LETTER / Gastrointestinal imaging

Spontaneous and simultaneous regression of multiple hepatic haemangiomas: First case reported

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KEYWORDS
Heaptic haemangioma; Sclerosis; MRI

Benign and asymptomatic, the haemangioma is the most common liver tumour. In the general population, the prevalence is between 1−2% and 20% [1]. Its characteristics are well known in imaging (ultrasound, CT scan and MRI) and, when present, never require a histological verification. It may have unusual aspects such as the giant haemangioma with central cystic degeneration, the rapidly filling haemangioma or calcification [1]. Less frequent, hyalinised haemangiomas (as well as sclerotic or sclerosing haemangiomas) may represent the ultimate stage in the involution of haemangiomas. They are most often isolated and associated with a reduction in size [2]. This article presents a case of multiple haemangiomas where the size and radiological semiology are simultaneously modified.

Case report

In 2008, a 47-year-old woman, without any noteworthy antecedents except for mood disorders treated with aripiprazole (Abilify®) and lithium carbonate (Teralithe®), consulted for abdominal pain without fever. The liver assessment was normal, the serologies for hepatitis A, B and C were negative. An ultrasound examination revealed the presence of three hyperechogenic focal hepatic lesions with posterior reinforcement: two lesions under 4 cm in size, compatible with typical haemangiomas and one 10 cm lesion, compatible with a giant haemangioma.

The MRI carried out immediately detected three formations. The semiology was typical of haemangiomas: well limited, in T1 hypointensity, fluid T2 hyperintensity, with enhancement in discontinuous peripheral lumps at arterial time, followed by progressive filling. A histological sample was not taken due to the presence of several typical haemangiomas in an asymptomatic patient.

In June 2009, a second liver MRI without contrast injection was carried out and revealed the three lesions in typical T2 signal hyperintensity (Fig. 1).

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Figure 1. MRI in 2009: T2 sequence: a: lesion A: a giant haemangioma of the right liver, cystic centre, measuring 94 × 104 mm; b: lesion B: a haemangioma of the hepatic dome measuring 32 × 36 mm; c: lesion C: a small sub-capsular haemangioma of the left liver measuring 21 × 12 mm (arrow).

In November 2011, a third liver MRI (Fig. 2) revealed a change in the aspect of the haemangiomas (Table 1): reduction in the size of all of the lesions (up to −41% between 2009 and 2011), modification of the T2 signal intensity and disappearance of the typical enhancement kinetics for the giant haemangioma.

In view of the typical appearance of the haemangiomas in the first MRI and since there is no risk of degeneration of the haemangiomas [1], a biopsy for aetiological purposes was not taken in this asymptomatic patient. After 12 months, the patient is still asymptomatic.

Discussion

In general, haemangiomas do not increase in size over time [1,2]. Several cases have been reported of haemangiomas with a significant increase in size, generally within

<table>
<thead>
<tr>
<th>Lesion</th>
<th>Size in 2009 (mm)</th>
<th>Size in 2011 (mm)</th>
<th>Variation in the largest diameter 2011/2009 (%)</th>
<th>Signal in T2 (2011 compared with the 2009 MRI)</th>
<th>Enhancement</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>104 × 94</td>
<td>75 × 72</td>
<td>−28</td>
<td>Very heterogeneous: hypo/iso/hypersignal</td>
<td>Periphery: in ring At the centre: heterogeneous, incomplete, centripetal</td>
</tr>
<tr>
<td>B</td>
<td>36 × 33</td>
<td>25 × 18</td>
<td>−31</td>
<td>Reduction in the hypersignal</td>
<td>Typical of haemangioma</td>
</tr>
<tr>
<td>C</td>
<td>22 × 12</td>
<td>13 × 9</td>
<td>−41</td>
<td>Hypersignal stable</td>
<td>Typical of haemangioma</td>
</tr>
</tbody>
</table>
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Figure 2. MRI in 2011: sequences T2, and T1 after gadolinium injection (portal time): a, b: lesion A: reduction in size (75 x 72 mm), heterogeneous T2 signal intensity, incomplete and heterogeneous enhancement; c, d: lesion B: reduction in size (25 x 18 mm), weaker T2 signal hyperintensity than in 2009, typical enhancement of a haemangioma, here in peripheral clumps; e, f: lesion C (arrows): reduction in size (13 x 9 mm), T2 hyperintensity, typical enhancement of a haemangioma with homogenisation at portal time (haemangioma not visible).

A specific hormonal context (pregnancy, estroprogestative contraception) [1,3]. Among the rare cases reported of a decrease in the size of haemangiomas, two of them describe the reduction in the size of a single haemangioma after treatment (VEGF inhibitor [4], chemotherapy [5]), and Doyle reported the reduction in the size of haemangiomas after sclerotic transformation without specifying if this phenomenon is secondary or spontaneous [2]. Other benign liver lesions are likely to regress over time: nodular and focal hyperplasia, without the determination of a formal cause for the reduction in size in the cases reported [6,7]; the haemangioma may decrease or even disappear after the
suspension of estroprogestative pill [8]. As far as we are aware, no cases of spontaneous and simultaneous reduction in the size of several liver haemangiomas have been reported.

In the case of our patient, although the size of the three haemangiomas decreased only the largest, lesion A, presents a major change in its signal in T2. It becomes very heterogeneous, and its enhancement loses its typical characteristics of haemangioma. Lesion C presents a sharp reduction of T2 signal intensity compared with the previous examination. Lesion B maintains typical signal and enhancement kinetics of haemangioma.

The hypothesis of a simultaneous sclerosis of haemangiomas has been put forward. The sclerosis or hyalinisation of haemangiomas represents the final stage in the regressive phenomena that haemangiomas may undergo (necrosis, thrombosis, fibrosis, etc.) [2]. The sclerotic haemangioma then consists of an acellular eosinophilic hyaline substance with rare sclerotic vessels [9]. A series of ten sclerotic haemangiomas prospectively monitored (where only two patients each had two haemangiomas) showed that, in general, they do not comply with the typical imaging of haemangiomas as regards the spontaneous signal intensity and the enhancement kinetics of the lesions [2]. The monitoring demonstrates a decrease in the size of the lesions over time [2].

Although several cases presenting a reduction in the size of liver haemangiomas have been reported, no cases of simultaneous and spontaneous regression of several liver haemangiomas have been described in the literature. Besides the medicinal causes already mentioned, another factor, whether endogenous or exogenous, that may affect or provoke this phenomenon has not been reported. In the case of our patient, a medicinal or hormonal aetiology was searched for in the questioning. Her treatment consists of aripiprazole (Abilify®) and lithium carbonate (Teralithe®). They are not known to have any effect on benign liver tumours. Our patient does not take a hormonal treatment and is not clinically menopaused. In addition, no cases of the sclerosis of haemangiomas have been related to the hormonal status of patients.

Conclusion
The regression in the size of haemangiomas is rare and no cases of the spontaneous and simultaneous reduction in the size of several haemangiomas have been described in the same patient. This exceptional case report indicates that factors are likely to trigger or influence this phenomenon.

Disclosure of interest
The authors declare that they have no conflicts of interest concerning this article.

References