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ABOUT COVER

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The primary aim of World Journal of Clinical Cases (WJCC, World J Clin Cases) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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CASE REPORT

Primary pulmonary meningioma: A case report and review of the literature

Dan-Bin Zhang, Tao Chen

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Abstract

BACKGROUND

Primary pulmonary meningioma (PPM) is a rare disease that is usually benign. The most common presentation of PPM is isolated pulmonary nodules or masses, so the disease can mimic any other lung tumor on imaging, especially lung cancer or metastasis.

CASE SUMMARY

A 47-year-old asymptomatic woman presented with a well-defined, lobulated pulmonary mass with calcification in the left lower lobe. The mass measured 69 mm × 57 mm × 61 mm and was found during a chest computed tomography (CT) performed for physical examination. Contrast-enhanced CT and positron emission tomography (PET)/CT revealed mild enhancement of the mass, with accumulation of 18-fluoro-2-deoxy-D-glucose (18F-FDG). Transbronchial biopsy suggested a provisional diagnosis of low-grade neuroendocrine tumor. Subsequent enhanced head magnetic resonance imaging revealed no positive lesions. An open cuff resection of the left lower lobe and wedge resection of the lingual segment were performed. Histopathological and immunohistochemical examination revealed that the mass was a PPM.

CONCLUSION

PPM should be considered in the differential diagnosis of isolated pulmonary masses found incidentally on CT and should be diagnosed based on a combination of radiological and histological features. Surgical resection is currently the main treatment strategy. No recurrence of benign PPMs has been reported after complete resection.

Key Words: Primary pulmonary meningioma; Contrast-enhanced computed tomography; Positron emission tomography; Case report

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Core Tip: Primary pulmonary meningioma (PPM) is a rare tumour that usually presents as an asymptomatic solitary pulmonary mass. Limited knowledge of the disease can make diagnosis difficult. Here, we present the case of a 47-year-old woman with PPM.

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INTRODUCTION

Primary ectopic meningiomas are rare tumors that occur in the head, neck, skin, peripheral nerves, bone, retroperitoneum, and lungs. They account for approximately 2% of meningiomas[1,2]. Primary pulmonary meningiomas (PPMs) are rare. Since the first case report in 1982 by Kemnitz et al[3], only 67 cases of PPMs have been reported domestically in the medical literature. Among these cases, only five were malignant meningiomas, and PPMs were more likely to be benign.

PPMs usually appear as isolated pulmonary nodules that are accidentally detected on chest radiographs or computed tomography (CT). Despite advancements in radiological examination such as enhanced CT and positron emission tomography (PET), it remains difficult to assess indeterminate isolated pulmonary nodules or masses, and many benign PPMs are misdiagnosed. The present paper reports a rare case of PPM. We also summarized the clinical imaging characteristics of PPMs in the literature to provide a reference for PPM diagnosis.

CASE PRESENTATION

Chief complaints

A 47-year-old woman had a pulmonary mass on physical examination 1 mo ago.

History of present illness

The patient was hospitalized due to chest CT findings of a pulmonary mass in the left lower lobe of the lung upon physical examination 1 mo prior.

History of past illness

The patient had a free previous medical history.

Personal and family history

The patient had no personal and family history.

Physical examination

Physical examination revealed no obvious positive signs.

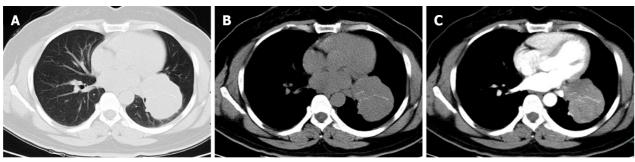
Laboratory examinations

All tumor marker results were within the normal range.

Imaging examinations

Contrast-enhanced chest CT revealed a 6.9 cm diameter mass with a well-circumscribed margin in the left lower lobe of the lung. The adjacent left lower lobar bronchus and lingual segment of the left upper lobar bronchus were compressed by the mass. The lesion was confined to the lung parenchyma and showed striated calcification. After contrast enhancement, the mass showed mild homogeneous enhancement, from a pre-contrast attenuation of 40 HU to a postcontrast attenuation of 60 HU (Figure 1).

On 18-fluoro-2-deoxy-D-glucose (FDG) PET imaging, the standardized uptake value (SUV) of the mass increased unevenly, with a maximum value of 4.4, which suggested malignant lesion (Figure 2). No other lesions were detected on PET/CT. Moreover, enhanced magnetic resonance imaging (MRI) of the brain showed no evidence of intracranial tumors or metastases. Bronchoscopy revealed partial obstruction of the lower left lobe by the mass and narrowing of the lingual opening in the upper left lobe. A subsequent transbronchial biopsy result suggested a low-grade neuroendocrine tumor (Figure 3).



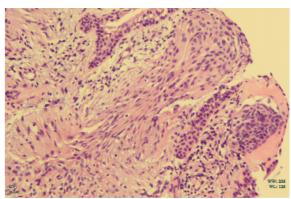
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Figure 1 Contrast-enhanced chest computed tomography images of (A, B) unenhanced and (C) enhanced scan. A 6.9-cm diameter wellcircumscribed mass in the left lower lobe of the lung shows mild homogeneous enhancement.



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Figure 2 Positive uptake by the mass on ¹⁸F-fluorodeoxyglucose-positron emission tomography suggesting malignancy.

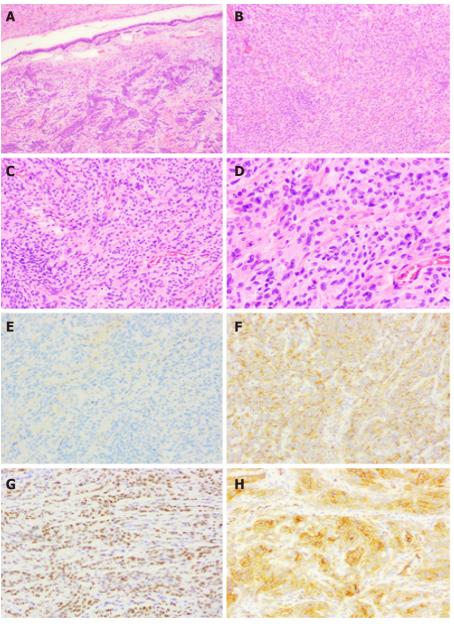


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Figure 3 The transbronchial biopsy result: Hematoxylin and eosin staining showed that a few nested epithelioid cells and abnormal cells were observed in the tissue (200x).

FINAL DIAGNOSIS

The final diagnosis of the presented case was PPM.



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Figure 4 Histological features of primary pulmonary meningioma. A-D: Macroscopically, primary pulmonary meningioma (PPM) showed as spindle or oval cells organized in bundles and whorls on hematoxylin-eosin staining (25x; 50x; 100x; 200x); E-H: Immunohistochemically (200x), PPM showed negativity for E: Cytokeratin, positive for F: Epithelial membrane antigen; G: Progesterone receptor; H: Somatostatin Receptor 2 (SSTR2).

TREATMENT

Considering the CT and PET features of the mass and the results of transbronchial biopsy, an open cuff resection of the left lower lobe and wedge resection of the lingual segment were performed. Gross examination revealed a 6.5 cm, off-white, tenacious texture mass. Microscopic examination revealed a tumor with focal bronchial cartilage involvement, no pleural involvement, and fusiform nests of cells arranged in fascicles or whorls. Immunohistochemistry showed positivity for epithelial membrane antigen (EMA), progesterone receptor (PR), somatostatin receptor 2 (SSTR2), D2-40, and CD34, and negativity for S-100, cytokeratin (CK), glial fibrillary acidic protein, CgA, SOX10, and SMA; the Ki-67 index was about 5%-10% positive (Figure 4). These morphological and immunohistochemical features were suggestive of a PPM. Preoperative contrast-enhanced chest CT, contrast-enhanced brain MRI, and PET-CT did not reveal evidence of intracranial or spinal meningioma.

Tab	Table 1 Patient characteristics					
No.	Ref.	Age (Gender)	Symptom	Size (cm)	Histology	Follow-up
1	Kemnitz et al[3]	59 (F)	Weakness, loss ofappetite, weight loss	4.0	В	30
2	Chumas et al[14]	58 (F)	None	4.0	В	12
3	Zhang et al[15]	58 (F)	None	2.5	В	18
4	Kodama et al[16]	53 (M)	None	2.6	В	84
5	Drlicek et al[17]	41 (M)	None	2.5	В	72
6		62 (F)	None	6.0	В	72
7	Flynn et al[18]	63 (F)	Coughing	3.0	В	44
8		74 (F)	None	1.7	В	37
9	Maiorana et al[19]	68 (M)	None	1.8	В	24
10	Kaleem et al[20]	45 (F)	None	1.2	В	10
11	Lockett et al[21]	65 (M)	None	0.8	В	5
12	Ueno et al[22]	61 (F)	None	0.4-1.5	В	36
13	de Perrot et al[5]	57 (F)	None	0.9	В	30
14	Prayson et al[23]	51 (M)	None	6.5	M	10
15	Spinelli et al[24]	71 (F)	Bronchitis	1.5	В	96
16	Falleni et al[7]	59 (M)	None	2.5	В	30
17	Cesario et al[25]	56 (M)	None	2.0	В	72
18	CURA et al[26]	58 (F)	None	2.0	В	N
19	Comin et al[27]	33 (M)	Hemoptysis and thoracic pain	2.0	В	47
20	Rowsell et al[28]	51 (M)	None	4.0	В	8
21	Picquet et al[9]	54 (F)	None	1.4	В	6
22	Kaneda et al[29]	59 (F)	None	1.4	В	14
23	van der Meij et al[30]	40 (F)	Dyspnea, coughing dysphagia	5.0	M	40
24	Meirelles et al[31]	48 (M)	None	1.5	В	N
25	Incarbone <i>et al</i> [32]	24 (M)	Hemoptysis	2.4	В	42
26	Izumi et al[33]	18 (F)	Hemoptysis on exertion	3.3	В	15
27	Weber et al[4]	108 (F)	Asthenia, lack of appetite, loss of weight and anxiety	15.0	M	N
28	Lepanto et al[10]	60 (F)	None	1.6	В	12
29	Kim et al[34]	61 (F)	Chest pain	2.5	В	84
30	Jiang et al[35]	63 (F)	None	3.5	В	N
31	Juan et al[11]	55 (M)	None	4.5	В	6
32	Oide et al[36]	44 (M)	None	2.0	В	N
33	Huang et al[37]	44 (F)	Chest pain	2.5	В	6
34	Žulpaitė et al[38]	43 (F)	None	4.5	M	24
35	Hong et al[39]	54 (M)	Cough and sputum	1.6	В	24
36	Luo et al[40]	65 (F)	Cough	3.5	В	N
37	Xu et al[41]	65 (F)	Chest pain and tightness	0.7	В	N
38	Ohashi et al[42]	60 (F)	None	2.0	В	36
39	Bae et al[43]	43 (F)	None	1.9	В	26
40	Cimini et al[13]	80 (M)	None	1.4	В	N
41		80 (M)	None	1.2	M	N

42	Wang et al[44]	64 (F)	None	3.4 (cystic nodules 0.8-2)	В	N
43	Fujikawa et al[12]	62 (F)	None	0.8	В	20
45	Han et al[6]	64 (F)	None	0.6	В	28
44		75 (F)	None	0.6	В	2
46	Gürçay et al[45]	55 (F)	Cough	2.0	В	N
47	Jiang et al[8]	70 (M)	None	1.5	В	N
48	Bas <i>et al</i> [46]	57 (M)	Cough	1.0	В	N
49	Oh et al[47]	54 (M)	None	0.5-1.3	В	24
50	Present report	46 (F)	None	6.9	В	3

F: Female; M: Male; None: No symptoms; B: Benign; M: Malignant; N: Not reported.

OUTCOME AND FOLLOW-UP

The patient was disease-free after 3 mo of follow-up.

DISCUSSION

A total of 68 patients diagnosed with PPM were reported in the English literature from 1982 to 2021. All of these patients received histological assessment confirming PPM. Eighteen cases were excluded because (1) They underwent no radiological examination; or (2) They received no radiological evaluation of the CNS negative for meningioma. Ultimately, 50 patients (including the case reported above) were included in the analysis.

Patient characteristics

The study group comprised 50 patients: 19 men and 31 women. The age range was 18-108 years (median age: 58.0 years). Thirty-five patients were asymptomatic and only occasionally showed pulmonary nodules or masses on chest CT or X-ray. Thirteen patients had respiratory symptoms, including chest pain, chest tightness, hemoptysis, cough, and sputum). In addition, two patients had non-specific symptoms[3,4]. There were nine patients with a history of malignancy: two had suffered lung adenocarcinoma[5,6], two colorectal cancer[7,8], two breast cancer[9,10], one buccal cancer[11], one papillary thyroid carcinoma[12], and one thymoma and kidney cancer[13] (Table 1)[14-47].

Radiological characteristics

Most PPMs were benign, and only five cases were malignant [4,13,23,30,38]. Benign PPMs were generally well-circumscribed on radiological studies, with diameters ranging from 0.4 to 6 cm (median: 2 cm). The five malignant PPMs ranged in diameter from 1.5 to 15 cm (median: 6.4 cm). On chest CT scan, benign PPMs usually appear as isolated, rounded, solid, well-defined nodules or masses, with or without lobulation. Five cases were lobulated [24,31,32,37,40], two manifested as ground glass density [11,45], and two showed burrs on the edges[6]. In addition, one recent study reported that the PPM showed multiple cystic lesions with a solid component [44]. The CT features of the lesions were not described in the remaining eight cases (Table 2)[14-47].

The CT enhancement patterns were described in 11 patients: six cases showed homogeneous enhancement, one showed heterogeneous enhancement [39], two showed mild enhancement [37,44], one showed mild concentric enhancement [8], and one showed no significant enhancement [13].

¹⁸F-fluorodeoxyglucose-PET was performed in 12 patients, including our reported case. The PET scans of four patients showed no accumulation of 18F-FDG in lung lesions[8,10,45,46]. Seven patients showed metabolically active lesions suspicious for malignancy, with a reported SUV range from 2.46 to 12.9 in seven cases. No other extra-pulmonary sites with increased FDG uptake were detected in any of the patients.

The prognosis of benign PPM resection is good, with almost no recurrence or metastasis. Follow-up was reported in 35 benign cases, ranging from 2 to 96 mo (median: 24 mo). However, two malignant PPMs relapsed[23,30]. The above summary is presented in Table 3.

Primary ectopic pulmonary meningiomas are very rare, and only 67 cases (including our report) of PPM have been reported in the English language medical literature. The present study reported a case with very complete clinical procedure and imaging data, including preoperative enhanced CT examination, PET-CT examination, bronchoscopy biopsy, and postoperative pathological results. There were rare signs of calcification on CT, false positives on PET-CT and errors in our biopsy results. This

Table 2 Radiological characteristics

	Ref.		CT feature	Enhancement feature	PET/CT
1	Kemnitz et al[3]	RL-P	Well-circumscribed	N	N
2	Chumas et al[14]	RL-P	Well-circumscribed	N	N
3	Zhang et al[15]	LU-P	Well-circumscribed	N	N
4	Kodama et al[16]	LU-P	N	N	N
5	Drlicek et al[17]	LL-N	Well-circumscribed	N	N
6		LL-N	N	N	N
7	Flynn et al[18]	LU-C	Well-circumscribed	N	N
8		LL-P	Well-circumscribed	N	N
9	Maiorana <i>et al</i> [19]	N-P	Well-circumscribed	N	N
10	Kaleem et al[20]	LL-P	Well-circumscribed	N	N
11	Lockett et al[21]	LL-P	Well-circumscribed	N	N
12	Ueno et al[22]	Bil-N	N	N	N
13	de Perrot et al[5]	RL-P	Well-circumscribed	N	N
14	Prayson et al[23]	RU-P	Smooth margins and focal necrosis	N	N
15	Spinelli et al[24]	N-P	Lobulated margins	N	N
16	Falleni et al[7]	LU-P	Well-circumscribed	N	N
17	Cesario et al[25]	LU-P	Well-circumscribed	N	N
18	CURA et al[26]	RU-C	Well-circumscribed	Enhancement	High uptake (no value)
19	Comin et al[27]	LU-P	N	N	N
20	Rowsell et al[28]	RL-C	N	N	N
21	Picquet et al[9]	LL-P	Well-circumscribed	N	N
22	Kaneda et al[29]	N-P	Well-circumscribed	N	N
23	van der Meij <i>et al</i> [30]	RH-C	N	N	N
24	Meirelles et al[31]	RL-C	Lobulated margins	N	High uptake (12.9)
25	Incarbone <i>et al</i> [32]	RU-P	Lobulated margins	N	High uptake (10.14)
26	Izumi et al[33]	LU-C	Well-circumscribed	N	N
27	Weber et al[4]	RL-C	N	N	N
28	Lepanto et al[10]	LL-P	N	N	Low uptake (1.2)
29	Kim et al[34]	RU-P	Well-circumscribed	Homogeneous enhancement enhancement	N
30	Jiang et al[35]	LU-P	Well-circumscribed	N	N
31	Juan et al[11]	LU-P	Ground-glass opacity	N	N
32	Oide et al[36]	LU-P	Well-circumscribed	N	N
33	Huang et al[37]	RL-P	Calcifications, mild peripheral lobulation	Mild enhancement	N
34	Žulpaitė et al[38]	LU-P	N	Homogeneous enhancement	N
35	Hong et al[39]	LU-P	Well-circumscribed	Heterogeneous enhancement	N
36	Luo et al[40]	RL-P	Heterogeneous lobulated	N	N
37	Xu et al[41]	RL-P	Well-circumscribed	N	N
38	Ohashi et al[42]	RL-P	N	N	N

39	Bae et al[43]	RL-C	Oval-shaped	Well-enhancement	Mildly high uptake (2.48)
40	Cimini et al[13]	RU-P	N	No significant enhancement	High uptake (4.63)
41		LU-N	N	Enhancement	Mildly high uptake (2.46)
42	Wang et al[44]	RL-N	Multiple thin-, smooth-walled cysts or cystic nodules with solid component	Mild enhancement	N
43	Fujikawa et al[12]	LL-P	Well-circumscribed	N	N
45	Han et al[6]	RL-P	Burrs on the edges	N	N
44		RL-P	Burrs on the edges	N	N
46	Gürçay et al[45]	RU-P	Peripheral ground-glass	N	Low uptake (1.89)
47	Jiang et al[8]	RL-P	Well-circumscribed	Mild centripetal enhancement	Low uptake (0.6)
48	Bas <i>et al</i> [46]	LL-P	Well-circumscribed	N	Low uptake (no value)
49	Oh <i>et al</i> [47]	scattered	Well-circumscribed	N	Mildly high uptake (3.1)
50	Present report	LL-C	Well-circumscribed	Mild homogeneous enhancement	High uptake (4.4)

 $RL: Right\ lower\ lobe;\ RU:\ Right\ lower\ lobe;\ LL:\ Left\ lower\ lobe;\ P:\ Peripheral\ or\ subpleural;\ C:\ Centrilobar;\ N:\ Not\ reported.$

suggests that we need to be cautious when excluding PPM only through auxiliary examination or even needle biopsy in clinical work.

The pathogenesis of PPMs remains unclear. One hypothesis is that the tumors develop from multipotent mesenchymal cells. Another states that PPMs originate from minute pulmonary meningothelial nodules that are occasionally found in approximately 1% of autopsies and excised lung specimens[48]. However, the incidence of meningiomas is much lower than that of meningeal epithelial nodules. Moreover, previous genotypic comparisons have failed to demonstrate pulmonary meningeal epithelial nodules or intracranial meningiomas, further supporting the hypothesis[49].

To date, approximately 90% of PPMs reported in the literature have been benign, while five have been malignant [4,13,23,30,38]. Most patients with PPM have no obvious symptoms, while some have respiratory or non-specific symptoms. Clinical symptoms may be related to the lesion location. As previously reported, benign PPMs are usually located in the peripheral pulmonary region, with no involvement of the bronchi, blood vessels, or pleura. Some PPM patients have a known history of malignancy[8], so a comprehensive and careful evaluation of pulmonary lesions must be carried out to avoid the misdiagnosis of metastasis.

Radiologically, PPMs usually appear as isolated, solid, and well-defined parenchymal coin-like lesions, ranging in size from 0.4 to 6.5 cm. Approximately 74.0% of PPMs are less than 3 cm in diameter. The lesions may present with burrs, lobulation, ground-glass density, or calcification, but these features are uncommon. Furthermore, one study reported a PPM presenting as multiple cystic lesions[44]. PPMs have diverse enhancement CT manifestations. They usually show different degrees of enhancement, or even no significant enhancement. Hence, the pattern of lesion enhancement may not help to determine whether the lesion is benign or malignant. On 18F-FDG PET, most PPMs exhibit high or mildly high metabolic activity, as in in our reported case. Only four PPMs showed low uptake of ¹⁸F-FDG[8,10,45, 46]. However, one recent study reported a patient with both benign and malignant PPMs, both characterized by increased glucose uptake[13]. This suggests that the malignancy of PPMs may not be related to ¹⁸F-FDG uptake.

Pathological identification is necessary to allow PPM diagnosis; however, diagnosis can sometimes be difficult using needle biopsy alone [32]. False positives are sometimes reported, in addition to negative reports. For instance, in the case reported by Žulpaitė et al [38], a false positive diagnosis of paraganglioma was given based on preoperative transthoracic needle biopsy. The present patient was misdiagnosed as having low-grade neuroendocrine tumor based on preoperative bronchoscopic biopsy.

CONCLUSION

In conclusion, the accurate diagnosis of PPM is challenging because the tumors are rare and show variable radiological manifestations. A single ¹⁸F FDG PET or contrast-enhanced CT examination may not be sufficient to evaluate patients with PPM. Surgical resection is the main treatment strategy, and no relapse has been reported in benign cases after complete resection. In clinical practice, attention should

Table 3 Clinical and image	aina c	characteristics of	nrimary nulmonari	/ meningioma natients

Variables		Number	Ratio (%)
Gender (n = 50)	Female	31	62.0
	Male	19	38.0
Age $(n = 50)$	≤ 40 yr	4	8.0
	40-60 yr	26	52.0
	≥ 60 yr	20	40.0
Symptoms ($n = 50$)	No	35	70.0
	Yes	15	30.0
Size $(n = 50)$	≤3 cm	37	74.0
	> 3 cm	13	26.0
Histology $(n = 50)$	Benign	45	90.0
	Malignant	5	10.0
Site $(n = 47)$	RL	15	31.9
	RU	6	12.8
	LL	10	21.3
	LU	13	27.7
	Other	3	6.4
Location $(n = 45)$	Peripheral	36	80.0
	Centrilobar	9	20.0
Main CT features ($n = 38$)	Well-circumscribed	27	71.1
	Lobulated	5	13.2
	Burrs	2	5.3
	Ground-glass density	2	5.3
	Calcification	2	5.3
PET/CT (n = 12)	High uptake	8	66.7
	Low uptake	4	33.3

RL: Right lower lobe; RU: Right upper lobe; LL: Left lower lobe; LU: Left upper lobe.

be paid to common isolated pulmonary nodule or mass, especially in asymptomatic patients. PPM should be considered in the differential diagnosis of lung diseases.

FOOTNOTES

Author contributions: Zhang DB was responsible for collecting the medical history of the patient and drafting the paper; Chen T reviewed the literature and revised the manuscript; all authors read and approved the final manuscript.

Informed consent statement: Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Conflict-of-interest statement: All authors declare no conflict of interest related to this study.

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