Adrenal-renal Fusion: A Rare and Challenging Case for the Adrenal Surgeon

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ABSTRACT

Adrenal-renal fusion is a rare entity wherein the capsule of the adrenal gland is fused to the kidney. Here, we report a case of adrenal-renal fusion making intraoperative dissection challenging. We also report on four other cases of adrenal-renal fusion at our institution and a review of the literature. Although rare, radiologists and surgeons must be aware of this condition and consider it as a possibility, especially when dealing with upper pole renal lesions in order to avoid misdiagnosis and unnecessary resections.

Keywords: Adrenal fusion, Adrenal-renal fusion, Intrarenal adrenal.

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BACKGROUND

Adrenal-renal fusion is a rare entity wherein the capsule of the adrenal gland is fused to the kidney. The condition is usually found incidentally and has no clinical significance. However, reports have shown this finding can be misleading on preoperative imaging. Here, we report a case of adrenal-renal fusion making intraoperative dissection challenging. We also report on four other cases of adrenal-renal fusion at our institution and a review of the literature.

CASE REPORT

We present a 54-year-old male with a long-standing history of difficult-to-control hypertension. Over a 10-year period, the patient had been on four different anti-hypertensive medications and on recent laboratory evaluation was found to have hypokalemia. Further laboratory analysis showed an aldosterone-to-renin ratio of 31. He was, therefore, diagnosed with primary hyperaldosteronism. A computerized tomography (CT) of the abdomen and pelvis was obtained, which suggested a small nodule in the right adrenal gland. To further elucidate the source, adrenal vein sampling was performed which lateralized to the right adrenal gland. The patient was taken to the operating room for a planned laparoscopic, transabdominal right adrenalectomy. The operation proceeded in the typical fashion. After the adrenal vein was ligated, attention was then turned to dissecting the adrenal gland away from the retroperitoneal fat and kidney. Typically, there is a layer of fat between the two organs and the adrenal gland was noted to be tightly adherent to the kidney. This made dissection from the kidney challenging, and ultimately the capsule of the kidney had to be violated to remove the adrenal gland. As this is not the typical course of a laparoscopic adrenalectomy, there was a clinically insignificant increase in blood loss. However, the patient tolerated the operation well and was discharged on postoperative day 1.

On pathologic examination, a 1.7 × 1.5 × 0.7 cm portion of red-brown tissue attached to the surface of the adrenal gland was noted to be grossly consistent with renal parenchyma (Figs 1 and 2). Therefore, the patient was deemed to have adrenal-renal fusion, which explained the difficulty in the dissection in the operating room.

REVIEW OF THE LITERATURE AND PATHOLOGIC REVIEW OF OUR INSTITUTIONAL EXPERIENCE

On review of our institutional pathology experience with adrenal-renal fusion, four more cases were found between the years 2005 and 2014. A search of the literature resulted in 13 additional cases. Combined with the 5 cases from the authors’ institution, this totals 18 cases (Table 1). Eleven...
of the patients were female and 7 were male. The age ranged from 35 to 83 years, with a mean age of 56 years. The most common findings were renal cell carcinoma (n = 5) and normal pathology (n = 5), followed by adrenal cortical adenoma (n = 3) and renal cyst (n = 2). The location of the fusion was most commonly at the upper pole of the kidney (n = 13). (This does not make any sense. It is always in upper pole since, i.e., the anatomical position of the adrenal gland.) In all cases where distinction was made, the adherent or intrarenal adrenal tissue was adrenal cortical tissue. There were no reported cases of adrenal medullary tissue present in the fused portion of tissue. The length of the fusion ranged from 0.3 to 3.6 cm.

RADIOLOGIC REVIEW OF OUR INSTITUTIONAL EXPERIENCE

The CT in case #1 revealed a mass that seemed to arise from the kidney. The adrenal gland appeared separate from the kidney. The CT in case #2 did not demonstrate a definite fat plane separating either kidney from their respective adrenal glands. Case #3 did not have any preoperative imaging available. The CT in case #4 demonstrated apparent separation of the adrenal gland from the kidney. The CT in case #5 revealed a renal cyst but did not demonstrate a definite fat plane separating either kidney from their respective adrenal glands.

DISCUSSION

Adrenal-renal fusion is a rare finding that was first described by Rokitansky in 1855, who divided the fusion into two forms: A developmental type and a postinflammatory fibrosis type. It has been hypothesized that the developmental fusion form results from a failure of complete encapsulation of the two organs due to lack of stimulation by retroperitoneal mesenchymal tissue during the embryological development. The exact incidence of renal-adrenal fusion is unknown as it has only been described in the literature in several case reports and one case series. This review represents the second case series in the literature.

Typically, a fused adrenal gland and kidney are found incidentally after a resection and though the fusion itself usually causes no physiological symptoms and is clinically insignificant, its morphological anomaly can confuse radiologists on CT and magnetic resonance scans, leading to misdiagnosis. In addition to their close apposition, scanner resolution, diaphragmatic motion, and the averaging of adjacent voxels limit the ability of radiologists to always confidently visualize a fat plane entirely separating the two organs. While a large congenital fusion might be diagnosed prospectively, subtle fusions secondary to scarring could easily be missed. This is a location where it can even be difficult to confidently localize some lesions to either the kidney or the adrenal gland. Mahadevia et al reported a case in which an adrenal lesion mimicked a renal lesion on radiological imaging due to adrenal-renal fusion whereby an abdominal CT scan of a patient who presented with abdominal pain showed a focal lesion in the upper pole of the left kidney, which appeared to be a renal mass. Diagnosis of a renal cell carcinoma was made and the patient underwent laparoscopic partial left nephrectomy at which time it was found that the mass was actually arising from the adrenal gland. Pathology revealed that the mass was an adrenal cortical adenoma. In this case, the authors suggested that there was no way of making the correct diagnosis of adrenal adenoma before reviewing the specimen histologically after surgical resection.
Table 1: Patient demographics and pathologic diagnosis

<table>
<thead>
<tr>
<th>Case</th>
<th>Age</th>
<th>Sex</th>
<th>Reason for resection</th>
<th>Tumor laterality</th>
<th>Size (cm)</th>
<th>Pathologic diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>54</td>
<td>M</td>
<td>Primary hyperaldosteronism with interventional radiology localization to right side</td>
<td>R</td>
<td>$0.9 \times 0.8 \times 0.7$</td>
<td>Adrenal cortical adenoma; adrenal-renal fusion</td>
</tr>
<tr>
<td>2</td>
<td>41</td>
<td>F</td>
<td>Renal mass</td>
<td>R</td>
<td>$3.0 \times 2.0 \times 1.5$</td>
<td>Adrenal cortical neoplasm arising in an adrenal-renal fusion; adrenal-renal fusion</td>
</tr>
<tr>
<td>3</td>
<td>45</td>
<td>F</td>
<td>Renal mass</td>
<td>R</td>
<td>$1.7 \times 1.5 \times 1.0$</td>
<td>Clear cell renal cell carcinoma; adrenal-renal fusion</td>
</tr>
<tr>
<td>4</td>
<td>68</td>
<td>M</td>
<td>Renal mass</td>
<td>R</td>
<td>$1.5 \times 1.3 \times 1.2$</td>
<td>Clear cell renal cell carcinoma; Adrenal-renal fusion and adjacent renal subcapsular hematoma</td>
</tr>
<tr>
<td>5</td>
<td>43</td>
<td>F</td>
<td>Renal cortical cyst</td>
<td>R</td>
<td>$0.6 \times 0.4 \times 0.4$</td>
<td>Adrenal-renal fusion and adjacent cortical cyst</td>
</tr>
<tr>
<td>6</td>
<td>47</td>
<td>F</td>
<td>Incidentally found on postmortem examination</td>
<td>R</td>
<td>No comment</td>
<td>Partial adrenorenal fusion was also demonstrated. Over most of their apposed surfaces, the two organs were separated by a thin fibrous capsule devoid of fat cells but multiple gaps appeared randomly in the capsule leading to intimate mingling of the kidney and adrenal</td>
</tr>
</tbody>
</table>

Colberg et al\(^2\)

7 83  F  Benign renal mass  R  No comment  Fusion of the adrenal gland with the kidney and adrenal cortical clear cells admixed with renal parenchyma. Renal cystic changes were noted at the junction of the fusion

Fan et al\(^4\)

8 62  M  Renal cyst  R  No comment  A triangular atrophic adrenal gland was identified with two limbs embedded in the cyst wall. The cyst wall was composed of fibrocollagenous tissue that appeared continuous with kidney capsule. Furthermore, areas of adrenal-renal union and fusion were evident

Mahadevia et al\(^4\)

9 76  F  Adrenal cortical adenoma  L  No comment  Fusion of the kidney and adrenal gland, as demonstrated by a lack of an outer renal capsule. Adrenal adenoma composed of clear cells also contained renal tubules and was partly intrarenal

Ye et al\(^6\)

10 44  M  Transplant nephrectomy  N/A  Microscopic  Ectopic adrenal tissue with focal necrosis; subcapsular

11 35  F  Renal cyst  R  0.8  Intrarenal adrenal gland tissue with adjacent simple renal cyst

12 55  F  Kidney biopsy  L  0.3  Intrarenal adrenal rest; subcapsular

13 75  M  Renal cell carcinoma  L  A small amount  A small amount of adrenal tissue adherent to the medial renal capsule; subcapsular

14 72  M  Urothelial carcinoma  R  0.7  A small subcapsular adrenal rest; subcapsular

15 51  M  Renal cell carcinoma  L  A small portion  Renal-adrenal fusion

16 52  F  End-stage kidney disease  L  Microscopic  Ectopic adrenal tissue in renal capsule and cortex; subcapsular and extracapsular

17 49  F  Adrenal cortical adenoma  L  $2 \times 2 \times 1$  Cortical adenoma arising in adherent and intrarenal adrenal gland; renal-adrenal fusion

18 59  F  Renal cell carcinoma  R  1.5  Small focus of ectopic adrenal tissue; subcapsular

In addition to the potential for the capsule of the two organs to become fused, it is possible that tissue from one organ can actually be found embedded in the adjacent organ. Colberg et al\(^2\) reported a case of complete renal-adrenal fusion that was discovered incidentally during a partial nephrectomy performed after diagnosing a solid mass in the upper pole of the right kidney. Histologic examination of the specimen revealed that the adrenal and renal cortical clear cells and the renal parenchyma were mixed with renal cystic changes at the junction of the fusion. Further immunohistochemical staining revealed the presence of some intrarenal adrenal tissues that had been completely embedded in the kidney showing as an ectopic adrenal tissue. Several other cases in this review presented in this manner. This highlights the importance of awareness of this condition as intrarenal adrenocortical clear cells may easily be confused with clear cell renal carcinoma.
Although the classic etiology for renal-adrenal fusion is a result of a development abortion, fusion can also occur as the result of a postinflammatory state. Fan et al\(^4\) reported a superior renal cyst diagnosed in a patient on CT examination. Upon resection, the cyst was found to be inflamed and thickly fibrotic. The histologic examination of the cyst was remarkable in that it showed the cyst to be of adrenal origin, suggesting that the mass was an adrenal gland that had undergone atrophy. The authors propose that the adrenal-renal fusion, in this case, was caused by cystic degeneration of the adrenal gland. The fibrosis that followed the degeneration was likely so strong that it obliterated the space that normally separates the adrenal gland from the kidney, leading to the fusion. One case in the author’s experience (case 4) presented in a similar fashion whereby the fusion appeared at an area of subcapsular hematoma, suggesting a postinflammatory reaction.

In conclusion, although renal-adrenal fusion is relatively rare in the literature, radiologists and surgeons must be aware of this condition and consider it as a possibility, especially when dealing with upper pole renal lesions in order to avoid misdiagnosis and unnecessary resections. It is the authors’ belief that adrenal-renal fusion may, in fact, be more common than reported. On review of our own series, in addition to the case presented here, we found four additional cases of adrenal-renal fusion, suggesting that there may be additional cases of adrenal-renal fusion that are not noticed by surgeons postoperatively. This entity can be particularly challenging during a laparoscopic adrenalec- tomy. Surgeons performing adrenalectomy should be aware of this possibility and consider this diagnosis when the dissection plane between the adrenal and renal tissue is particularly challenging.

REFERENCES