Benign adrenal and suprarenal retroperitoneal schwannomas can mimic aggressive adrenal malignancies: case report and review of the literature

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SUMMARY The suprarenal retroperitoneum and adrenal gland is a rare site of origin for benign schwannomas which frequently present as larger and more aggressive lesions than schwannomas identified elsewhere. These tumors are often surgically excised. We present a case of an 81-year-old asymptomatic man presenting with an incidental 10 cm left suprarenal retroperitoneal mass identified on CT. The mass was indiscernible from the adrenal gland, demonstrating heterogeneous enhancement with a centrally cystic/necrotic core, and punctate calcifications. Subsequent core needle biopsy demonstrated a benign adrenal schwannoma. The lesion has been managed conservatively with imaging follow up and without complication. DISCUSSION: Our review of the literature identifies 121 reported in vivo benign adrenal and suprarenal schwannomas published to date with imaging features available for 90 cases (74%). All cases were encapsulated with the average size measuring over 6.5 cm. Fifteen percent (13/84) of reported lesions measured over 10 cm at presentation. Punctate calcification was present in 50% (26/52) of reporting cases. Nearly 50% (40/86) of cases demonstrate cystic/necrotic appearances on imaging. Despite aggressive appearances, our case demonstrates that biopsy and surveillance may represent a reasonable alternative to surgery in suboptimal surgical candidates.

Keywords adrenal, schwannoma, retroperitoneal, suprarenal, malignancy, nerve

1. Introduction

Non-malignant masses account for around 20% of primary retroperitoneal (RP) tumors with benign schwannomas representing a subset of only 5% of primary RP tumors (1). Adrenal and suprarenal RP schwannomas represent an even rarer origin subset. Schwannomas presenting in this location tend to be larger and more heterogeneous at time of presentation than schwannomas presenting elsewhere in the body (2-5). Surgical resection is often the treatment of choice for these tumors (2).

2. Case Report

An 81-year-old otherwise asymptomatic man presented via ambulance to our tertiary care hospital after falling from a ladder. He remained hemodynamically stable throughout his presentation. On account of his mechanism and multifocal pain, he underwent a trauma scan of his chest, abdomen, and pelvis. An incidental 8 × 5 × 10 cm (anteroposterior × transverse × craniocaudal) left suprarenal lesion indiscernible from the left adrenal gland was identified (Figure 1). The lesion demonstrated peripheral arterial and portal venous enhancement with a centrally cystic/necrotic core. There were punctate calcifications scattered throughout. The right adrenal gland was normal. No regional lymph node enlargement was present. No other primary or metastatic lesions were seen elsewhere throughout the body.

A 24-hour urine metanephrine study was negative for PCC. Bloodwork was otherwise non-contributory. A subsequent ultrasound-guided core needle biopsy was performed. Histopathology demonstrated a tumor composed of spindle cells with minimal atypia and/or mitotic activity (Figure 2). The spindle cells were dense, wavy, and with tapered ends. Hyalinized blood vessels were readily present. Subsequent immunohistochemical analysis demonstrated a diffusely positive S100 stain while markers for other RP tumors including MART1, HMB45, DOG1, CD117, MDM2, actin, desmin,
caldesmon, AE1/AE3, CD34 and STAT6 were all negative. Pathological features were consistent with a benign adrenal schwannoma. A glial fibrillary acidic protein (GFAP) stain was performed for the purposes of this report and was positive.

Following consultation with urology, the lesion was managed conservatively with observation. The patient has remained free of symptoms relating to the tumor. As of a six-month follow-up CT scan, no change in size or appearance of the lesion has been documented and the patient remained otherwise clinically asymptomatic.

3. Discussion

3.1. Literature review

An English language search of MEDLINE and Google Scholar from inception to July 24, 2019 with search terms including "schwannoma" AND ("retroperitoneal" OR "adrenal") was performed to evaluate for in vivo cases of adrenal or suprarenal RP schwannomas. A total of 121 reported cases were identified with 90 cases documenting imaging characteristics (Table 1) (2-44). The rarity of these reports is demonstrated by Li et al. who identified only 19 adrenal schwannomas in a series of almost 4,000 adrenal lesions (26). Patients in our literature review ranged in age between 14-81, with our report representing the oldest known patient described to date. As seen in our study, male gender accounts for less than half of the identified reports.

3.2. Clinical and imaging findings

Clinical and imaging features are summarized in Table 2. Similar to more than half of reported cases, the RP schwannoma in our patient was identified incidentally when investigating for another purpose. Most patients who did present with symptoms complained of non-specific symptoms including flank and abdominal pain and rarely abdominal distention. It is possible that several of these patients’ tumors were also identified incidentally with the symptoms attributable to an alternative otherwise undetected cause. This would correspond with a study by Zhou et al. assessing pathologic features of 31 patients with adrenal schwannomas identifying 84% of patients as presenting incidentally (Table 3) (44). The high frequency of incidental and often delayed presentation likely accounts for why the average size of suprarenal RP schwannomas is larger than the average size of schwannomas identified elsewhere. In our review, the average size of identified tumors was over 6.5 cm with 15% (13/84) of reporting cases demonstrating tumors at least 10 cm in size.

All reported cases including ours demonstrated well encapsulated margins typical of a benign schwannoma. Punctate calcifications described in our case were present in 50% (26/52) of reporting imaging cases. The number of cases with calcifications may even be under-reported as several studies evaluated lesions with only ultrasound and/or MRI features but lacking spatial resolution necessary to identify small calcifications. This would correspond with Li et al.’s study where 84% (16/19) of their patients had some calcification...
| Author, Year | Ref. | Age/Gender | Presentation | Size (cm) | Margin | Heterogeneity | Solid/ Cystic | Calcification | Suspected Diagnosis | Diagnosis |
|--------------|------|------------|--------------|-----------|--------|---------------|--------------|--------------|-------------------|-----------|
| Abdessater 2018 (2) | 50/F | Asymptomatic | 10 | En | Heg | Cystic | Present | ACC | Surgery |
| Adas 2013 (3) | 32/F | Flank Pain | 10 | En | Heg | Cystic | NR | ACC | Surgery |
| Babuya 2017 (4) | 69/M | Abdominal Pain | 4 | En | Hog | Solid | None | NAA | Surgery |
| Bakhshi 2011 (5) | 34/F | Asymptomatic | 9 | En | Heg | Cystic | NR | NAA | Surgery |
| Bedard 1986 (6) | 63/F | Abdominal Pain | 6 | En | Hog | Solid | NR | NC | Surgery |
| Damdaran 2015, Case 1 (7) | 36/F | Flank Pain | 9 | En | Heg | Cystic | NR | ACC | Surgery |
| Damdaran 2015, Case 2 (7) | 50/F | Flank Pain | NR | En | Hog | NR | NR | NAA | Surgery |
| Fernandez 2016 (8) | 56/F | Asymptomatic | 10 | En | Heg | Cystic | NR | ACC | Surgery |
| Fu 2015 (9) | 71/M | Asymptomatic | 15 | En | Heg | Cystic | NR | NC | Surgery |
| Garg 2007 (10) | 50/F | Abdominal Pain | 9 | En | Hog | Solid | NR | ACC | Surgery |
| Gazuza 2007 (11) | 42/M | Flank Pain | 12 | En | Heg | Cystic | NR | ACC | Surgery |
| Goh 2006, Case 1 (12) | 46/M | Asymptomatic | NR | En | Heg | Cystic | NR | ACC | Surgery |
| Goh 2006, Case 2 (12) | 28/F | Asymptomatic | NR | En | Heg | Cystic | Present | ACC | Surgery |
| Goh 2006, Case 3 (12) | 49/M | Asymptomatic | NR | En | Heg | Cystic | None | ACC | Surgery |
| Goh 2006, Case 4 (12) | 58/M | Flank Pain | NR | En | Heg | Cystic | None | ACC | Surgery |
| Grasso 2015 (13) | 45/M | Asymptomatic | 12 | En | Heg | Cystic | None | ACC | Surgery |
| Hsiao 2008 (14) | 49/M | Asymptomatic | 5 | En | Hog | Solid | None | NAA | Surgery |
| Igawa 1998 (15) | 45/M | Abdominal Pain | 6.5 | En | Hog | Solid | NR | ACC | Surgery |
| Ikemoto 2002 (16) | 62/F | Abdominal Pain | 12 | En | Hog | Solid | None | NC | Surgery |
| Inoukichi 2006 (17) | 35/F | Asymptomatic | 7 | En | Heg | Cystic | Present | ACC | Surgery |
| Jakowski 2008 (18) | 51/F | Asymptomatic | 5.5 | NR | NR | NR | NR | NC | Surgery |
| Jeshtadi 2014 (19) | 55/F | Flank Pain | 6.5 | En | Hog | Solid | NR | NC | Surgery |
| Khurram 2015 (20) | 64/M | Asymptomatic | 2 | En | Hog | Solid | None | NAA | Surgery |
| Kleinman 2011 (21) | 31/F | Asymptomatic | 4.5 | En | Heg | Solid | NR | NAA | Surgery |
| Konstantinos 2012 (22) | 71/F | Flank Pain | 8 | En | Heg | Cystic | Present | ACC | Surgery |
| Korets 2007 (23) | 70/M | Hernaturna | 3 | En | Heg | Cystic | None | NC | Surgery |
| Kostakopoulos 1993 (24) | 38/F | Abdominal Pain | 18 | En | Heg | Cystic | None | ACC | Surgery |
| Lau 2006, Case 1 (25) | 73/M | Abdominal Pain | 9 | NR | NR | NR | NR | NC | Surgery |
| Lau 2006, Case 2 (25) | 26/F | Abdominal Pain | 10 | NR | NR | NR | NR | NC | Surgery |
| Li 2015, Case 1 (26) | 38/F | NR | 7 | En | Hog | Solid | Present | NAA | Surgery |
| Li 2015, Case 2 (26) | 31/F | NR | 4 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 3 (26) | 50/F | NR | 4 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 4 (26) | 55/F | NR | 6 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 5 (26) | 50/F | NR | 9 | En | Heg | Cystic | None | Schwannoma | Surgery |
| Li 2015, Case 6 (26) | 23/M | NR | 6 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 7 (26) | 54/F | NR | 7 | En | Hog | Solid | None | ACC | Surgery |
| Li 2015, Case 8 (26) | 66/F | NR | 6.5 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 9 (26) | 56/F | NR | 5.5 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 10 (26) | 61/M | NR | 6 | En | Hog | Solid | Present | Schwannoma | Surgery |
| Li 2015, Case 11 (26) | 65/F | NR | 8 | En | Hog | Solid | None | Teratoma | Surgery |
| Li 2015, Case 12 (26) | 34/M | NR | 5 | En | Heg | Cystic | Present | ACC | Surgery |
| Li 2015, Case 13 (26) | 64/F | NR | 6 | En | Hog | Solid | Present | NAA | Surgery |
| Li 2015, Case 14 (26) | 44/F | NR | 6 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 15 (26) | 46/F | NR | 6.5 | En | Hog | Solid | Present | ACC | Surgery |
| Li 2015, Case 16 (26) | 50/F | NR | 7 | En | Hog | Cystic | Present | ACC | Surgery |
| Li 2015, Case 17 (26) | 40/F | NR | 5 | En | Hog | Solid | Present | NAA | Surgery |
| Li 2015, Case 18 (26) | 32/F | NR | 6 | En | Hog | Solid | Present | Schwannoma | Surgery |
| Li 2015, Case 19 (26) | 58/F | NR | 7 | En | Hog | Solid | Present | ACC | Surgery |
| Liu 2012, Case 1 (27) | 14/M | Abdominal Pain | 7 | En | Hog | Solid | None | NAA | Surgery |
| Liu 2012, Case 2 (27) | 51/M | Abdominal Distension | 4 | En | Heg | Cystic | NR | ACC | Surgery |
| Liu 2012, Case 3 (27) | 41/M | Flank Pain | 7 | En | Hog | Solid | NR | NAA | Surgery |
| Oberoi 2019 (28) | 50/F | Asymptomatic | 12 | En | Heg | Solid | NR | ACC | Surgery |
| Onodera 2008 (29) | 62/F | Asymptomatic | 4 | En | Hog | Solid | None | NAA | Surgery |
| Pittasch 2000 (30) | 56/F | Abdominal Pain | 12 | En | Heg | Solid | NR | ACC | Surgery |
| Richter 2011 (31) | 30/F | Abdominal Pain | 14 | En | Heg | Solid | None | NAA | Surgery |
| Said 2017 (32) | 64/M | Asymptomatic | 9 | En | Heg | Cystic | Present | ACC | Surgery |
| Shabana 2019 (33) | 32/M | Flank Pain | 5 | En | Heg | Solid | NR | NAA | Surgery |
| Suzuki 2007 (34) | 33/M | Asymptomatic | 8 | En | Hog | Solid | None | NC | Surgery |
| Tang 2018, Case 1 (35) | 47/F | Asymptomatic | 5.5 | En | Heg | Cystic | None | NC | Surgery |
| Tang 2018, Case 2 (35) | 65/F | Asymptomatic | 5 | En | Heg | Cystic | None | NC | Surgery |
| Tang 2018, Case 3 (35) | 69/F | Asymptomatic | 4 | En | Heg | Cystic | None | NC | Surgery |
| Tang 2018, Case 4 (35) | 50/F | Abdominal Pain | 8 | En | Heg | Cystic | None | NC | Surgery |
| Tang 2018, Case 5 (35) | 31/F | Asymptomatic | 3.5 | En | Heg | Solid | None | NC | Surgery |

NR: Not Reported; En: Encapsulated; Heg: Heterogenous; Hog: Homogenous; NC: not clear; ACC: adrenal cortical carcinoma; NAA: nonfunctional adrenal adenoma; PCC: Phaeochromocytoma.
identify a higher proportion of schwannomas. The high frequency of central cystic changes may relate to a frequently delayed presentation with increased size of these tumors at presentation. A study by Hirose et al. identify a higher proportion of GFAP positive schwannomas in the retroperitoneum compared to elsewhere (45). They attribute this to the origin and subtype of RP schwannomas and could provide an alternative reason for the more aggressive appearance of many suprarenal RP schwannomas at presentation. Of identified cases in our review, a GFAP stain was only performed prospectively by Fernandez et al. (26). Our case also demonstrated marked heterogeneity with a centrally cystic/necrotic core, a feature which has been described in nearly 50% (40/86) of reported suprarenal RP schwannomas. The high frequency of central cystic changes may relate to a frequently delayed presentation with increased size of these tumors at presentation. A study by Hirose et al. identify a higher proportion of...
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Table 3. Clinical and pathological characteristics of suprarenal retroperitoneal schwannomas published by Zhou et al. (44)

| Cases | Age/Gender | Presentation | Size (cm) | Margin | Heterogeneity | Solid/Cystic | Calcification | Suspected Diagnosis | Diagnosis |
|-------|------------|--------------|-----------|--------|---------------|--------------|---------------|---------------------|-----------|
| Case 1 | 45/F       | Asymptomatic | 3         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 2 | 53/M       | Asymptomatic | 8         | En     | NR            | Cystic       | None          | NAA                 | Surgery   |
| Case 3 | 66/M       | Hypotension  | 5         | En     | NR            | Cystic       | Present       | FAA                 | Surgery   |
| Case 4 | 43/M       | Asymptomatic | 6.5       | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 5 | 62/F       | Asymptomatic | 5         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 6 | 32/F       | Asymptomatic | 6         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 7 | 42/F       | Asymptomatic | 6         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 8 | 44/F       | Flank Pain   | 5         | En     | NR            | Solid        | Present       | FAA                 | Surgery   |
| Case 9 | 26/F       | Asymptomatic | 3         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 10| 50/M       | Flank Pain   | 4         | En     | NR            | Solid        | None          | FAA                 | Surgery   |
| Case 11| 58/M       | Asymptomatic | 2.5       | En     | NR            | Solid        | None          | FAA                 | Surgery   |
| Case 12| 56/F       | Asymptomatic | 1         | En     | NR            | Solid        | Present       | FAA                 | Surgery   |
| Case 13| 38/M       | Asymptomatic | 3.5       | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 14| 61/F       | Asymptomatic | 12        | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 15| 57/F       | Cushing Syndrome | 6      | En     | NR            | Solid        | None          | FAA                 | Surgery   |
| Case 16| 47/F       | Asymptomatic | 12        | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 17| 48/F       | Asymptomatic | 8         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 18| 40/M       | Asymptomatic | 3         | En     | NR            | Solid        | Present       | FAA                 | Surgery   |
| Case 19| 42/M       | Asymptomatic | 3         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 20| 42/F       | Asymptomatic | 4         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 21| 52/F       | Asymptomatic | 4.5       | En     | NR            | Solid        | Present       | FAA                 | Surgery   |
| Case 22| 31/M       | Asymptomatic | 7         | En     | NR            | Cystic       | None          | NAA                 | Surgery   |
| Case 23| 69/M       | Asymptomatic | 2.5       | En     | NR            | Cystic       | None          | NAA                 | Surgery   |
| Case 24| 67/M       | Asymptomatic | 6         | En     | NR            | Cystic       | None          | PCC                 | Surgery   |
| Case 25| 46/F       | Asymptomatic | 12        | En     | NR            | Solid        | Present       | NAA                 | Surgery   |
| Case 26| 29/F       | Abdominal Pain| 2         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 27| 31/F       | Asymptomatic | 4         | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 28| 54/M       | Asymptomatic | 8.5       | En     | NR            | Solid        | None          | NAA                 | Surgery   |
| Case 29| 67/F       | Asymptomatic | 7         | En     | NR            | Cystic       | Present       | NAA                 | Surgery   |
| Case 30| 71/M       | Asymptomatic | 1         | En     | NR            | Cystic       | None          | NAA                 | Surgery   |
| Case 31| 33/F       | Asymptomatic | 4         | En     | NR            | Solid        | None          | NAA                 | Surgery   |

En: Encapsulated; NR: Not Reported; ACC: adrenal cortical carcinoma; NAA: nonfunctional adrenal adenoma; FAA: functional adrenal adenoma; PCC: Pheochromocytoma.

Table 4. Differential diagnosis table for retroperitoneal adrenal and suprarenal schwannomas

| Adrenal adenoma | • Typically small.
|                 | • Typically homogeneous.
|                 | • Typically low density (HU < 10 on non-contrast imaging) and/or demonstrates washout characteristics (> 60% absolute washout; > 40% relative washout).
|                 | • Increasing size raises concern for malignancy.

| Adrenal cortical carcinoma | • Typically large (> 6 cm).
|                            | • Irregularly shaped (main differentiating factor from RP suprarenal schwannomas).
|                            | • Typically heterogeneous with central necrosis and/or hemorrhage.
|                            | • Calcification in up to 30%.

| Adrenal metastasis | • Variable appearance.
|                   | • Typically < 50% washout.

| Pheochromocytoma | • Typically large.
|                 | • Typically heterogeneous.
|                 | • Avid enhancement.
|                 | • Calcification in < 10%.
|                 | • Typically “light bulb bright” on T2 MRI sequence.

| Suprarenal Schwannoma | • Typically large (mean 6.5 cm).
|                       | • Encapsulated.
|                        | • Nearly 50% are heterogeneous with central necrosis and/or hemorrhage.
|                        | • Calcification in ~50%.
|                        | • Can be indistinguishable from adrenal metastasis on imaging.

Demonstrating positivity (8). Hirose et al. identified GFAP positivity in 92% of their RP cellular/fascicular type schwannomas, which would be consistent with GFAP positivity seen in our cellular type schwannoma.

3.3. Differential diagnoses
Irrespective of cause, our case and review of the literature demonstrates the high proportion of aggressive appearing
suprarenal schwannomas at imaging presentation. These tumors are frequently misattributed as malignant etiologies of adrenal origin such as adrenal cortical carcinoma and metastasis. Other adrenal lesions such as adrenal adenomas and PCCs are frequently considered as well. Solid lesions of retroperitoneal origin such as lymphoid tumors, sarcomas, teratomas, and other non-schwannoma neurogenic tumors can be considered but rarely represent the predominant differential in these lesions (46). Features for typical differential diagnoses for adrenal and suprarenal retroperitoneal schwannomas are described in Table 4.

4. Conclusion

Although rare, our case and review demonstrates that benign schwannoma is a differential consideration for suprarenal malignant lesions. All other known reported cases utilized resection for pathological diagnosis. While publication bias is likely a factor, resection may not be a preferable approach in older patients and/or poor surgical candidates. As in our case of an 81-year-old man reluctant to undergo aggressive surgery, a core needle biopsy and close imaging follow up may be sufficient for management despite the aggressive appearance on initial presentation.

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