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Case Report

Ancient schwannoma in the right iliac fossa: A case report and review of literature ☆,☆☆

Marwan Alaswad[a,*], *Tariq Saleh*[a], *Fatima Alaidaros*[a], *Abdullah Al Otry*[a], *Ayman Z. Azzam*[b,c], *Tarek M. Amin*[b]

[a] *College of Medicine, Alfaisal University, Riyadh, Saudi Arabia*
[b] *Department of Surgery, Surgical oncology, King Faisal Specialist Hospital and Research Centre, Riyadh, Saudi Arabia*
[c] *Department of General Surgery, Faculty of Medicine, Alexandria University, Alexandria, Egypt*

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ABSTRACT

Ancient schwannomas are a rare variation of schwannomas, with the distinction being based on histopathological examination of the excised specimen. On histopathological examination, ancient schwannomas exhibit degenerative changes such as calcification, hyalinization, and cystic necrosis, along with S100 positivity. Complete surgical excision is the mainstay treatment for ancient schwannomas and carries a favorable prognosis. Recurrence is the most common complication, often arising from incomplete surgical excision. Herein, we present a case of a 41-year-old male who presented to our center as a case of a retroperitoneal mass for further investigations and diagnostic workup. Imaging showed a retroperitoneal mass in the right iliac fossa. We proceeded with ultrasound guided needle biopsy, and examination of the specimen confirmed the diagnosis of ancient schwannoma. Subsequently, the patient underwent surgery, and complete surgical excision was achieved. On follow-up 3-months later, the patient is doing well, and no signs of recurrence were found.

Introduction

Schwannomas are rare, benign neurogenic tumors that develop from the cells of the neural sheaths in the peripheral nervous system [1]. Although around 25%-50% of schwannomas occur in the head and neck, this type of tumor can rarely arise in any region of the body [1]. Typically, they are known to be solitary, encapsulated, and slow growing [2]. Ancient schwannomas, an uncommon variety of schwannoma,

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* Corresponding author.
E-mail address: Marwanalaswad21@outlook.com (M. Alaswad).
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Table 1 – Cases of ancient schwannomas arising in the iliac fossa.

Author	Age:	Gender:	Symptoms:	Location:	Diagnosis:	Treatment:
Hanif et al. [6]	28	Male	RLQ pain	Right iliac fossa	CT guided biopsy	Conservative with analgesia
Rai et al. [7]	53	Male	RLQ pain and positive Tinel's sign	Right iliac fossa	US and CT	Surgical resection
Mesina et al. [8]	33	Male	LLQ pain and paresthesia of left thigh	Left iliac fossa	US and CT	Surgical resection
Bhatia et al. [9]	64	Female	Asymptomatic	Right iliac fossa	CT with contrast	Surgical resection
Ginesu et al. [10]	62	Female	RLQ pain	Right iliac fossa	CT with contrast	Surgical resection
Our case	41	Male	RLQ pain	Right iliac fossa	US guided biopsy	Surgical resection

exhibit unique histopathological changes including spindle cells with Antoni A and B fibers, with areas of degenerative changes, such as calcification, hyalinization, and cystic necrosis [3]. These features could be confused with malignancy leading to an inaccurate diagnosis [1]. For that reason, histopathological investigations of an excised specimen or a biopsy is the gold standard modality of diagnosis and complete surgical removal is the main treatment [4]. We report a rare case of an ancient schwannoma occurring in the right iliac fossa.

Case presentation

A 41-year-old male patient presented to our institution complaining of on and off right lower abdominal pain for the past 3 months. There was no associated fever, nausea, or vomiting. Patient was referred to us from an outside institution as a case of retroperitoneal mass for further investigations and diagnostic workup. The patient is medically free and has no history of previous surgeries. Vitals, general, respiratory, and cardiovascular examinations were all normal. Abdomen was soft and lax with no tenderness. Labs, including tumor markers CA 12-5, CA 15-3, CA 19-9, and CEA were all normal.

CT abdomen showed a 4.1 × 2.8 × 4.9 cm oval, homogenous, well-defined right lower quadrant retroperitoneal mass overlying the right psoas muscle with no signs of invasion (Fig. 1). PET CT was done and showed a well-defined, mild FDG avid hypodense mass within right iliac fossa (Fig. 2). Subsequently, MRI showed a well-defined, hypervascular right lower quadrant retroperitoneal mass, with homogenous enhancement, and low T1 and T2 signal intensity (Fig. 3). T1 diffusion weighted MRI scan precontrast and postcontrast, along with T2 MRI scan showed a 4.8 × 2.6 × 4.3 cm well-defined smooth outlined right lower quadrant mass corresponding to the PET-CT scan observation; which demonstrates thin low T2 signal intensity wall and clustered contiguous low T2 signal intensity component, low T1 signal intensity, lack of signal drop on the opposed phase T1 image, lack of diffusion restriction, and progressive intense fairly homogeneous enhancement; compatible with a neoplastic process. This appearance is nonspecific and differential diagnosis may include neurogenic tumor, with no evidence of metastatic disease (Fig. 4).

The decision was made to proceed with US guided needle biopsy. Histopathological examination of the specimen showed a mass composed of spindle cells with a focal nuclear palisading pattern composed of hypercellular areas which express Antoni A and hypocellular areas which express Antoni B, with degenerative changes (Fig. 5). Immunohistochemistry showed tumor cells displaying uniform, intense positivity for S100 protein (Fig. 6), focally positive TLE1 and SMA, negative CD34 and STAT6, and a very low Ki-67 proliferative index of less than 2%. A final diagnosis of ancient schwannoma was made based on these findings.

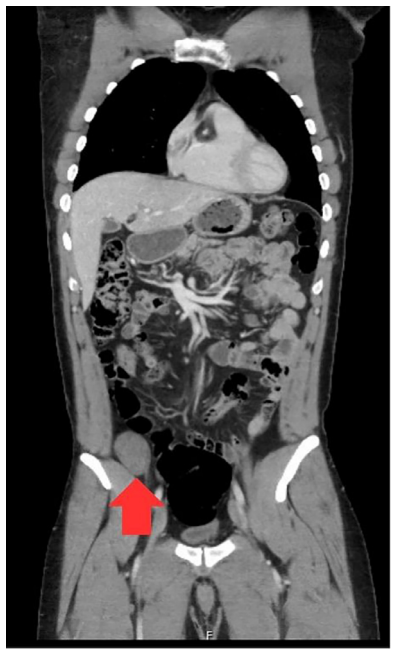

Fig. 1 – Coronal CT showing a well-defined homogenous mass in the right lower quadrant (arrow).

The patient subsequently underwent surgery, and complete surgical excision of the mass was achieved. Histopathological examination of the excised mass showed similar findings to the prior biopsy, confirming the diagnosis of ancient schwannoma. The surgery was uncomplicated, with no injuries to the surrounding structures, and the patient was stable on discharge. On follow up 3-months later, the patient is doing fine and there were no signs of recurrence.

Discussion

Ancient schwannomas, first described by Ackerman et al. in 1951, are a rare variety of schwannomas, which are benign tumors that arise from Schwann cells in the peripheral nervous

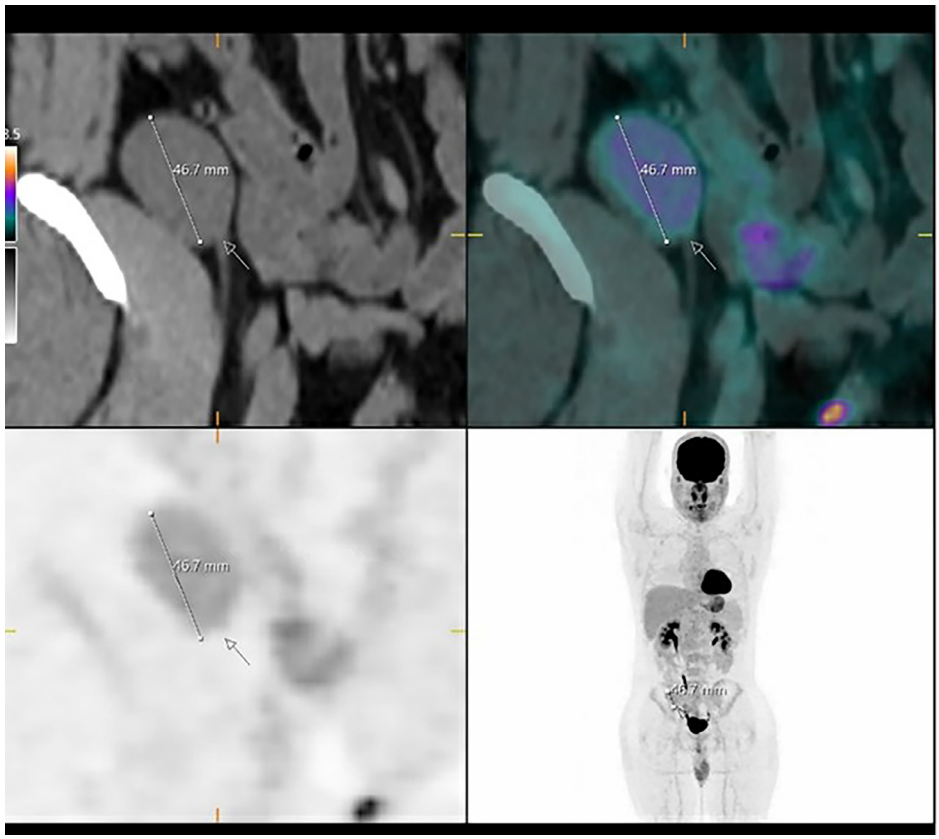

Fig. 2 – PET-CT showing a hypodense mass with mild FDG activity in the right iliac fossa.

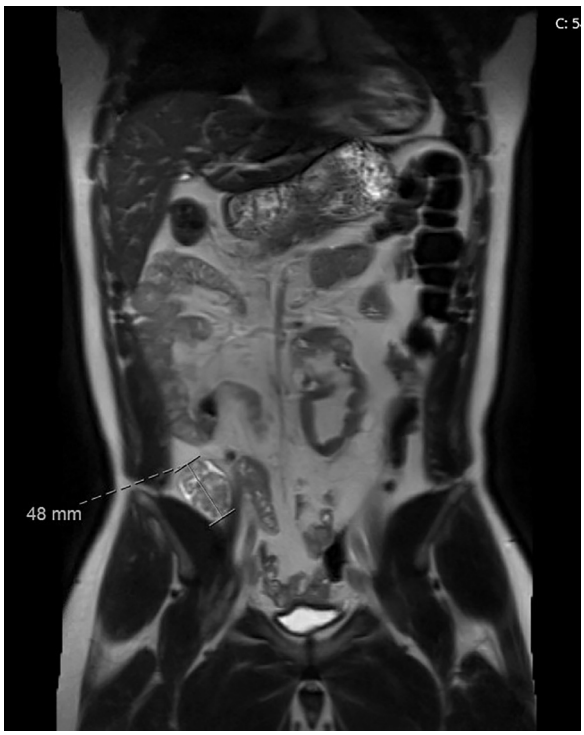

Fig. 3 – Coronal MRI showing a well-defined, hypervascular mass in the right lower quadrant.

system [1,3]. Ancient schwannomas have been reported in various regions of the body, most commonly in the head and neck region [1]. We present a case of ancient schwannoma arising in the iliac fossa, a rare location for this tumor with only 5 reported cases in the literature, summarized in Table 1. Patients with schwannomas are typically asymptomatic but can commonly present with pain and Tinel's sign, which is the sensation of tingling along the distribution of the affected nerve [1,5].

Histologically, ancient schwannomas are characterized by encapsulated regions with atypia and nuclear hyperchromatism, and as they grow larger, they may demonstrate various degenerative changes such as cystic degeneration and hemorrhage, all of which may be confused with malignant neoplasms, such as sarcoma [1,3]. These degenerative changes contribute to the growth and aging of the tumor, hence termed ancient schwannoma [11]. Additionally, ancient schwannomas exhibit areas of high cellularity and low cellularity, termed Antoni A and Antoni B areas, respectively [12]. On immunohistochemistry, ancient schwannomas are positive for S100 [12].

Upon radiological examination with CT, ancient schwannomas appear well-defined with enhancement after IV contrast medium infusion. MRI is the most useful modality for further evaluation of ancient schwannomas. Ordinary schwannomas exhibit hypointensity in T1 and hyperintensity in T2, corresponding to Antoni B areas [5]. Ancient schwannomas, however, exhibit inhomogeneous signal intensity, re-

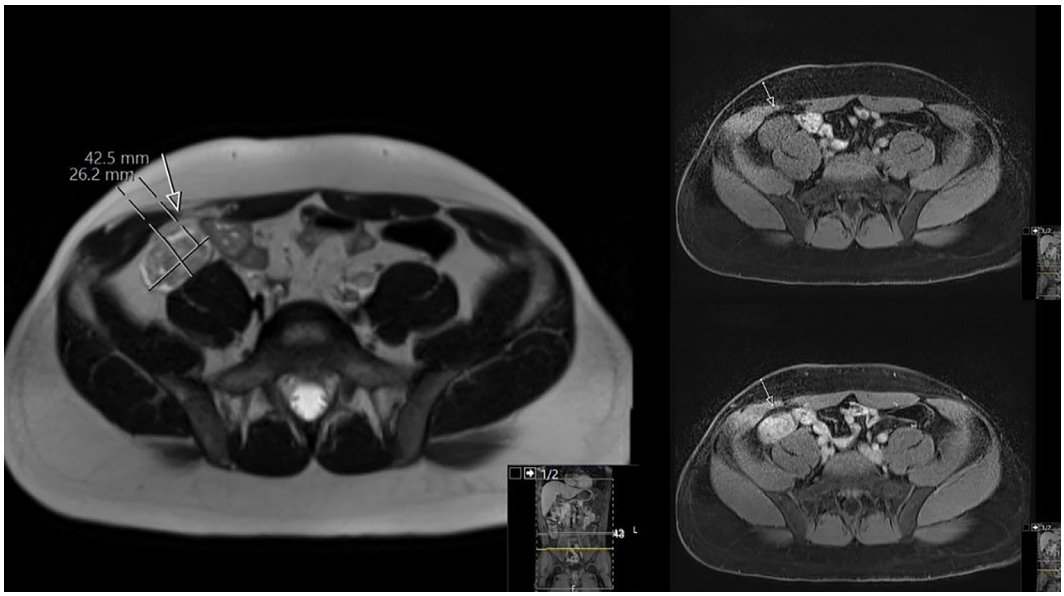

Fig. 4 – Multiparametric axial T2 MRI image (Left image) and T1 MRI diffusion weighted scan precontrast (Right upper image) and postcontrast (Right lower image) showed a 4.8 x 2.6 x 4.3 cm well-defined smooth outlined right lower quadrant mass corresponding to the PET-CT scan observation; which demonstrates thin low T2 signal intensity wall and clustered contiguous low T2 signal intensity component, low T1 signal intensity, lack of signal drop on the opposed phase T1 image, lack of diffusion restriction, and progressive intense fairly homogeneous enhancement; compatible with a neoplastic process.

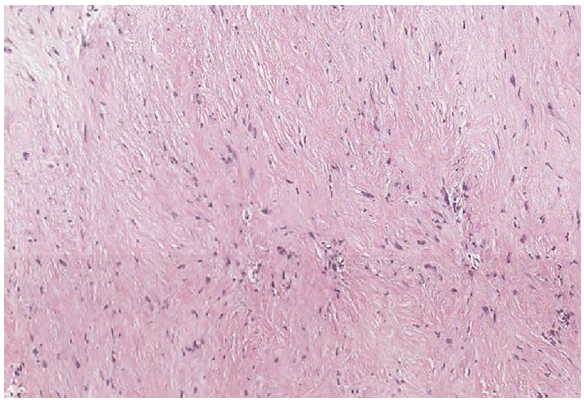

Fig. 5 – A mass composed of spindle cells with a focal nuclear palisading pattern composed of hypercellular areas which express Antoni A fibers and hypocellular areas which express Antoni B fibers.

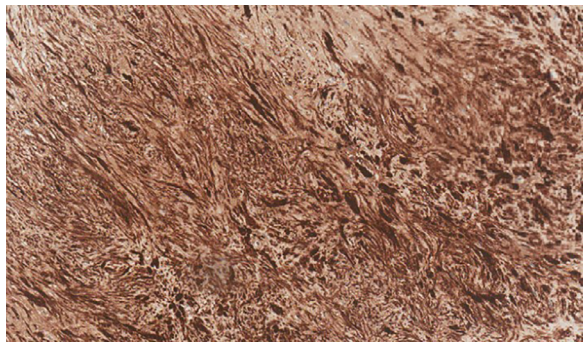

Fig. 6 – Tumor cells display uniform, intense positivity for S100 protein.

flecting the varying amounts of Antoni A and Antoni B areas seen in ancient schwannomas [13]. Scintigraphy is useful in the examination of large neurogenic tumors, as Isobe et al. reported 7 cases of ancient schwannomas that were positive for technetium-99m dimercaptosuccinic acid and negative for ^{67}Ga citrate, which is consistent with the findings of Kobayashi et al. in 11 patients with schwannomas 3 cm or larger [5,14].

Complete surgical excision is the mainstay of treatment and carries a favorable prognosis [8]. Recurrence is the most common complication, often following incomplete excision [15].

Conclusion

Ancient schwannomas, a rare variety of schwannomas, are benign tumors arising from Schwann cells in the peripheral nervous system. They can arise anywhere in the body; however, they are most commonly encountered in the neck. We report a case of ancient schwannoma arising in the iliac fossa, which is an extremely rare location for ancient schwannoma, with only 5 cases reported in the literature. Various imaging modalities can be used to detect ancient schwannomas; how-

ever, MRI is the most effective. Furthermore, histopathologic assessment is a must, and is characterized by degenerative changes and areas of Antoni A and B fibers. Complete surgical excision is the mainstay treatment option and carries an excellent prognosis.

Patient consent

Informed written consent was obtained from the patient for publication of this case report.

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