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## Mesenchymal hamartoma of liver in a man: Comparison with cases in infants

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### Abstract

**A 53 year old man with a large mesenchymal hamartoma is reported. Only a few bile ducts could be found in the periphery of the lesion and no hepatocytes were identified within the lesion. As far as is known, this is the only adult male patient reported to date. On the basis of the reported findings of mesenchymal hamartoma in other adults, it is suggested that there could be changes in the morphology of this lesion with age: progressive loss of hepatocytes; degeneration of bile duct epithelium; and cystic change of the mesenchymal component. The haematopoietic element is considered to be part of the fetal hepatic haematopoiesis that occurs in the hamartoma.**

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Mesenchymal hamartoma is a rare lesion of the liver, occurring almost exclusively in young children. It seldom presents beyond the age of 5, the average being 10 to 15 months. Presentation at birth has also been reported. The male to female ratio was found to be 2:1 in one study, but other series have shown an equal sex incidence. It is usually large and discovered as a progressively enlarging abdominal mass. Histologically it consists of myxoid and oedematous connective tissue with dilated vessels, lymphatics, and bile ducts. Clusters of hepatocytes and scattered haematopoietic cells are also often seen.

### Case report

A 53 year old Chinese man was admitted into Queen Mary Hospital, Hong Kong, complaining of dull-aching right upper quadrant pain with fever which he had had for one day. His health had been good. He was found to have an enlarged liver about 10 cm below the xiphisternum. There was tenderness but no guarding or rebound. Laboratory investigations showed no abnormalities. Abdominal ultrasonography and a computed tomography scan showed a 20 × 14 × 10 cm heterogeneous hypodense lesion in the right lobe of the liver with well defined, irregular, curved band-shaped density areas. The caudate lobe was affected and the left lobe was displaced laterally. Contrast injection showed irregular marginal enhancement. Angiography (hepatic, superior mesenteric, and right renal) showed that the mass was hypovascular with no arterio-venous shunting. The main portal vein, inferior vena cava, and the right kidney were displaced but not affected.

A laparotomy was performed and a large soft mass replacing the right lobe was resected. The patient was well two years after the operation with no evidence of recurrence on ultrasound examination.

The resected lobe weighed 1520 g and measured 27 × 15 × 13 cm. There was a well circumscribed tumour measuring 20 cm at its largest diameter. The cut surface showed soft, greyish, oedematous tumour tissue with extensive cystic change. The cysts were filled with clear fluid. There was no necrosis, calcification, or haemorrhage (fig 1).

Histological examination showed loosely

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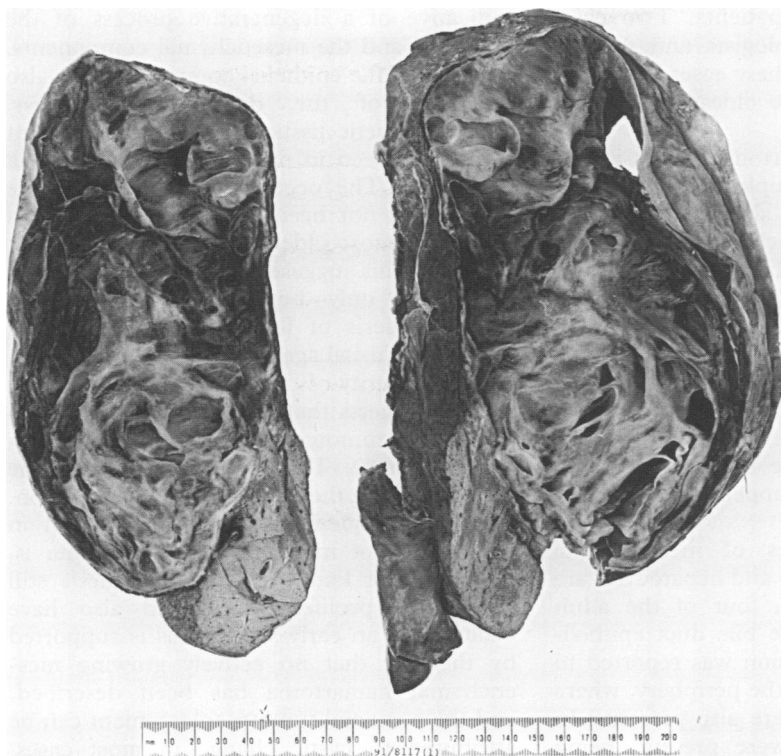


Figure 1 Transected surface of the specimens showing a well circumscribed multicystic tumour.

arranged spindle or stellate cells in a myxoid and vascular background. They had small pyknotic nuclei and scanty cytoplasm with little pleomorphism. Mitotic activity was not seen. There was hyaline thickening around the vascular channels. The myxoid material was rich in hyaluronic acid. The cystic spaces had no endothelial, mesothelial, or epithelial lining. There was a rim of compressed hepatic tissue with regenerative bile ducts, blood vessels, and nerves around the tumour. The adjacent liver parenchyma showed only non-specific changes. Extensive sampling showed no hepatocytes or haematopoietic cells within the tumour. Bile ducts surrounded by fibrous tissue were occasionally found at the periphery (fig 2).

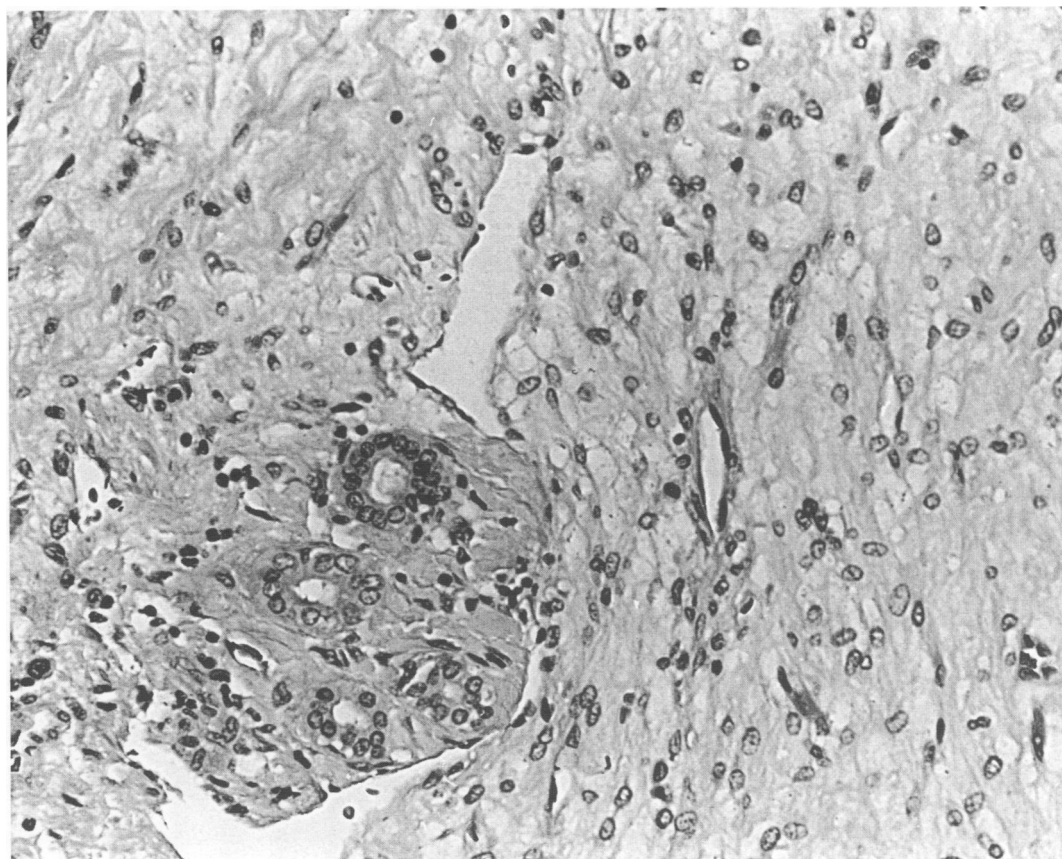
Immunohistochemical staining showed that the spindle and stellate cells were positive for vimentin and negative for desmin, factor VIII, or S-100 protein.

Ultrastructurally, the tumour cells had pinocytotic vesicles, Golgi apparatus, and moderately developed rough endoplasmic reticulum. Granular material and bands of collagen fibrils were present in the extracellular matrix with interspersed small vessels.

#### Discussion

Morphologically, this lesion is identical to a mesenchymal hamartoma, with the absence of hepatocytes and scarcity of bile ducts limited to the periphery. Five cases of mesenchymal hamartoma have been reported in adults.<sup>1-5</sup> Several cases have been reported in

Figure 2 Periphery of the tumour showing mesenchymal cells in a loose matrix with moderate vascularity, and two small bile ducts haematoxylin and eosin.



Japanese<sup>6</sup> and Spanish patients. Two cases were described by radiologists and are not included for review. All these cases have been found in women and the oldest patient was 69.<sup>4</sup>

Mesenchymal hamartoma is widely believed to be a non-neoplastic hamartomatous lesion resulting from abnormal embryonal development of mesenchymal tissue of the portal tracts. Cytogenetic analysis in two cases has shown a balanced translocation between chromosome 19 and 11 or 15. In both cases the breakpoint in chromosome 19 seems to be identical.<sup>7,8</sup> The bile duct epithelium present has been considered to be part of the proliferative process accompanying the mesenchymal tissue. The hepatocytes could either be passively entrapped or aberrantly induced.<sup>9</sup>

In the infantile cases of mesenchymal hamartoma the bile ducts and hepatocytes are active and abundant. In four of the adult cases reported degenerate bile duct epithelium was present. One lesion was reported to have more epithelium at the periphery, where islands of hepatocytes were also present.<sup>3</sup> In two cases hepatocytes were present, being confined to the periphery in one case<sup>1</sup> and distributed randomly in another.<sup>2</sup> Hepatocytes were absent in another case.<sup>4</sup> In our case there were only small bile ducts at the periphery and a complete absence of hepatocytes. We are not certain whether these peripheral bile ducts were incorporated from the adjacent compressed liver or were part of the tumour. The decrease in non-mesenchymal elements is a major difference in morphology between mesenchymal hamartomas found in infants and those found in adults. The fifth case was rather unusual, with large spaces lined by well formed mucous epithelium in a mesenchymal stroma.<sup>5</sup> Such well formed epithelium lining large glandular spaces has not been seen in the other four cases, nor in ours. Differentiation from bile duct adenoma with mesenchymal stroma would be very difficult.

Another major reported difference is that mesenchymal hamartoma in older patients is usually more cystic and in younger patients it's more solid.<sup>10</sup> Our case conforms to this pattern and shows extensive cystic change. This is also true for the other reported adult cases. This myxomatous and cystic change is

indicative of a degenerative process of the epithelial and the mesenchymal components. The loss of the epithelial component may also be part of the degenerative process. Haematopoietic tissue is another component sometimes seen in mesenchymal hamartoma in infants. The occurrence in different age groups has not been well studied, but it is absent in most older children and all adult patients. This suggests that the haematopoietic cells may only be part of the hepatic haematopoiesis of fetal liver which persists beyond the usual age.

The maturity of the mesenchyme in all age groups suggests that the maximum proliferation of this tumour has already taken place at the fetal stage. This is in keeping with the postulation on the histogenesis of this lesion. The time sequence for the proliferation in adult cases of mesenchymal hamartoma is, however, not known. If the hypothesis still holds, the proliferation should also have occurred at an early stage. This is supported by the fact that no actively growing mesenchymal hamartoma has been described, and the postnatal tumour enlargement can be attributed to cystic change in most cases. Interestingly in the 69 year old patient, abdominal surgery had been performed 20 years before and no liver tumour had been noticed.<sup>4</sup>

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