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Case Report

Recurrence of retroperitoneal mature cystic teratoma in an adult: A case report

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ABSTRACT

Mature cystic teratoma is one of the most common tumors of the ovaries, testis, mediastinum, and retroperitoneum; however, secondary retroperitoneum lesions are rare entities in adults. We report a case of a 22-year-old female who was previously diagnosed with a mature teratoma of left ovarian was hospitalized due to dull abdominal pain in right hypochondria. Radiological evaluation revealed a mass in the right upper abdominal and flank region with an extension from the posterior aspect of the duodenum, composed of greasy and cystic elements. A tumor was resected through the Kocher's laparotomy and the pathology report confirmed the diagnosis of a mature cystic teratoma with no evidence of malignancy or immature components.

Introduction

Teratomas are uncommon neoplasms comprised of a mixture of dermal cells derived from the 3 germ cell layers (ectoderm, mesoderm, or endoderm), mature teratomas are rare neoplasms characterized by inclusion of any well-differentiated parenchymal tissues [1,2]. These tumors are most commonly found in testes and ovaries but extragonadal sites have also been reported including intracranial, cervical, mediastinal, retroperitoneal, and sacrococcygeal [3–5]. Accounting for only 4% of all teratomas, retroperitoneal lesions are rare and more common among children rather than adults [2,6]. We report a case of a large symptomatic secondary retroperitoneal mature cystic teratoma in a 22-year-old woman.

Case report

A 22-year-old woman presented with right hypochondria pain. Four years ago, she underwent surgery to remove the left ovary dermatome cysts. She began to feel uncomfortable and aching in the lower right quadrant for about 3 months after she first palpated the lesion 2 and 3 months ago. Recently, she noticed an increase in the size of the mass and referred to the clinic. She denied any associated symptoms including fever, loss of

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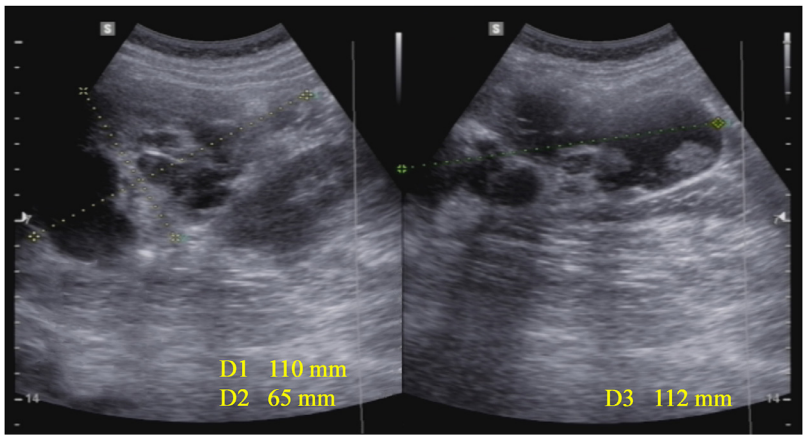

Fig. 1 – Mature cystic teratoma (dermoid) in a 22-year-old woman in whom symptomatic right hypochondria mass was palpated at a routine physical examination. Transverse transabdominal ultrasonography image showed a cystic structure (calipers) of 112 x 110 x 65 mm dimensions in the middle of the liver and right kidney. It was heterogeneous, well circumscribed with irregular borders.

appetite, weight loss, nausea, and vomiting. She denied taking any medications, smoking or alcohol consumption and her family history was unremarkable.

Her weight is 53 kg, gravidarum 0, parity 0. She was normotensive with a blood pressure of 110/60 mmHg, her pulse rate was 72 per minute, her respiratory rate was 18 per minute and her temperature was 37.5°C. Her physical examination was unremarkable except for a firm, nonmobile fullness palpated in her right upper quadrant region without any tenderness or abdominal guarding. The extent of the mass could not be established.

Laboratory results showed that Ht, Hb, PLT, WBC, Glu, Protein, Urea, Cr, Na, K, and bHCG have fluctuated in normal ranges. Tumor markers AFP, CA 15-3, CA 19-9, CA 125, and Carcinoembryonic antigen (CEA) have fluctuated in normal ranges. Serum levels of HBsAg were significantly elevated reaching the plateau of 5122 ng/mL (normal ranges: <0.9 ng/mL). Except that several urine parameters include urine-specific gravity (1.015), pH (9), protein (25 mg/dL), leukocytes (500 LEU/uL), and erythrocytes (25 Ery/uL) were mildly changed. Ultrasonography showed a mass of 112 × 110 × 65 mm dimensions in the middle of the liver and right kidney. It was heterogeneous, well circumscribed with irregular borders. The mass did not appear to arise from the right kidney. The mass was adherent to the segment VI of the right lobe of the liver. The mass has been described as a multilocular mixture lesion. Doppler interrogated images were also obtained and revealed minimal internal vascularity (Fig. 1).

A computerized tomography (CT) scan of the abdomen and pelvis with contrast for the patient that revealed a large retroperitoneal mass lying in the middle of the liver and right kidney measuring about 120 × 80 × 65 mm in diameter. It consisted of both cystic and solid elements, showing fat density but not seen calcification density. No abnormal enhancement was identified. The mass did not invade surrounding organs (such as the liver, the right kidney, the right adrenal glands). There was no retroperitoneal lymphadenopathy, ascites. The mass fixed to the broad clip of the old surgery (Fig. 2).

Magnetic resonance imaging of upper and lower abdomen has also been performed. Similarly to ultrasonography and CT scan report, it also shows large multilocular cystic spaces mass with mild enhancing a smooth wall and thin septations as well as a little nonenhancing solid contents. The cystic has been hemorrhagic heterogeneous signal intensity in locales with many different phases. Note the absence of detectable calcification or matted tuft of hair in mass (Figs. 3 and 4).

Laparotomy with excision of the retroperitoneal mass was performed by the right subcostal line (Kocher's laparotomy). A huge retroperitoneal mass was observed. The surgeon found a tumor section stick to the surface of the liver and the stem of mass is derived from the retroperitoneal. Subsequently, complete removal of the mass from the liver surface was successful, and adhesions were released and the tumor was separated from the surrounding tissue. No ascites were observed and based on the findings a decision was made for total excision of a tumor. Finally, a 12 × 8 × 6.5 cm mass was totally excised and sent to the lab for pathologic evaluations. The mass had well-circumscribed smooth borders and rubbery consistency. Cut-section of the mass revealed multilocular cystic spaces, whitish-grey walls, scattered yellowish adipose tissue collections, and mucus secretions. The cyst wall was up to 4-mm thick (Fig. 5).

Histopathologic examination shows that within a neoplasm composed of various tissues including epithelial tissue, fat, cartilage tissue mixed with fibrous tissue, and muscle tissue along with vast areas indicative of previous bleeding with aggregation of hemosiderin. The final diagnosis was made as a mature cystic teratoma with no evidence of malignancy or immature components (Fig. 6).

The patient was followed 2 weeks after the surgery, during which she developed no significant complications.

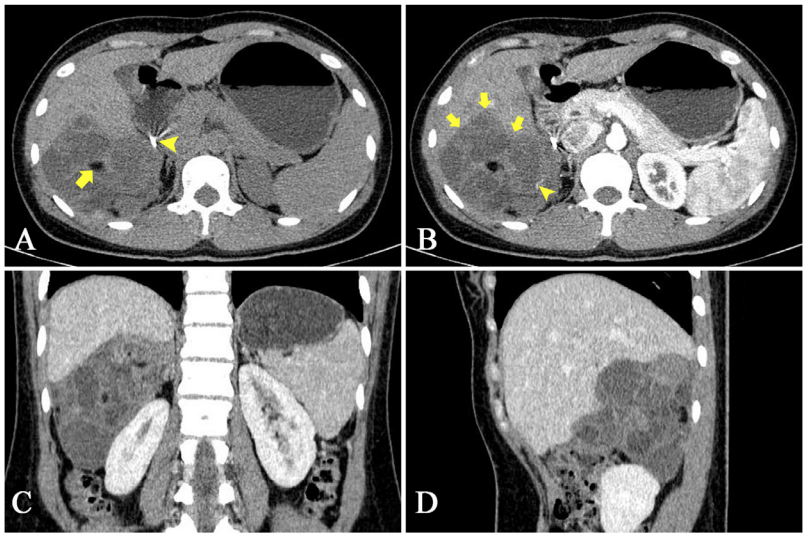

Fig. 2 – CT scan through the abdominal shows a mature cystic teratoma in the right hypochondria. (A) On the non–enhanced CT axial plane shows it contains fat (arrow). Presence next to a tumor has a clip of the old surgery lost seats (arrowhead). (B) The enhanced axial CT in arterial phase shows bound of the mass is not clear with a liver (arrows). The lesion shows with mild hypervascular (arrowhead). (C, D) The tumor on the sagittal and coronal images in late portal phase.

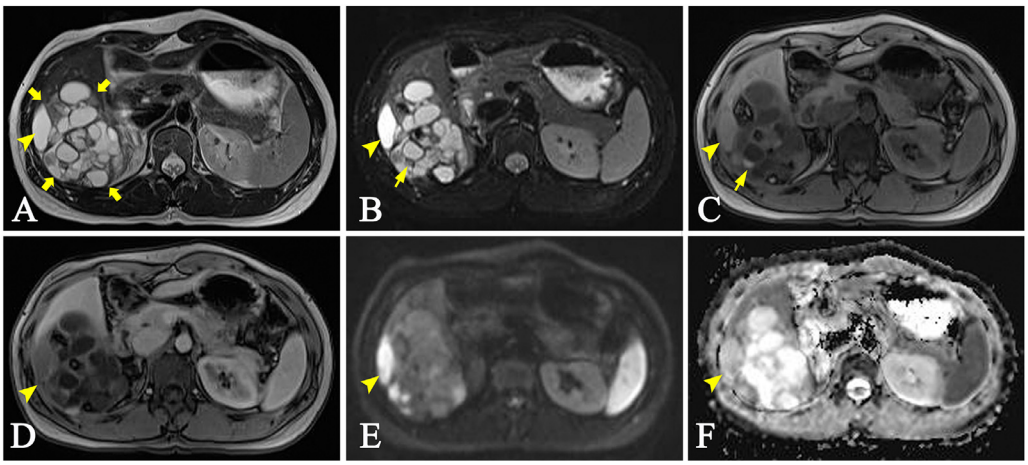

Fig. 3 – MRI of a tumor in the axial plane on T2 TSE sequence (A), T2 TSE FS (B), non–enhanced T1 Vibe Dixon sequence (C), enhanced T1 Vibe Dixon in late arterial phase (D), Diffusion weighted imaging (DWI) (E), and Apparent diffusion coefficient (ADC) (F). The mass shows a multilocular lesion (arrows in (A)). A part of the lesion (arrowhead) increased signal on T2W, increased slightly on T1W, and increased signal on DWI and ADC (T2 shine-through effect). It also shows the signal intensity of hemorrhage or fat (thin arrows in (B) and (C)).

Discussion

Teratomas are a common form of germ cell tumor, containing all 3 germ cell layers. Teratomas are classified as mature or immature, depending on the degree of differentiation of its components [7]. Retroperitoneal teratomas are uncommon in the adult population. Primary retroperitoneal teratomas represent 1%-11% of the retroperitoneal neoplasms [1], but secondary retroperitoneal teratomas are rarer and most adult cases are in females. There are usually asymptomatic but can cause nonspecific abdominal disorders including pain, nausea, and vomiting. In the majority of cases, they present as asymptomatic, making the diagnosis at an earlier stage more difficult [8]. This disorder is important to diagnose because 26% are malignant when there are detected in adults [9]. Teratomas in this location tend to be well developed.

The retroperitoneal teratoma was identified by chance in this patient and was characterized by benign cystic features with grease. A transperitoneal laparoscopic resection of a tumor was performed. During the surgery, the tumor exhibited the same physical appearance and was proximal to the liver, and as we had not previously encountered this situation, complete removal of the mass from the liver surface was performed.

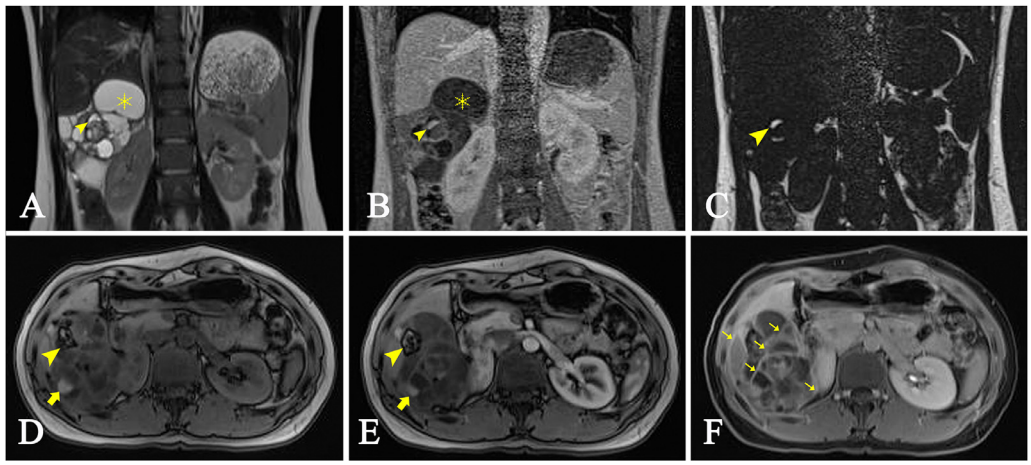

Fig. 4 – MRI of a tumor in the coronal plane on T2 Haste (A), T1-weighted in-phase (B), and out-of-phase (C) show cystic (asterisks) and greasy (arrowheads) element. On the non–enhanced (D) and enhanced axial T1-weighted images in the arterial phase (E) and in delay phase (F) show the mixture signal intensity (arrowheads and thick arrows). Wall and septals of tumor show enhance (thin arrows).

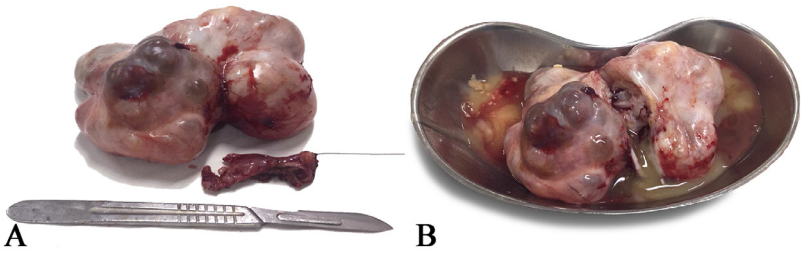

Fig. 5 – (A) Photograph of the gross specimen shows the mass with a multilocular lesion. The stem of mass is derived from the retroperitoneal. (B) It shows multiseptate cystic mass filled with sebum, yellowish pasty material and several solid nodular masses findings that account for the fat echogenicity and signal intensity seen at radiography imaging. (Color version of figure is available online.)

In our case, a retroperitoneal mixed mass was detected with ultrasound. This lesion was initially considered as a primary tumor. Detailed anamnesis revealed a previous mature ovarian teratoma 4 years previously, converting the initially primary retroperitoneal mass as a high suspicion of a secondary location of the initial teratoma, which was finally confirmed by surgery. As in our case, the review of the literature concludes that the clinical presentation of such a retroperitoneal mass is nonspecific with a dull pain in the right upper abdominal. The role of imaging is to orientate the diagnosis.

The present study reported a rare case of a retroperitoneal teratoma accompanied by abdominal pain, which was treated successfully by a complete resection of a tumor. The potential mechanism was unclear, but we hypothesized that the former surgery and surgical clips lost may have been involved. The diagnosis of a retroperitoneal teratoma could be made on the basis of imaging studies, and the gold standard treatment strategy for this neoplasm is surgical resection without rupture.

Radiology can give information about the structure of the mass, which can be of 2 types: solid or cystic. Ultrasound (US), CT, and magnetic resonance imaging contribute to detecting the 3 components of the solid type (tissular, fatty, and calcified components) and to identifying the cystic type (with or without fat or calcium component). The cystic type may contain enhanced septations [9]. In our case, there is the absence of calcification in mass. However, all other features are directed toward the teratoma with originating in the retroperitoneal cavity. The presence of the different components help to suggest the correct diagnosis on the basis of imaging, but surgery and pathology are required for the final diagnosis. Differential diagnosis includes a secondary location of ovarian carcinoma, sarcoma (when fat is present, then liposarcoma has to be evocated), ovary metastases, neurogenic mass, tuberculosis, Kaposi sarcoma, Castleman disease, and growing teratoma syndrome (development of a mature teratoma following chemotherapy for a nonseminomatous tumor).

When we consider retroperitoneal cystic masses, the following possibilities are to be included: the neoplastic conditions include cystic lymphangioma, mucinous cystadenoma, cystic teratoma, cystic mesothelioma, Müllerian cyst, epidermoid cyst, tailgut cyst, bronchogenic cyst, pseudomyxoma retroperitoneum, and perianal mucinous carcinoma; non-neoplastic disorders include pancreatic pseudocyst, lympho-

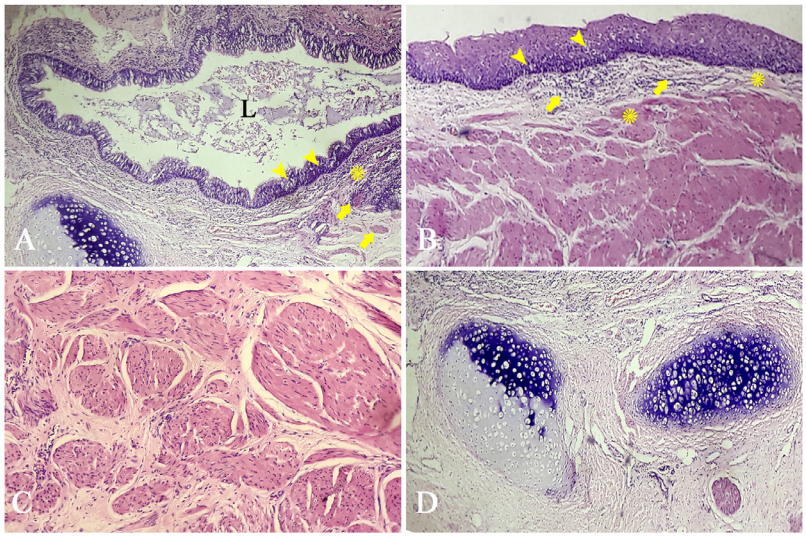

Fig. 6 – Mixed germ cell tumors (mature cystic teratoma) in a 22-year-old woman. (A and B, ×10 and ×40, hematoxylin-eosin stain) Photomicrograph of the cyst wall shows squamous cell lining (arrowheads), sebaceous glands (arrows), and intervening muscle (asterisks). Sebaceous material and cell debris are seen filling the cyst lumen (L) (×10 and ×40, hematoxylin-eosin stain). (C, ×40, hematoxylin-eosin stain) The image shows the structure of the muscle tissue. (D, ×10, hematoxylin-eosin stain) Photomicrograph of cartilage structure.

cele, , and hematoma [9,10]. At histology, fibrosis, necrosis, or mature teratoma can be observed [11]. In most of the cases, surgical is made immediately, with a good outcome.

Conclusion

The secondary localization of retroperitoneal teratoma is uncommon. Although the diagnosis can be made preoperatively by the characteristics of a tumor on the imaging modalities, a definitive diagnosis is established upon histologic assessment. Imaging modalities contribute for the suggestion of the correct diagnosis, by showing cystic retroperitoneal masses with septation, multiple calcifications, and parietal enhancement. In patients with the previous story of ovary neoplasm, the secondary location of the disease is one of the diagnostic possibilities. Surgical resection is the mainstay in the treatment of mature retroperitoneal teratomas.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2019.03.008.

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