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Successful treatment of a giant ossified benign mesenteric schwannoma

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Abstract

Primary benign schwannoma of the mesentery is extremely rare. To date, only 9 cases have been reported in the English literature, while mesenteric schwannoma with ossified degeneration has not been reported thus far. In the present study, we present the first giant ossified benign mesenteric schwannoma in a 58-year-

old female. Ultrasound, computed tomography and magnetic resonance imaging were used, but it was still difficult to determine the definitive location and diagnose the mass. By laparotomy, a 10.0 cm × 9.0 cm × 9.0 cm giant mass was found in the mesentery and was then completely resected. Microscopically, the tumour located in the mesentery mainly consisted of spindle-shaped cells with a palisading arrangement. Some areas of the tumour were ossified, and a true metaplastic bone formation was observed, with the presence of bone lamellae and osteoblasts. Immunohistochemical investigation of the tumour located in the mesentery showed that the staining for the S-100 protein was strongly positive, while the stainings of SMA, CD34, CD117 and DOG-1 were negative. The cell proliferation index, measured with Ki67 staining, was less than 3%. Finally, a giant ossified benign mesenteric schwannoma was diagnosed. After surgery, the patient was followed up for a period of 43 mo, during which she remained well, with no evidence of tumour recurrence.

Key words: Schwannoma; Mesentery; Ossification; Laparotomy; S-100

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Core tip: To date, only 9 cases of mesenteric schwannomas have been reported in the English literature; an ossified mesenteric schwannoma has not been reported. In the present study, we present the first giant ossified benign mesenteric schwannoma. It was challenging to determine the location and obtain a precise diagnosis of the mesenteric schwannoma prior to surgery. We completely resected the mesenteric schwannoma by laparotomy. In this paper, a literature review was conducted to deepen our understanding of mesenteric schwannomas.

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INTRODUCTION

Schwannomas are mesenchymal neoplasms of the peripheral nerve sheath^[1], which mainly develop in young to middle-aged patients with no obvious gender difference. Schwannomas are usually solitary sporadic lesions. About 3% occurred in patients with NF-2, 2% in those with schwannomatosis, and 5% in association with multiple meningiomas with or without NF2. Most of schwannomas, whether sporadic or inherited, display inactivating germline mutations of the tumour suppressor gene *NF2* located on chromosome 22 which encodes the protein merlin or schwannomin^[2].

This protein, localized to regions of the cell membrane engaged in cell contact and mobility, is expressed in Schwann cells, meningeal cells and the lens of the eye. The mechanism by which the loss of this protein results in tumorigenesis is not well understood. Schwannomas are usually benign (> 90%) and occupy approximately 5% of benign soft-tissue neoplasms^[3,4]. The tumour can sometimes show secondary degenerative changes, including cyst formation, hyalinization, haemorrhage and calcification^[5]. Schwannomas usually occur in the head, neck and extremities^[6], while schwannomas in the bowel mesentery are extremely rare. To our knowledge, only nine cases of schwannomas located in the bowel mesentery have been reported^[7-15]. In addition, mesenteric schwannoma with ossified degeneration has not been reported thus far. In the present study, we present the first giant ossified benign mesenteric schwannoma.

CASE REPORT

On January 14, 2014, a 58-year-old woman visited our department due to a lesion detected incidentally in her abdominal cavity during a routine health examination in the local hospital. On physical examination, a lesion was palpable in her upper left abdomen, sized 12 cm × 6 cm, and she felt slight tenderness. Four years previously, she had undergone resection of a lipoma in her back. Blood test findings, including tumour markers, were unremarkable. Ultrasound (US) revealed a solid lesion in the upper left abdomen, with clear margin, and also showed some cystic and strong echo areas in the lesion. Color doppler flow imaging (CDFIs) showed blood signals in the lesion. In a native computed tomography (CT) scan, the lesion in the upper left abdomen appeared well-defined and was 9.6 cm in diameter, while regions of high density were visible, compatible with calcification and/or ossification (Figure 1A). Contrast-enhanced CT study revealed a lesion with slight and inhomogeneous enhancement (Figure 1B). On T1-weighted images, the upper left abdominal lesion appeared hypointense, while appeared inhomogeneous and hyperintense on T2-weighted images (Figure 2). According to these imaging results, the abdominal lesion was primarily considered to be a teratoma.

After sufficient pre-surgical preparation, an exploratory laparotomy was performed. We found a giant mass originating from the mesentery and surrounded by a fibrous capsule. The territory of tumour blood supply was the branch of superior mesenteric artery. We carefully separated tissues around the tumour, ligated the vessels supplying the tumour and resected the tumour completely from the mesentery. Intraoperative frozen pathology revealed an ossified mesenteric schwannoma.

Macroscopically, the mass was 10.0 cm × 9.0 cm × 9.0 cm in size and yellowish-white in colour. Microscopically, in the mesenteric tumour, both

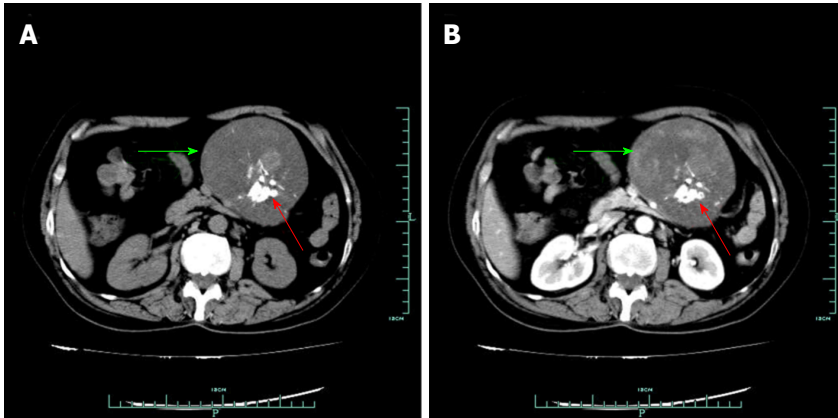


Figure 1 Computed tomography findings. A: In a native CT scan, the mass in the upper left abdomen appeared well defined (green arrow), while regions of high density were visible (red arrow); B: On the contrast-enhanced CT, the mass was inhomogeneous and slightly enhanced, and regions of high density were also visible (green arrow). CT: Computed tomography.

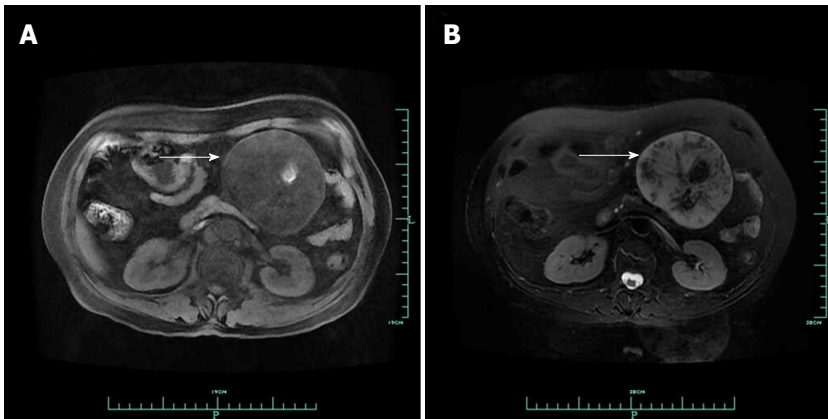


Figure 2 Magnetic resonance imaging findings. A: On MRI, the mass (white arrow) in the upper left abdominal cavity appeared hypointense on T1-weighted images; B: The mass (white arrow) appeared inhomogeneous hyperintense on T2-weighted images. MRI: Magnetic resonance imaging.

hypercellular and hypocellular areas were visible. The tumour mainly consisted of spindle-shaped cells with a palisading arrangement; atypical cells or signs of malignancy were not observed (Figure 3A). Some areas of the tumour were ossified, and a true metaplastic bone formation could be seen, with the presence of bone lamellae and osteoblasts (Figure 3B). Immunohistochemical investigation of the tumour showed a strong positivity for S-100 protein (Figure 4), while SMA, CD34, CD117 and DOG-1 were negative. The cell proliferation index, measured with Ki67 staining, was less than 3%. Finally, a giant benign ossified mesenteric schwannoma was diagnosed. After surgery, the patient recovered smoothly and left the hospital 8 d later. She was followed up for a period of 43 mo, during which she was well, with no evidence of tumour recurrence during the follow-up time.

DISCUSSION

Schwannoma is an encapsulated tumour arising from Schwann cells^[16]. Malignant peripheral sheath nerve

tumours (MPNSTs) are uncommon and are always associated with von Recklinghausen's disease^[17]. Schwannomas mostly develop in young to middle-aged patients^[18] and typically arise in the head, neck, and extremities^[19]. The tumours in the ligament^[20], mesentery^[10] and intra-abdominal organs^[21-23] are extremely rare. Secondary degenerative changes, including cyst formation, calcification, haemorrhage, and hyalinization, can sometimes be shown. However, mesenteric schwannoma with ossified degeneration has not been reported thus far, and to our knowledge, only nine cases of schwannomas in the bowel mesentery have been reported^[7-15]. In the present study, we present a giant ossified benign mesenteric schwannoma. Brief clinical characteristics of ten cases of mesenteric schwannoma in the English-language literature, including our case, are outlined in Table 1 and the summarized clinical characteristics are present in Table 2. The mean age of these patients was 52.89 ± 12.89 years (range 38-80 years), and the male-female ratio was 4:5. Most patients were asymptomatic (70.00%). The mean tumour size was

Table 1 Clinical characteristics of the 10 patients with mesenteric schwannomas

Ref.	Year	Sex/Age	Symptom	Imaging method	Size(cm)	Preoperative diagnosis	Treatment	Histology	Follow-up (mo)	Status
Present case	2017	F/58	Asymptomatic	US, CT, MRI	10 × 9 × 9	Teratoma	Surgery	Benign	43	Survived
Medina-Gallardo <i>et al</i> ^[7]	2017	F/80	Asymptomatic	CT	NA	NA	Laparoscopic operation	Benign	NA	NA
Tepox Padrón <i>et al</i> ^[8]	2017	F/38	Asymptomatic	MRI	11.3 × 8.4 × 4.1	NA	Surgery	Benign	24	Survived
Wang <i>et al</i> ^[8]	2014	M/54	Abdominal pain and hematochezia	CT, Colonoscopy	NA	NA	Ascending, transverse and splenic flexure colectomy	Benign	12	Survived
Tang <i>et al</i> ^[6]	2014	F/43	Mild abdominal pain	CT	4.0 × 4.0 × 1.9	GIST or leiomyoma	Laparoscopic operation	Benign	10	Survived
Lao <i>et al</i> ^[6]	2011	M/45	Asymptomatic	CT, MRI and Angiography	2.2 × 1.7	NA	Surgical excision	Benign	NA	NA
Kilicoglu <i>et al</i> ^[8]	2006	M/56	Nausea, vomiting, abdominal pain, and constipation	US	22 × 19 × 4	Intra-abdominal mass	Surgery	Benign	11	Survived
Minami <i>et al</i> ^[8]	2005	F/54	Asymptomatic	CT, MRI	8.0 × 7.0 × 4.8	Benign solid tumour	Enucleation	Benign	5	Survived
Ramboer <i>et al</i> ^[6]	1998	NA	Asymptomatic	MRI	NA	NA	NA	Benign	NA	NA
Murakami <i>et al</i> ^[8]	1998	M/48	Asymptomatic	United States, CT, MRI	4.5 × 4.0 × 4.0	Benign solid tumour	Laparotomy	Benign	24	Survived

M: Male; F: Female; NA: Not available; US: Ultrasound; CT: Computed tomography; MRI: Magnetic resonance imaging; GIST: Gastrointestinal stromal tumour.

8.86 ± 6.68 cm (range 2-22 cm). All of the tumours were benign.

Obtaining an accurate preoperative diagnosis of a mesenteric schwannoma prior to surgery is nearly impossible. The final diagnosis is based on the histo-pathological examinations of resected specimen^[8]. Microscopically, these tumours present two morphologic features: Antoni type A areas and Antoni type B areas^[24]. Antoni type A areas consist of closely packed spindle cells, with occasional nuclear palisading. Antoni type B areas are hypocellular and contain more mixoid tissue with high water content^[18,25], which can be cystic, haemorrhagic, calcified and even ossified^[5,24]. In the present study, some areas of the tumour were ossified, and a true metaplastic bone formation could be seen, with the presence of bone lamellae and osteoblasts, just as reported by Gurzu *et al*^[26]. Schwannomas show strong immunoreactivity for S-100 protein, while CD34, CD117, DOG-1 and SMA are negative^[27]. In all these cases, immunohistochemical staining is crucial to confirm the differentiation of the tumour cells. In the present study, the tumour located in the mesentery showed a strong positivity for S-100 protein, while SMA, CD34, CD117 and DOG-1 were negative. The cell proliferation index, measured with Ki67 staining, was less than 3%. Ki67 staining is potentially useful in distinguishing benign peripheral nerve sheath tumours from malignant peripheral nerve sheath tumours, particularly in differentiating problematic cases of cellular schwannoma and malignant peripheral nerve sheath tumours. Ki-67 labelling indices ≥ 20% are highly predictive of malignant mesenchymal cells, which show a diffuse growth pattern and, frequently, positivity for is gastrointestinal stromal tumours (GISTs). GISTs are composed of spindled mesenchymal cells, which show a diffuse growth pattern and, frequently, positivity for CD34, CD117 and DOG-1^[29]. Negative immunohistochemical staining for CD34, CD117 and DOG-1 can exclude the possibility of GIST.

Multiple imaging modalities, including US, CT, MRI, are helpful for establishing a probable diagnosis; however, an accurate preoperative diagnosis of the schwannoma is still difficult to obtain^[9]. On US, schwannomas are usually well-defined hypodense lesions^[12]. Plain CT scan shows a well-defined hypodense mass. Cystic, haemorrhagic, calcified and even ossified degeneration sometimes can be seen. On dynamic CT, Antoni A areas of schwannomas are typically well enhanced. Because of the loose stroma and low cellularity, schwannomas with Antoni B areas generally show low density and can be cystic^[9,11]. On T1-weighted images, schwannomas appeared hypointense, while appeared inhomogeneous and hyperintense on T2-weighted images^[14,30]. Celliac angiography is important to determine the arteries supplying the tumour^[11]. Fine-needle aspiration cytology, accompanied by immunohistochemical staining, may play an important role in defining the appropriate treatment procedure and prognosis^[31].

Surgery is curative for mesenteric schwannomas. In the present case, by laparotomy, a giant mass was found that had originated from the mesentery and was surrounded by a fibrous capsule. The patient was followed up for a period of 43 mo, during which she remained well, with no evidence of tumour recurrence.

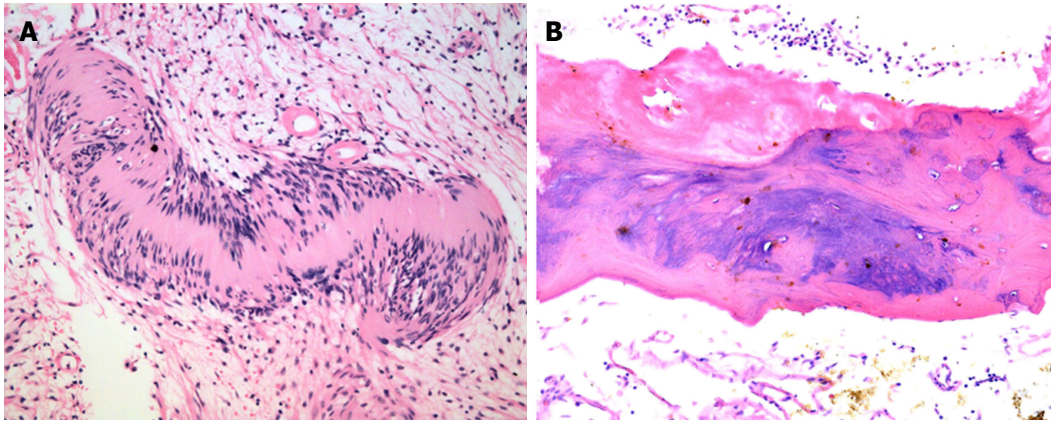


Figure 3 Microscopic examination. A: Microscopically, in the mesenteric tumour, both hypercellular and hypocellular areas were visible. The tumour mainly consisted of spindle-shaped cells with a palisading arrangement; atypical cells or signs of malignancy were not observed (HE $\times 40$); B: Some areas of the tumour were ossified, and a true metaplastic bone formation could be seen, with the presence of bone lamellae and osteoblasts (HE $\times 200$). HE: Hematoxylin and eosin.

Table 2 Summary of clinical data from all 10 cases of mesenteric schwannomas

	<i>n</i> (%) or mean \pm SD (range)
Age (yr) (<i>n</i> = 8)	
Mean	52.89 \pm 12.14 (38-80)
Sex (male/female), (male %) (<i>n</i> = 9)	4/5 (44.44%)
Symptoms (<i>n</i> = 10)	
Asymptomatic	7 (70.00)
Symptomatic	
Abdominal pain	3 (30.00)
Hematochezia	1 (10.00)
Nausea	1 (10.00)
Vomiting	1 (10.00)
Constipation	1 (10.00)
Mean size (cm) (<i>n</i> = 7)	8.86 \pm 6.68 (2-22)
Operation (<i>n</i> = 9)	
Laparotomy	6 (66.67)
Laparoscopic operation	2 (22.22)
Enucleation	1 (11.11)
Histology (<i>n</i> = 10)	
Benign	10 (100.00)
Malignant	0 (0.00)
Follow-up (mo) (<i>n</i> = 8)	18.43 \pm 12.00 (5-43)
Survived	7 (100.00)

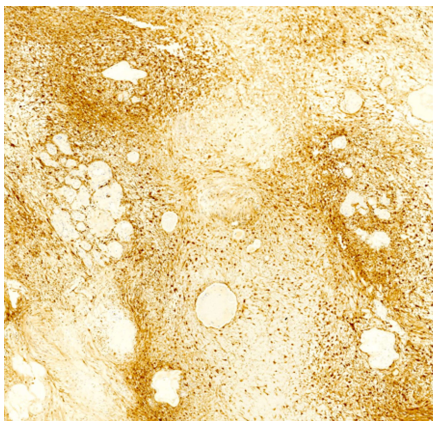


Figure 4 Immunohistochemical staining. The mesenteric tumour was strongly positive for S-100 ($\times 40$).

In conclusion, a schwannoma located in the

mesentery is extremely rare. To our knowledge, our study presents the tenth reported mesenteric schwannoma. In addition, a mesenteric schwannoma with ossified degeneration has not been reported thus far. In the present study, we present the first giant benign mesenteric schwannoma with ossified degeneration. It was difficult to acquire an accurate diagnosis of the mesenteric schwannoma preoperatively, because of the lack of specific symptoms, radiological characteristics and tumour markers.

ARTICLE HIGHLIGHTS

Case characteristics

A 58-year-old woman visited our department due to a lesion detected incidentally in her abdominal cavity during a routine health examination in the local hospital.

Clinical diagnosis

A lesion, palpable in the her upper left abdomen, was sized 12 cm \times 6 cm, and she felt slight tenderness.

Differential diagnosis

Gastrointestinal stromal tumour, intra-abdominal teratoma, sarcoma and neurogenic tumour.

Laboratory diagnosis

Blood test findings, including tumour markers, were unremarkable.

Imaging diagnosis

Ultrasound revealed a solid lesion in the upper left abdomen, with clear margin, and also showed some cystic and strong echo areas in the lesion. Color doppler flow imagings (CDFIs) showed blood signals in the lesion. In a native computed tomography (CT) scan, the lesion in the upper left abdomen appeared well-defined and was 9.6 cm in diameter, while regions of high density were visible, compatible with calcification and/or ossification. Contrast-enhanced CT study revealed a lesion with slight and inhomogeneous enhancement. On T1-weighted images, the upper left abdominal lesion appeared hypointense, while appeared inhomogeneous and hyperintense on T2-weighted images (Figure 2). According to these imaging results, the abdominal lesion was primarily considered to be a teratoma.

Pathological diagnosis

Macroscopically, the mass was 10.0 cm \times 9.0 cm \times 9.0 cm in size and yellowish-

white in colour. Microscopically, in the mesenteric tumour, both hypercellular and hypocellular areas were visible. The tumour mainly consisted of spindle-shaped cells with a palisading arrangement; atypical cells or signs of malignancy were not observed. Some areas of the tumour were ossified, and a true metaplastic bone formation could be seen, with the presence of bone lamellae and osteoblasts. Immunohistochemical investigation of the tumour showed a strong positivity for S-100 protein, while SMA, CD34, CD117 and DOG-1 were negative. The cell proliferation index, measured with Ki67 staining, was less than 3%. Finally, a giant benign ossified mesenteric schwannoma was diagnosed.

Treatment

The authors completely resected the mass located in the mesentery by laparotomy.

Related reports

Schwannomas in the bowel mesentery are extremely rare. To our knowledge, only 9 cases of schwannomas located in the bowel mesentery have been reported. In addition, mesenteric schwannoma with ossified degeneration has not been reported thus far.

Experiences and lessons

In the present study, the authors present the first giant ossified benign mesenteric schwannoma. It was difficult to obtain an accurate diagnosis of the mesenteric schwannoma preoperatively, because of the lack of specific symptoms, radiological characteristics and tumour markers.

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