CASE REPORT

Capsular retraction: an uncommon imaging finding in hepatic inflammatory pseudotumour

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ABSTRACT. Capsular retraction is an infrequent but characteristic feature of malignant liver lesions such as hepatic metastases and intrahepatic cholangiocarcinoma. Rarely, this finding may be observed in association with benign lesions, such as atypical haemangiomas. Capsular retraction has not previously been reported in association with hepatic inflammatory pseudotumour (IPT). Hepatic IPT is an uncommon benign hepatic lesion with a good clinical prognosis. In this report, we discuss the case of a 48-year-old woman with capsular retraction secondary to multifocal hepatic inflammatory IPTs.

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Case report

A 48-year-old woman presented with a 2-year history of intermittent right-upper quadrant abdominal pain. She denied any history of fever, chills, diarrhoea or haematochezia. Physical examination of the abdomen was unremarkable. Routine haematological studies were normal. Alanine aminotransferase, alkaline phosphatase and γ -glutamyl transferase levels were mildly elevated. Aspartate aminotransferase, serum albumin, bilirubin and α -fetoprotein levels were within normal limits. Serological tests revealed elevated C-reactive protein and immunoglobulin (Ig)G (elevated IgG subclasses 1 and 2) levels, but were negative for anti-mitochondrial antibody, anti-nuclear antibody, anti-smooth muscle antibody and hepatitis B and C viral markers.

Contrast-enhanced CT of the abdomen revealed multiple homogeneously enhancing lesions distributed throughout the liver. The largest enhancing lesion, measuring 1.9×1.6 cm, was surrounded by a 1 cm zone of relative fat sparing in Segment III in a subcapsular location and was associated with focal concavity of the anterior and inferior surface of the overlying liver (Figure 1). The other lesions ranged from 3–12 mm in diameter. The liver was severely steatotic. Follow-up double contrast-enhanced MRI (Sigma; General Electric Medical Systems, Milwaukee, WI) was performed at 3T to further characterise the lesions. All lesions were hypointense relative to the surrounding liver on T_1 weighted in-phase and T_2 weighted unen-

hanced images. T_1 weighted out-of-phase MR images confirmed the zone of relative fat sparing surrounding the Segment III lesion. Following intravenous infusion of ferumoxides, there was loss of signal on T_2^* weighted images in the centre of the Segment III lesion, indicating ferumoxide uptake (Figure 2). The lesion rim did not lose signal. After administration of a gadolinium-based contrast agent (GBCA), the lesion rim enhanced vividly during the hepatic arterial phase on T_1 weighted images and retained enhancement on delayed images obtained 10 min later. The lesion centre did not enhance after GBCA administration (Figure 3). Enhancing curvilinear structures extended from the anterior and inferior aspects of the lesion to the retracted overlying surfaces of the left lobe (Figure 3). The smaller lesions did not accumulate ferumoxides; however, similar to the largest lesion, they showed vivid enhancement on the arterial phase that persisted on delayed images obtained after 10 min (Figure 4). The main imaging differential diagnoses in our case included abscess, hepatic metastasis and, because of the underlying hepatic steatosis, hepatocellular carcinoma (HCC).

CT-guided liver biopsy of the Segment III lesion was performed. Histologically, fibroinflammatory tissue replaced the normal liver parenchyma. Sheets of plasma cells admixed with lymphocytes and histiocytes, and eosinophils occasionally infiltrated a variably sclerotic stroma composed of interlacing bundles of fibroblasts and collagen fibres (Figure 5). Occasionally, lymphoid follicles were present. Immunostains for CD20 and CD3 showed a mixed population of B- and T-lymphocytes. The plasma cell infiltrate was polytypic for immunoglobulin light chain expression following *in situ* hybridisation. An IgG4 immunostain highlighted a small subset of plasma cells showing positivity for IgG4, estimated at 5%

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Figure 1. (a) Dynamic post-contrast axial CT images in (a) late arterial and (b) hepatic venous phases. Note the three homogeneously enhancing lesions in Segments III and VIII and adjacent to the inferior vena cava (white arrows). The Segment III lesion is associated with capsular retraction along the anterior and inferior surface of the left lobe (black arrows).

of the total population. Overall, the histological and immunohistochemical findings were diagnostic of hepatic IPT.

All lesions were stable on follow-up MRI 6 months later.

Discussion

IPT is characterised histologically by a combination of polyclonal plasma cells, lymphocytes and histiocytes intermingled with bundles of collagenous tissue [1]. Typical sites of IPT include the lung, orbits, intestine, mesentery, spleen and liver [2]. Hepatic IPT has been described in all age groups, with a slight predominance in young adult males. The exact aetiology of IPT is

unclear; possible mechanisms include infection, biliary obstruction, chronic cholangitis and primary sclerosing cholangitis. The clinical presentation can be non-specific, and may include upper abdominal pain, fever, nausea, vomiting and weight loss. Laboratory studies usually reveal leukocytosis, as well as elevated C-reactive protein, IgG and liver transaminase levels. Histologically, hepatic IPTs are classified based on their dominant cellular pattern as xanthogranulomatous type, plasma cell granulomas type or hyalinised sclerosing type [3]. The long-term prognosis of hepatic IPT is good.

The clinical and imaging features of hepatic IPTs may overlap with those of malignant hepatic lesions, which results in a diagnostic dilemma [4, 5]. In our patient, the intermittent right-upper quadrant abdominal pain may

Figure 2. Two-dimensional multi-echo spoiled gradient-recalled echo axial images (a) before and (b) after administration of ferumoxides. Note the focal loss of signal (hypointensity) within the centre of the Segment III lesion on the post-ferumoxide image (b), indicating ferumoxide uptake (white arrow).

Figure 3. Dynamic post-contrast T_1 weighted spoiled gradient echo axial images during arterial and delayed venous phases at 5 min. (a) Arterial enhancing mass lesion (arrow) in Segment III with a central non-enhancing component and a perilesional halo of enhancement. Note the enhancing curvilinear structure (arrows) extending to the anterior liver surface associated with capsular retraction along the anterior and inferior surfaces. (b) On the delayed venous phase, note the persistent ring-like enhancement of this lesion (arrow).

possibly have been secondary to either low-grade hepatic inflammation or capsular involvement and inflammation from IPT. At cross-sectional imaging, hepatic IPTs may manifest as a solitary mass, multiple masses or periportal soft-tissue infiltrative lesions. The MRI appearance of hepatic IPTs is diverse and is possibly related to the variable combination of inflammatory infiltrates, necrosis and fibrosis. These lesions are hypointense on T_1 weighted images. On T_2 weighted images, their signal intensity ranges between hypointensity and mild hyperintensity.

Diverse IPT enhancement patterns have been reported following GBCA administration. Most reported IPTs have been hypovascular, with limited or no appreciable enhancement at the arterial phase, although there has been one report of an IPT with intense arterial enhancement [6]. Peripheral ring-like enhancement with a central non-enhancing core is a commonly reported morphological enhancement pattern [7, 8]; nodular enhancement of the central core has also been described. Other morphological enhancement features may include thick irregular septations and "stalactite" marginal enhancement. There are less published data regarding enhancement after ferumoxide administration. Kato et al [9] reported a case of hepatic IPT with intralesional ferumoxide uptake, and postulated that ferumoxide accumulation may delineate areas of less severe inflammatory and fibrotic infiltration. However, Mortelet et al [6] reported a case of hepatic IPT with no ferumoxide uptake. Thus, there are conflicting reports on the presence of active Kupffer cells within hepatic IPTs.

The patient described in this report presented with multiple hypervascular lesions, the largest of which caused capsular retraction. Capsular retraction adjacent to hepatic tumours is uncommon, with a reported frequency of 2.0–2.8% [10], and is thought to be caused by tumoural necrosis and desmoplastic reaction [11].

This finding is characteristic of malignant hepatic tumours, including untreated and treated lesions, and is only rarely associated with benign tumours, such as atypical haemangiomas [12]. It has not been reported previously with hepatic IPT. In the absence of an underlying tumour, capsular retraction is generally associated with benign processes such as confluent hepatic fibrosis, oriental cholangiohepatitis and bile duct necrosis. In the case reported here, the enhancing curvilinear structures extending from the anterior and inferior surfaces of the Segment III lesion probably represented bands of perilesional fibrosis elicited as part of a desmoplastic reaction. These fibrotic bands caused focal capsular retraction in the overlying liver surfaces.

Although capsular retraction associated with the dominant lesion raised concerns over a malignant aetiology, two imaging features suggested benignity. The first is that all of the lesions showed persistent GBCA enhancement into the late venous phase. Arterial-phase ring enhancement that *fades or washes out* on delayed phases suggests a malignant aetiology; this enhancement pattern is characteristic of metastatic disease and can also be observed in HCC and cholangiocarcinoma. Arterial-phase ring enhancement that *persists* on delayed phases suggests a benign aetiology; this enhancement pattern is characteristic of inflammatory processes, such as pyogenic abscesses, and post-procedural granulation tissue associated with tumour ablation. The second is that the dominant lesion accumulated ferumoxides. Ferumoxide uptake suggests intralesional Kupffer cells and is characteristic of benign solid liver nodules, especially cirrhosis-associated regenerative nodules, cirrhosis-associated dysplastic nodules and some focal nodular hyperplasias. Ferumoxide uptake is uncommon in HCC and rare in metastatic disease; therefore, this finding favoured a benign process.

Figure 4. (a,b) Dynamic post-contrast T_1 weighted spoiled gradient echo axial images during the arterial phase show multiple arterial enhancing lesions (arrows) varying in size from 4 mm to 12 mm in the liver.

Two small series have reported the regression of IPT lesions at follow-up imaging without intervention. Mortelet et al [6] reported normalisation of biochemical tests and reduced size and number of lesions in one patient after 1 year. Yamaguchi et al [13] reported spontaneous regression of IPT in three patients within a 2–12 month interval. In contrast, the lesions reported here remained stable at 6 months, suggesting that short-term follow-up imaging alone may not exclude a slowly growing malignant process. Thus, until the natural history and imaging findings of these lesions are better understood, biopsy may be necessary.

Conclusions

This report illustrates that hepatic IPTs may present with capsular retraction, an imaging feature usually

associated with malignant entities. Although previous histological studies have documented intralesional fibrosis within IPTs, extralesional bands of fibrosis and capsular retraction as an imaging finding of IPT has not been reported previously. Radiologists should be aware that IPTs may manifest with capsular retraction and, accordingly, should consider the possibility of a benign aetiology in their differential diagnosis of this finding. In this case, ancillary features that suggested a benign diagnosis, despite the capsular retraction, included ferumoxide uptake by the dominant lesion as well as persistent enhancement after GBCA administration by all lesions. However, the diagnostic workup still includes a liver biopsy for definitive diagnosis, as the imaging features are non-specific.

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Figure 5. Dense inflammatory infiltrate composed predominantly of plasma cells set in a fibroblastic background (haematoxylin and eosin, $\times 20$).

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