Diagnostic dilemmas in large adrenal pseudocysts: A case report

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1. Introduction

Adrenal cysts are uncommon with a reported incidence rate of 0.06–0.18%. However, in view of the increasing detection of such incidentalomas by imaging & autopsy, their true incidence is likely much higher (1–3). They are a diverse group of cysts classified into pseudocysts, endothelial cysts, epithelial cysts, and parasitic cysts, the commonest being, endothelial (vascular) cysts & pseudocysts. Majority are diagnosed in late adulthood (3–5). It is simple to diagnose adrenal cysts by cross-sectional imaging when they are smaller in size. If however, the cyst attains large sizes, it can create a lot of confusion regarding the organ of origin. The range of differentials would then include hepatic, splenic, renal, pancreatic, retroperitoneal, mesenteric, urachal cysts and adrenal tumors. This can delay correct management. Additionally, large adrenal cysts, more than 5 cm in diameter carry a 7% risk of malignancy, adrenal pseudocysts in particular have a known association with adrenal neoplasms (6,7). We present one such rare case of a giant right adrenal pseudocyst, initially misdiagnosed as an exophytic simple hepatic cyst.

2. Case report

A female patient aged 53 years presented with painless, progressive distention of abdomen with no significant past medical history. Sonography revealed a large, exophytic, thin walled, unilocular hepatic cyst with anechoic contents. Echinococcal cyst was excluded by serology. Further evaluation by MRI confirmed the diagnosis of an exophytic simple hepatic cyst. She underwent cyst aspiration, and was lost to regular follow-up thereafter. The patient presented 2 years later with the same complaint. On abdominal sonography, the cyst had grown back to its former size. In view of recurrence, surgical resection was planned. Pre-operative CT abdomen showed a large cystic lesion in the right hemi-abdomen measuring 18 x 15 x 12 cm with a volume of 3.2 l. The cyst was in contact with the entire inferior surface of the right lobe of liver (Fig. 1), displacing the IVC, head of pancreas to the left, and the transverse colon anteriorly (Fig. 2). The right kidney was malrotated and displaced inferiorly (Fig. 3). All the displaced organs were retroperitoneal. A systematic search did not reveal the right adrenal gland. An effort was made to identify the vascular feeders on CECT, only few small peripheral vessels were noted inferior & left lateral to the cyst (Fig. 1). Beak sign of the liver was inconsistent, present in the coronal images & absent in some of the sagittal images (Fig. 1). In view of the above mentioned findings, a differential diagnosis of giant right adrenal cyst versus large exophytic simple hepatic cyst was made. Pre-operative endocrinology workup ruled out adrenal dysfunction (7). Intra-operatively, the cyst was adrenal in origin (Fig. 6). Histopathology confirmed it to be an adrenal pseudocyst (Fig. 7). Retrospective evaluation of the CT images showed the stretched & thinned out limbs of the adrenal gland...
in the left lateral cyst wall, which had been interpreted as peripheral vessels supplying the cyst (Fig. 4). Post-operative period & follow-up at 6 weeks were uneventful.

3. Discussion

Cysts in the adrenal gland are rare and most of them produce no symptoms due to their small size (2,6). They can occur in any age group, but are usually detected in the 5th and 6th decades of life. Incidental adrenal cysts are increasingly encountered with the widespread use of CT & MRI. They usually become symptomatic when > 6 cm in diameter and the patients generally present with abdominal distention, pain or bowel disturbances (2,6,8). The cysts can be complicated by infection, hemorrhage or rupture (6). Adrenal dysfunction is rarely manifested (2,8). Our patient was unusual in that, despite such a large intra-abdominal mass, she had no pain or bowel complaints (9).

Non-neoplastic adrenal cysts are classified into four categories, namely vascular cysts (45%), pseudocysts (39%), epithelial cysts (9%) & parasitic cysts (7%) (2,9,10). Pseudocysts are so named because their walls are composed of fibrous tissue & lack an endothelial or epithelial lining (11). They are

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Fig. 1  CECT abdomen, coronal & sagittal reformats demonstrating size & extent of the cystic lesion. It is in contact with inferior surface of the right lobe of liver.

Fig. 2  Axial CT images showing the cyst displacing head of pancreas (orange arrow) & IVC (blue arrow) to the left & transverse colon (green arrow) anteriorly.

Fig. 3  The right kidney is malrotated & displaced inferiorly.
formed following hemorrhage into a normal or abnormal adrenal gland. Smaller cysts do not require treatment, only larger (>5 cm diameter) or symptomatic cysts require intervention (7,9). Percutaneous aspiration can be an initial management strategy, but is prone for recurrence (2). Surgical excision is curative (8).

Large lesions in the abdomen can pose a diagnostic challenge in cross-sectional imaging studies (2,6). To improve diagnostic accuracy, the first step would be to localize the organ as intraperitoneal or retroperitoneal, based on the pattern of displacement of adjacent structures. In our patient, all the displaced structures were retroperitoneal, pointing to the retroperitoneal origin of the lesion. The next step is to identify whether all the retroperitoneal structures can be individually identified. Large lesions arising from small organs can obscure the parent organ, known as the phantom organ sign (12). This was an invaluable clue in our patient, as her right adrenal gland could not be visualized. The other radiological sign helpful in detecting the organ of origin, is the beak sign, where the edge of the organ from which the mass arises assumes a beak shape (12). However these signs are not infallible, false positives do exist (12). In our case the beak sign in the liver was present in the coronal images & absent in some sagittal

Fig. 4  The stretched & thinned out limbs of the right adrenal gland.

Fig. 5  Beak sign in the liver was present in the coronal reformats, but was inconsistent in the sagittal images.
sections giving rise to a confusing picture (Fig. 5). Identifying the vascular supply to the lesion can also help in ascertaining the parent organ, for which angiographic studies are useful. A review of published literature of large adrenal cystic lesions showed many were either misdiagnosed or the organ of origin remained undetermined by imaging studies (2,6,8,10,13–18). We propose a new imaging sign which can aid in identifying large adrenal cysts. When the adrenal gland cannot be visualized separately, an attempt should be made to search for the presence of thinned out, stretched limbs of the adrenal gland in the cyst wall. It was clearly seen during retrospective re-evaluation of our patient’s pre-operative CT images (Fig. 4). We also propose this sign be named “adrenal limb-wall sign.” It would be specific for diagnosing giant adrenal cysts. In previously published case reports of large adrenal pseudocysts, the compressed adrenal limbs embedded in the cyst wall was easily visualized with the naked eye in the gross pathology specimens (19,20–22). It is therefore very likely they can be visualized in imaging studies also, if a dedicated attempt is made to look for it. Our new sign may reduce the incidence of misdiagnosis. The pseudocyst in our case contained serous fluid making it relatively easy to visualize the adrenal limbs in the cyst wall. Further studies are required to evaluate applicability of this imaging feature in large suprarenal cysts complicated by hemorrhage, infection or complex loculations.

4. Conclusion

To summarize, we present a rare case of a giant right adrenal pseudocyst incorrectly diagnosed by initial sonography & abdominal MRI as an exophytic simple hepatic cyst. Any large abdominal lesion can create a confusing picture in cross-sectional imaging. The phantom organ sign & displaced retroperitoneal structures aided in identifying the possible organ of origin in our case. However, the beak sign was misleading. It is suggested that in large retroperitoneal cystic lesions, the presence of stretched out limbs of the adrenal gland in the cyst wall should be specifically looked for, which we propose be called the adrenal limb-wall sign. It will aid in early proper management & a retroperitoneal surgical approach can be planned if required.

References

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