CASE REPORT

Gastric haemolymphangioma: a literature review and report of one case

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ABSTRACT. Gastric haemolymphangioma is a very rare benign tumour. Pathologically, it is composed of lymphatic vessels and blood vessels. Only a few cases of haemolymphangioma have been reported in the literature so far, all of which developed at sites other than the stomach. The authors believe that a haemolymphangioma occurring in the stomach has not been previously reported. The patient was a healthy 68-year-old male who had felt light epigastric discomfort for 3 months. A CT scan and a gastrofiberscope examination revealed a well-demarcated mass on the posterior wall of the stomach near the lesser curvature. The mass was successfully removed by surgery. During the 18-month follow-up period, the patient was asymptomatic with no recurrence. In this case report, we discuss the imaging findings as well as the pathological features of this unusual case, with a review of the related literature.

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Lymphangioma is a type of congenital malformation of the vascular system which is composed of newly formed lymph spaces and channels and is also called angio-lymphaticum. Usually this type of lesion is thought to be a benign and non-invasive disorder, although invasive lymphangioma has been recently reported. According to the reported pathological findings [1], lymphangiomas can be classified into four categories: capillary lymphangioma, cavernous lymphangioma, cystic lymphangioma (hygroma) and haemolymphangioma (combination of haemangioma and lymphangioma) [2–4]. Haemolymphangioma is a rare benign tumour and gastric involvement is rarely seen. A review of medical literature (online PubMed search) published prior to December 2010 could not find any reports on gastric haemolymphangioma. This is a case report of a gastric haemolymphangioma and we aim to describe the imaging findings of these lesions.

Case report

A 68-year-old male was admitted following 3 months of light epigastric discomfort which became more apparent after meals. The patient did not complain of any other symptoms such as nausea, emesis, acid regurgitation, abdominal pain or black stools. The patient had no history of trauma or surgery. The abdominal examination was negative and no mass was felt. All the laboratory results

were normal; examinations included a routine blood test, faecal occult blood test and a routine urine examination.

A plain CT scan of the upper abdomen showed a well-demarcated and homogeneous mass with lower attenuation on the posterior wall of the stomach near the lesser curvature, with a maximum cross-sectional area of 4.5×3.1 cm (Figure 1a). A contrast-enhanced CT scan was performed with a bolus injection of 90 ml of iohexol (300 mg ml^{-1} of iodine) via an antecubital vein at a flow rate of 2.0 ml s^{-1} . In both the arterial and portal vein phase, the lesion did not show any obvious enhancement, while a slight enhancement was seen on the 2 min delayed scans. The CT value of the mass increased from 14.1–18.3 HU to 15–23.3 HU and the enhancement had a ring-shaped cystic wall appearance. The perigastric fat layer and its space were clear and sharp, and there was no lymphadenopathy detected at neighbouring sites (Figure 1b–d). From the CT scans, a diagnosis of a submucosal benign cystic lesion was made.

A gastrofiberscope examination revealed a 4.0×2.0 cm well-demarcated protruding lesion, which was situated on the posterior wall of the stomach near the lesser curvature. The surface of the mass was smooth, soft and mildly translucent (Figure 2). Under endoscopic ultrasonography (EUS), the lesion was seen as a well-defined and homogeneous hypoechoic submucosal mass (Figure 3). The lesion was diagnosed as a benign submucosal mass in the posterior wall of the stomach.

Under EUS guidance, the lesion was successfully excised and the patient was discharged from hospital 4 days after surgery (Figure 4). During the 18-month follow-up period, the patient was asymptomatic with no recurrence.

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Figure 1. CT findings. (a) A pre-enhanced CT scan shows a well-demarcated and homogeneous lower attenuation mass with the maximum cross-sectional area of 4.5×3.1 cm. (b) Arterial phase: the lesion is not showing any obvious enhancement. (c) Delayed phase: the mass shows slight enhancement, with CT values increasing from 14.1–18.3 HU in the plain and arterial phase to 15–23.3 HU in the delayed phase; the ring-shaped cystic enhancement is clearly seen. No sign of serous membrane involvement can be found. (d) Coronal oblique multiplanar enhanced CT scan shows a slightly enhanced homogeneous mass with low attenuation protruding into the gastric cavity and located on the posterior wall of the stomach near the gastric angle.

Figure 2. Gastrofibrescopic findings. A well-demarcated protruding lesion 4.0×2.0 cm in size on the posterior wall of stomach near the lesser curvature. The surface of the mass was smooth, soft and mildly translucent.

Microscopically, dilated blood vessels and lymphatic vessels of different sizes could be seen in the submucous layer; some vessels had markedly expanded to form the appearance of a cyst. The adjacent layers, *i.e.* the mucosa and muscularis, remained relatively normal (Figure 5). The pathological diagnosis was of gastric haemolymphangioma.

Discussion

Haemolymphangioma is a rare benign tumour that appears to arise from congenital malformations of the vascular system. The occurrence of the tumour may be due to the obstruction of the venolymphatic communication between dysembryoplastic vascular tissue and systemic circulation [2–6]. A review of the literature up to December 2010 (PubMed) found 38 reports concerning this type of tumour, in which tumours had developed in the orbit, tongue, neck, mediastinum, pancreas, spleen, liver,

Figure 3. Endoscopic ultrasonography. A well-defined homogeneous submucosal mass with a smooth border and low echo (black arrow).

adrenal gland, retroperitoneum and extremities. Most of them were manifested as a cystic or cavernous lesion. The diagnosis is usually made at birth or early in childhood, and the most frequent location of the tumour is the cervical region [3–12]. However, there were no reports of tumours in the gastric region. In our case, the patient was a 68-year-old male and the tumour occurred in the gastric submucosal layer and appeared as a cystic lesion.

Clinically, the onset of haemolymphangiomas varies from a slowly growing cyst over a period of years to an aggressive enlarging tumour without invasive behaviour. The size of the tumour seems to vary according to the anatomical location and the relationship with neighbouring tissue. Small tumours are usually superficial, while large tumours are located deeper and have a cystic texture. The most common complications are spontaneous or traumatic haemorrhage, rupture, infection and lymphorrhya. In clinical examinations they usually present as a soft, lobulated and compressible mass [2–7]. In our patient, except for 3 months of light epigastric discomfort, there

Figure 4. Photograph of the localised endoscopic ultrasound guided resection specimen shows the integrated submucosal mass which contains pale yellow fluid.

Figure 5. Pathological examination. Dilated submucosal blood vessels (red arrows) and lymphatic vessels (blue arrows) in different sizes, some of them are inflated and are cystic in appearance (haematoxylin and eosin staining, $\times 100$).

were no abnormal laboratory findings or clinical signs. Histologically, the submucosal mass was composed of various dilated blood and lymphatic vessels; some of them were cystic in appearance.

In our case, a plain CT scan showed a well-demarcated and homogeneous mass with lower attenuation, which was located on the posterior wall of the stomach near the lesser curvature. There was no obvious enhancement in the arterial and portal vein phases on contrast-enhanced CT scans. However, slight enhancement appeared in the delayed phase scans, which might suggest that there are fewer tortuous blood vessels in the lesion. The lesion appeared on EUS as a well-defined homogeneous submucosal mass with a smooth border and low echo. Imaging examinations, including ultrasound, CT and MRI, are useful in confirming the diagnosis, revealing the nature of the tumour and displaying its extension and relationship to the surrounding tissue, which is helpful for planning the surgical strategy and follow up [3]. The differential diagnoses include, for example, gastric haemangioma, lymphangioma, gastric glomus tumour, lipoma and neurofibroma. Needle aspiration or biopsy can also be employed in differentiating haemolymphangioma from other fluid-filled masses. However, the final diagnosis will rely on the pathological findings.

Complete surgical excision is always the treatment of choice for localised haemolymphangiomas because it has the lowest recurrence rate. An important consideration is deciding the optimal time for surgery. Some authors have suggested that the mass should be excised promptly following a confirmed diagnosis, while others believe the tumour size should be regularly observed before surgical excision is definitely needed. In the latter case, there are the potential risks that infection or haemorrhaging may occur, which would make complete surgical excision more difficult. Other clinically available techniques include aspiration together with drainage, cryotherapy, injection of sclerotic agents and radiotherapy. The recurrence rates vary depending on the complexity of the mass, the anatomical location and the adequacy of the excision. Lesions that have been completely excised can have a recurrence rate of 10–27%, while lesions that have

been partially removed can recur in 50–100% of cases [2–12].

Endoscopic submucosal dissection as a new and potentially curative method for gastrointestinal neoplasm may be a safe and effective treatment for small localised benign lesions, and also for early gastric cancer [13–14]. In our case, the gastric haemolymphangioma, which was approximately 4.0 × 2.0 cm in size, successfully excised with EUS, and no complications or recurrence occurred during the 18-month follow-up period.

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