | Female | Axilla | A swelling | + | US | Total cystectomy | - | Six weeks adjuvant therapy |
| 29 | 54 | Male | Brain | Headache and fainting | + | CT-MRI | Total cystectomy | - | Four months adjuvant therapy |

US: Ultrasonography, CT: Computed tomography, MRI: Magnetic resonance imaging, IHA: Indirect hemagglutination test.

### Ethics

**Ethics Committee Approval:** The study obtained ethical permission from the Pamukkale University Non-Interventional Clinical Research Ethics Committee (number: E-60116787-020-453576, date: 31.10.2023).

**Informed Consent:** This retrospective study is based on anonymized medical records and does not involve any direct patient contact or intervention. Therefore, as per the institutional ethics committee's guidance, obtaining informed consent was not necessary.

### Footnotes

#### Author Contributions

Surgical and Medical Practices - R.S.A., R.B., S.T.Ş., A.E.Y.; Concept - R.S.A., R.B.; Design - R.S.A.; Data Collection or Processing - R.B., Ş.K., Y.S.K., A.E.Y.; Analysis or Interpretation - R.B.; Literature Search - R.B., S.T.Ş.; Writing - R.S.A., R.B.

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