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Giant Cyst with Unclear Positioning and Undefined Pathological Diagnosis

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

This is the report of a right-sided giant cystic lesion of a young woman without symptoms which was diagnosed incidentally during routine health screening before marriage. All the routine blood tests and chest x-ray were normal. Abdominal ultrasound (US) revealed a type-3 hydatic cyst in the right liver lobe, but serum tests were negative. On the contrary, Magnetic Resonance Imaging (MRI) revealed a "retroperitoneal cyst" adjacent to the right upper pole of the kidney rising upwards by making a massive bulge at the posterior part of the right lobe of the liver, displacing and rotating the right lobe through the diaphragm by neighboring liver segments of VI, VII, and VIII. At the operation, it was unexpectedly discovered that none of the abdominal US and MRI radiologic reports were wholly true or false! As a well-known fact, without any exception of its anatomical structures, a normal liver is located completely intraperitoneal part of the abdominal cavity. This case is the first where we could not find another in the searched literature that the upper inferior part of the right lobe of the liver was located "partially" in the retroperitoneal area. As there was no trauma history of the patient, this might be a congenital malformation affecting both the right liver lobe and right side of the diaphragm in which a giant fully retroperitoneal cyst was originated. All of the radiologic, surgical, and postoperative pathological diagnostic uncertainty and confusion is caused by this malformation.

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Keywords: Hepatic cyst; giant cyst; idiopathic cyst.

1. INTRODUCTION

Retroperitoneal cysts located on the right side adjacent to the liver mimicking liver cysts and cysts originating truly from the liver mimicking retroperitoneal cysts could be challenging for both the radiologists and pathologists to diagnose and so for the clinicians to treat. Here we report a giant right-sided cystic lesion of a young woman without symptoms which was diagnosed incidentally during routine health screening before marriage. The cystic lesion was not mimicking but truly retroperitoneal and genuinely originating from the liver simultaneously. Furthermore, there was no trauma history of the patient. Without any exception of its anatomical structures, a normal liver is located entirely in the intraperitoneal compartment of the abdominal cavity, which is a well-known fact. This case is the first where we could not find another in the searched literature that the inferior part of the right lobe of the liver was located in the retroperitoneal area where a giant, entirely retroperitoneal cyst was originated from and, also from the lower part of the diaphragm adjacent to the right triangular ligament of the liver and which was all the cause of the radiologic, surgical and postoperative pathological diagnostic uncertainty and confusion.

This case is the first case that we could not find in the literature, in which the lower part of the right lobe of the liver is located in the retroperitoneal region and a giant, completely retroperitoneal cyst originates from the lower part.

2. PRESENTATION OF CASE

A 22 years old female patient referred to our clinic with the complaint of a giant abdominal mass originating from the liver, which was detected incidentally during routine health screening before marriage. Abdominal ultrasound revealed a 171x135 mm cystic mass in the right liver lobe compatible with a type-3 hydatic cyst. As the echinococcus indirect hemagglutination (IHA) test was negative, a further Magnetic Resonance Imaging (MRI) was needed to clarify the situation. After the MRI test, a “retroperitoneal cyst” adjacent to the upper pole of the right kidney rising upwards by making a massive bulge at the posterior part of the right lobe of the liver, displacing and rotating the right lobe of the liver through the diaphragm by neighboring liver segments of VI, VII, and VIII compatible with a type-3 hydatic cyst was reported (Figs. 1, 2).

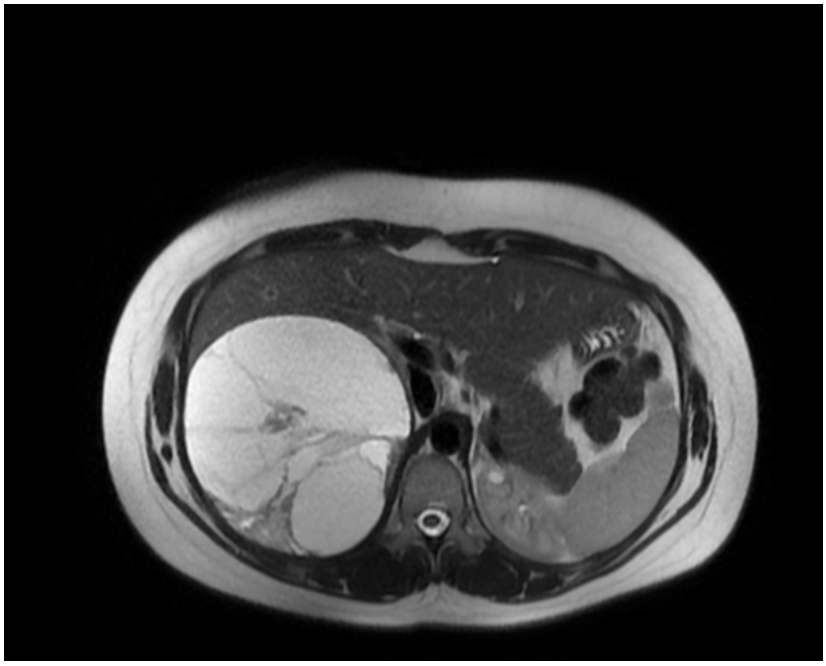

Fig. 1. Abdominal Magnetic resonance (MR) imaging (Preoperative)

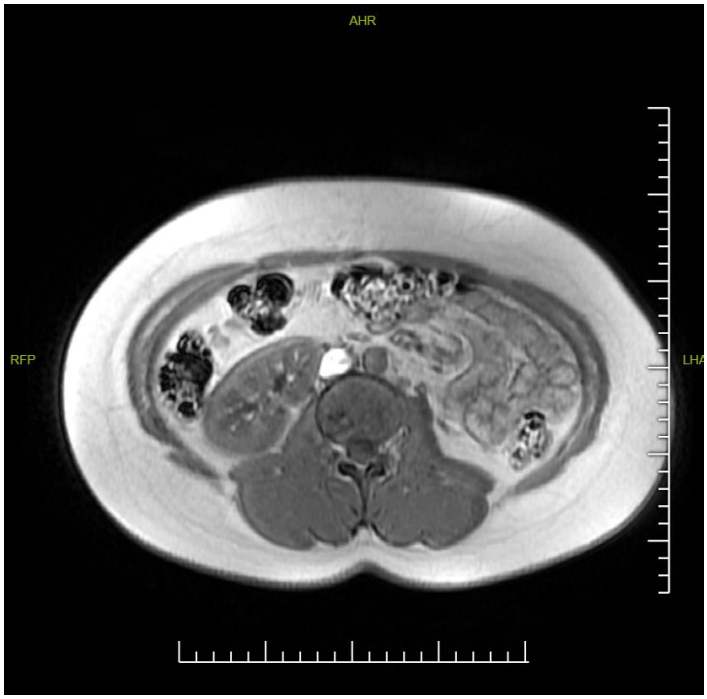

Fig. 2. Abdominal Magnetic resonance (MR) imaging (Preoperative)

All preoperative routine laboratory tests and chest X-ray were normal. Because of the diagnostic uncertainty in the imaging modalities and serum echinococcus IHA test, surgery was required for complete excision of the cyst for a definitive diagnosis of the disease and to cure it. The abdomen was entered with a midline incision to under general anesthesia where a giant cyst located entirely in the retroperitoneum was explored in both inspection and palpation at exploration, the operative team got the impression that the huge cyst had no connection with the liver. After making a huge retroperitoneal bulge in front of the right lobe of the liver, the impression of the cyst was that it elevated this liver part through the diaphragm and has hidden under it (Fig. 3).

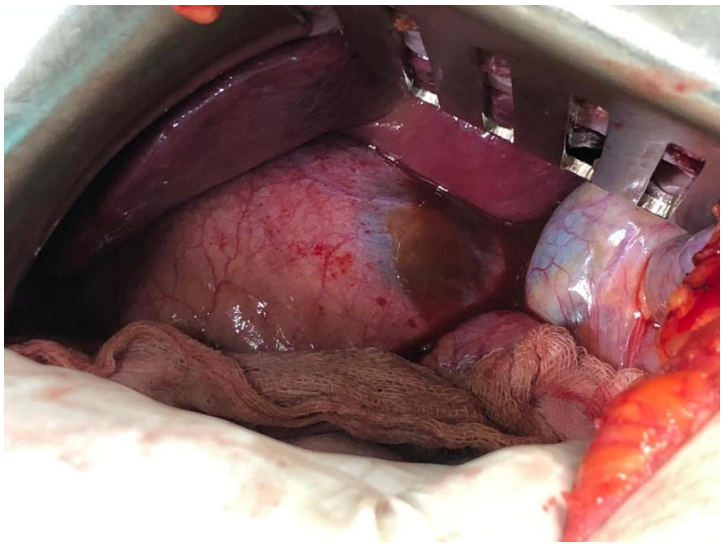

Fig. 3. Retroperitoneal view of the cyst - surgical view

By the intention of complete excision of the cyst retroperitoneal compartment was entered by sharp dissection, and the cyst was easily dissected from the upper pole of the right kidney. When the dissection was carried upwards, the cyst was found to have a tense contact with the right adrenal gland with an impression that it was originating from it. Right adrenalectomy was performed for complete excision of the cyst, and sharp dissection was continued upwards through the diaphragm, where the cyst was dissected easily from the vena cava medially and abdominal wall laterally by using a ligasure sealing device. On the contrary, when the dissection plane reached the diaphragm, it was observed that the cyst invaded the lateral lower part of the diaphragm. The invaded part was excised in order to perform a complete cyst excision. After performing partial diaphragmatic excision, the cyst was tried to be taken out of the abdominal cavity by placing the hand under it. However, this process could not be performed successfully because the right liver lobe moved together with the huge cyst. Then, in order to dissect tense soft tissue attachments between the cyst and liver, the superior anterior part of the peritoneum, which was between the right liver lobe and the massive bulge of the cyst, was incised from the peritoneal part of the abdominal cavity. However, unexpectedly, a dissection plane between the cyst and the liver could not be obtained. At this point of the operation, it was observed that this entire retroperitoneal cyst originated from the lower part of the right liver lobe. For complete excision, liver segments 4b, 5, 6 and 7 had to be excised. There was an unexpected and unusual situation for the operative team as the entirely retroperitoneal located giant cyst was not invading but originating from the liver and diaphragm. The need for the excision for such huge liver tissue segments was questionable as the patient was too young to expect a malignancy, and still, there was uncertainty about the diagnosis. Partial excision of the cyst called marsupialization was decided, and hepatectomy was postponed to a second operation in case of a need after the postoperative pathological diagnosis. In case of an unusual presentation of a hydatid cystic disease, 3% saline water impregnated operative towels were placed around the cyst to prevent infestation during cystic fluid aspiration. After aspirating 50cc of cystic fluid that seemed to contain bile, 50cc of 3% saline water was injected into the cyst and waited for 20 minutes to kill the possible cestodes. After the entire aspiration of the cystic fluid, the extrahepatic cystic wall was incised near to the liver, and the cyst was excised partially by leaving the liver parts of it by using a ligasure device where a few bile duct openings were seen and sutured. Also, the diaphragmatic opening was sutured by mattress sutures, and a chest tube was inserted. After placing a soft drain into the cystic cavity, the abdominal wall was sutured anatomically, and the operation was ended. The postoperative period was uneventful, and the patient was discharged on the fifth postoperative day. Pathologic report of the cyst was not a hydatic disease. The pathological report revealed that the cystic wall was also neighboring but not originating from the right adrenal gland. There was no sign of malignancy, but a definitive diagnosis of the cyst could not be obtained pathologically.

Since the malignancy risk of the cyst was not known, the patient was taken to the follow-up program with radiological and biochemical examinations every 6 months for the first 2 years and yearly thereafter. Postoperative visits in the outpatient on the 6^{th}, 12^{th} mounts revealed no recurrence of cyst by ultrasonography.

3. DISCUSSION AND CONCLUSION

Although radiologic imaging techniques are sensitive in detecting hydatid cysts, hydatid disease demonstrates various imaging features that vary according to growth stage, associated complications, and the affected tissue [1,2]. Ultrasound is the first diagnostic technique for hepatic HD, and no other imaging techniques are requested when the appearance is typical [3]. Although abdominal CT, with its higher rate of accuracy, is the diagnostic tool of choice, due to its multiplanar capabilities and the excellent contrast resolutionfor soft tissues, MRI has particular importance if the diagnosis of HD is questionable as it is more accurate in demonstrating parietal features and defining anatomical relationships. However, with atypical imaging findings, hydatid cysts in an unusual location or of an unusual dimension may complicate the differential diagnosis. The retroperitoneal hydatid cyst is a rare entity even in endemic areas [4]. Simple cysts are more prevalent in women. A definitive role for open surgery technique in selected patients is indicated especially in giant cysts that had taken up most of the abdomen, and displaced other organs [5].

Furthermore, retroperitoneal cystic masses have an important diagnostic and therapeutic

challenge as differential diagnosis include a large variety of neoplastic lesions (cystic lymphangioma, mucinous cystadenoma, cystic teratoma, cystic mesothelioma, müllerian cyst, epidermoid cyst, tailgut cyst, bronchogenic cyst, cystic change in solid neoplasms, pseudomyxoma retroperitonei) and non-neoplastic lesions (pancreatic pseudocyst, non-pancreatic pseudocyst, lymphocele, urinoma, hematoma) [6]. For this reason, a definitive diagnosis requires a combination of imaging, serologic and immunologic studies. Sometimes, even pathology reports are insufficient in making an accurate diagnosis.

In our case, ultrasonography reported a cystic mass confined to the right liver lobe compatible with a type-3 hydatic cyst. However, as serological test IHA was negative for hydatid disease, MRI is needed to identify the lesion. MRI concluded a “retroperitoneal cyst” adjacent to the upper pole of the right kidney rising upwards by making a massive bulge at the posterior part of the right lobe of the liver, displacing and rotating the right lobe through the diaphragm. The cyst was not originating from the right lobe of the liver.

The treatment options for hydatid cysts of the liver depend on stage, localization, size, and complications of the cysts and include nonoperative and operative methods [7]. Operative methods include classical surgical techniques (total or subtotal cyst-pericystectomy, partial hepatectomy, capsulorrhaphy, capitonnage, omentoplasty); minimally invasive techniques, such as laparoscopic or robotic procedures; and other treatment modalities, such as puncture, aspiration, injection, and respiration of scolicidal solutions. Chemotherapy alone with the benzimidazole family has limited efficacy [8,9,10].

In our case, despite the use of the broad diagnostic armamentarium, the cystic lesion could not be accurately classified. As a giant cyst at risk of rupture in pleural and/or abdominal cavities is one of the surgery indications, we decided to operate on the patient. It is recommended that laparoscopic fenestration should be used in easily accessible areas, superficial cysts especially located in the anteriorly or laterally of the abdomen, and that the traditional open surgery should be used in potential malignant cases where the cysts are posterior or are located near big vascular structures [11]. This case was not suitable for minimally invasive techniques, so open surgery was preferred to make an accurate diagnosis and treatment. Unroofing is associated with a high recurrence rate (>20%) [12]. Surgical cystectomy is the treatment of choice for large deep seated cysts [13]. The estimated disease was a giant liver seronegative hydatic cyst or a giant retroperitoneal seronegative hydatic disease originating from one of the retroperitoneal structures, most probably the right adrenal gland. In order to cure the patient, complete excision of the cyst was decided. Therefore, right adrenalectomy was carried out together with partial diaphragmatic excision as the cyst was found to invade the lateral lower part of the diaphragm. In the dissection plane, it was observed that the cyst unexpectedly originated from the lower part of the right liver lobe. In order to perform a complete excision, liver segments 4b,5, 6 and 7 had to be excised. At this point of the operation, the need for the excision for such a huge liver tissue was questionable as the patient was too young to expect a malignancy, and still, there was uncertainty about the diagnosis. Therefore, partial excision of the cyst was the preferred technique. Postoperative pathologic examination was inadequate to determine the exact diagnosis. There was no sign of malignancy. It was not a hydatid disease and was not originating from the right adrenal gland either.

This case is the first where we could not find another in the searched literature that the upper inferior part of the right lobe of the liver was located “partially” in the retroperitoneal area. As there was no trauma history of the patient, this might be a congenital malformation affecting both the right liver lobe and right side of the diaphragm in which a giant fully retroperitoneal cyst was originated, and it was all the cause of the radiologic, surgical, and postoperative pathological diagnostic uncertainty and confusion. In the present case, even the pathologic examination could not adequately diagnose the lesion.

Diagnostic uncertainty is a disturbance for a surgeon. The surgery decision is then determined by the presence of symptoms, the concern about future complications like rupture, infection, the possibility of malignancy or malignant change. We think this cystic mass overlapped a possible congenital anatomic malformation and is the first in the literature.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

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

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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