CASE REPORT

Anastomosing haemangioma with fatty changes in the perirenal space: a lesion mimicking liposarcoma

Kishida N, Sentani K, Terada H, Honda Y, Goto K, Hatanaka Y, et al. Anastomosing haemangioma with fatty changes in the perirenal space: a lesion mimicking liposarcoma. BJR Case Rep 2017; 3: 20170022.

CASE PRESENTATION

The patient was a 75-year-old woman without a chief complaint. Abdominal ultrasound for cancer screening revealed a retroperitoneal hypoechoic mass measuring 26 × 22 × 18 mm in the perirenal space, at the level of the left renal upper pole. Subsequently, she was referred to our hospital for examination.

INVESTIGATIONS, IMAGING FINDINGS

Abdominal CT and MRI were performed. The mass was well circumscribed, with fat stranding in the surrounding adipose tissue, and demonstrated heterogeneous enhancement (Figure 1a,b). The ventral side of the mass exhibited avid contrast enhancement equivalent to the renal cortex in the corticomedullary phase, with prolonged enhancement (Figure 2a–c). The ventral side of the mass was heterogeneous and lower in intensity than the cerebrospinal fluid in T2 weighted image (WI) (Figure 2d) and of low intensity in T1WI (Figure 2e). The dorsal side of the mass exhibited low intensity in T1WI and high intensity in T2WI and, although almost no enhancement area was observed, overall, it was believed to be a poorly enhanced cystic structure. Plain CT did not indicate a fat component within the mass. On the dorsal side of the mass, there appeared to be a decrease in intensity from in-phase to out-of-phase in T1WI; however, it was impossible to conclude preoperatively whether fat was present in the mass (Figure 2e, f).

On 18F-fludeoxyglucose positron emission tomography/CT (FDG-PET/CT), minor accumulation was observed in the mass [standardized uptake value maximum (SUVmax) 2.5] (Figure 3). No significant accumulation was observed in other regions.

DIFFERENTIAL DIAGNOSIS

Owing to avid contrast enhancement, paraganglioma was considered in the differential diagnosis; however, on...
I-metaiodobenzylguanidine scintigraphy, significant accumulation was not observed. Therefore, we thought that the possibility of paraganglioma was low.

Dedifferentiated liposarcoma was the probable diagnosis because of the following reasons:

- Liposarcoma occurs with high frequency in the retroperitoneum.
- The mass appeared to be dedifferentiated liposarcoma.
- The surrounding adipose tissue with fat stranding appeared to be well-differentiated liposarcoma.

However, the diagnosis was uncertain because accumulation on FDG-PET/CT was weak in the mass.

Owing to the hypervascular mass, solitary fibrous tumour was added to the differential diagnosis. Schwannoma due to the possibility of including cystic changes, and accessory spleen due to the gradually increasing contrast enhancement effect were considered. However, there were no additional characteristic findings to suggest these were more likely than liposarcoma.

Malignant lymphoma can also sometimes occur in the retroperitoneum; however, primary malignant lymphoma is rare, and usually exhibits strong accumulation on FDG-PET/CT. Therefore, we thought that the possibility of malignant lymphoma was low.

**TREATMENT, OUTCOME**

Although the tumour size remained unchanged at 1-year follow-up, it was impossible to rule out malignancy; therefore, surgery was performed. The perirenal adipose tissue was peeled back from the renal capsule of the upper pole and excised en bloc with the tumour.

Pathologically, a brownish-coloured solid mass including a mixture of dense capillaries and mature adipose tissue was observed (Figure 4a). The pathologist did not identify any features within the tumour, such as necrosis, haemorrhage or cystic changes, which were suspected in the radiological findings. The tumour was composed of an anastomosing proliferation of various-sized capillary vessels that were lined with hobnail endothelial cells (Figure 4b). Mature adipose tissue was also observed. No mitotic...
activity was observed. Similar to the preoperative diagnosis, lipo-
sarcoma was considered pathologically in the differential diag-
nosis. On immunohistochemistry, the tumour cells were positive
to p16 but negative for MDM2 or CDK4. In addition, the
amplification of MDM2 gene was not detected in fluorescence
in situ hybridization. Together with histopathological findings,
we finally diagnosed the tumour as anastomosing haemangioma
with fatty changes.

DISCUSSION
Anastomosing haemangioma is a rare subtype of capillary hae-
mangioma recently defined by Montgomery et al.1 Anastomos-
ing haemangioma presents with anastomosing sinusoidal capillaries in an architecture reminiscent of the splenic
parenchyma. Pathologically, differentiation from angiosarcoma
is problematic; however, anastomosing haemangioma does not
present with mitotic activity, has little to no cellular atypia, and
presents with benign pathological findings.2 Earlier studies have
reported that it often occurs in the urogenital organs, especially
the kidneys.3–5 John et al. stated that the lesion also occurred in
deep soft tissue such as the retroperitoneal adipose and
paraspinal tissues.6

O’Neill et al reported on five cases that were well circumscribed
and hyperdense on plain CT, with avid contrast enhancement
and heterogeneity.2 “Well circumscribed” is a description con-
sistent with well-marginated pathological findings for anasto-
mosing haemangioma, while “hyperdense” is consistent with
haemorrhage in macroscopic findings.1,2,6–9 Heterogeneity
within the mass is believed to be due to fatty components, vascu-
lar thrombi, hyaline globules and cystic changes.1,2

In our case, as in other reports, anastomosing haemangioma was
accompanied by fatty changes, and radiological images revealed
tumour with fat stranding in the surrounding adipose tissue. 
Because it occurred in the retroperitoneum, it was difficult to
differentiate from liposarcoma. In previous reports of anasto-
mosing haemangioma, features were not clearly evident,
although some reported cases exhibited fatty changes. In the
summary of radiological findings, O’Neill et al. reported fatty
changes in 40% (two of five) of cases diagnosed with anastomos-
ing haemangioma. Because of the fatty changes, past reports
have described angiomylipoma7 and liposarcoma2 in the pre-
operative differential diagnoses, with renal and retroperitoneal
occurrence, respectively. No report has summarized a series of
radiological images from a large number of cases, and there is
still no report clearly describing how these fatty changes affect
radiological differentiation. However, it must be recognized that
clinically, retroperitoneal primary anastomosing haemangioma
with fatty changes is difficult to differentiate from liposarcoma,
as demonstrated in our case.

The mass in our case was also well circumscribed, with heteroge-
neity and findings of a gradually increasing contrast enhance-
ment effect, similar to the report by O’Neill et al.7 However, a
preoperative diagnosis could not be reached. Other than

Figure 3. 18F-fluorodeoxyglucose positron emission tomography/CT image of retroperitoneal anastomosing haemangioma. Minor accumulation was observed in the mass [standardized uptake value maximum (SUVmax) 2.5].

Figure 4. Pathological findings of anastomosing haemangioma in the perirenal space. [haematoxylin and eosin staining, original magnification (a, x25; b, x400)]. (a) The mass included a mixture of dense capillaries and mature adipose tissue. (b) The tumour was composed of anastomosing proliferation of various-sized capillary vessels that were lined with hobnail endothelial cells.
liposarcoma, paraganglioma, solitary fibrous tumour due to the comparatively intense staining and schwannoma due to the possibility of including cystic changes could be considered. Owing to the gradually increasing contrast enhancement effect, the differential diagnoses could also include accessory spleen, which may be convincing because of the pathological similarity. Malignant lymphoma may be the differential diagnosis in case of malignancy, or even metastasis with patients who have a history of cancer. For paraganglioma, 123I-metaiodobenzylguanidine scintigraphy is useful for differentiation. If FDG-PET/CT shows strong accumulation or appears to be continuous with other lesions, haemangioma can be ruled out, and a malignant lesion (malignant lymphoma or metastasis if multiple) may be more likely. However, if there is weak accumulation, findings are often non-specific and not useful for differentiation.

Percutaneous biopsy enabled O’Neill et al to diagnose the eight cases that were initially suspected to be anastomosing haemangioma; they reported no complications such as haemorrhage. If anastomosing haemangioma is strongly suspected on the basis of findings such as the site of occurrence, well-circumscribed margins, hyperdensity on plain CT, avid contrast enhancement and fatty or cystic changes, biopsy should be considered as a potential approach to avoid unnecessary surgery.

In conclusion, the anastomosing haemangioma in our case was accompanied by fatty changes with retroperitoneal occurrence; therefore, liposarcoma may be the most likely differential diagnosis. If a retroperitoneal mass is well circumscribed with avid and prolonged contrast enhancement, and includes fatty or cystic changes, anastomosing haemangioma should also be considered in the radiological differential diagnosis. In such cases, biopsy must be considered as a useful option for determining a definitive diagnosis.

**LEARNING POINTS**

1. Anastomosing haemangioma, which often occurs in the retroperitoneum, is well circumscribed, exhibits avid contrast enhancement, and is heterogeneous due to fatty changes; consequently, liposarcoma can be difficult to differentiate.

2. With retroperitoneal tumours accompanied by fatty changes and including a strongly enhanced area, the possibility of anastomosing haemangioma may be considered. Tumours with prolonged enhancement and minor accumulation on FDG-PET/CT may suggest anastomosing haemangioma.

3. Biopsy must be considered as a useful option for determining a definitive diagnosis.

**CONSENT**

Written informed consent for the case to be published (including images, case history and data) was obtained from the patient(s) for publication of this case report, including accompanying images.

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