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Perinatal evolution of mesenchymal hamartoma of the chest wall

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Abstract Mesenchymal hamartoma of the chest wall (MHCW) is a rare condition. Previously, surgical resection has been advocated with considerable post-operative morbidity. Evidence for conservative management is lacking because the natural history of MHCW is unknown. We present serial measurements of an antenatally detected MHCW (8 antenatal ultrasounds and 2 postnatal computed tomographic scans). The study demonstrates that the relative tumor size peaked at birth and then decreased postnatally. Based on this evidence, we believe that MHCW can be managed conservatively in an asymptomatic patient.

Chest wall lesions in children are not common. Mesenchymal hamartoma of the chest wall (MHCW) is an extremely rare but benign lesion. Most of MHCW have been previously managed with radical surgical resection to minimize potential compression on the lung tissue. This resulted in severe chest deformity and occasional mortality [1-3]. The role of conservative management is uncertain because the natural history of the lesion is unknown. We report a case of antenatally detected MHCW that showed evidence of involution on computed tomographic (CT) imaging and serial ultrasound studies in the first 6 months of life.

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1. Case report

A 34-year-old woman with a history of insulin dependant gestational diabetes underwent routine prenatal ultrasound at 24 weeks of pregnancy. A right chest mass was shown in her male fetus (Fig. 1A). Serial ultrasound imaging showed that the mass (volume = height × width × depth × 0.52) grew at a rate faster than the body growth (abdominal circumference), especially during the last two months of the pregnancy (Fig. 2). Planned Cesarean section at 38 + 3 weeks was carried out. At birth, the deformity of the chest contour was subtle and there was no associated abnormalities. The baby had minimal respiratory symptom and was able to breath unaided on room air.

A postnatal (Day 1) CT of the chest showed a multifocal tumor, originating from the right fifth to seventh ribs.

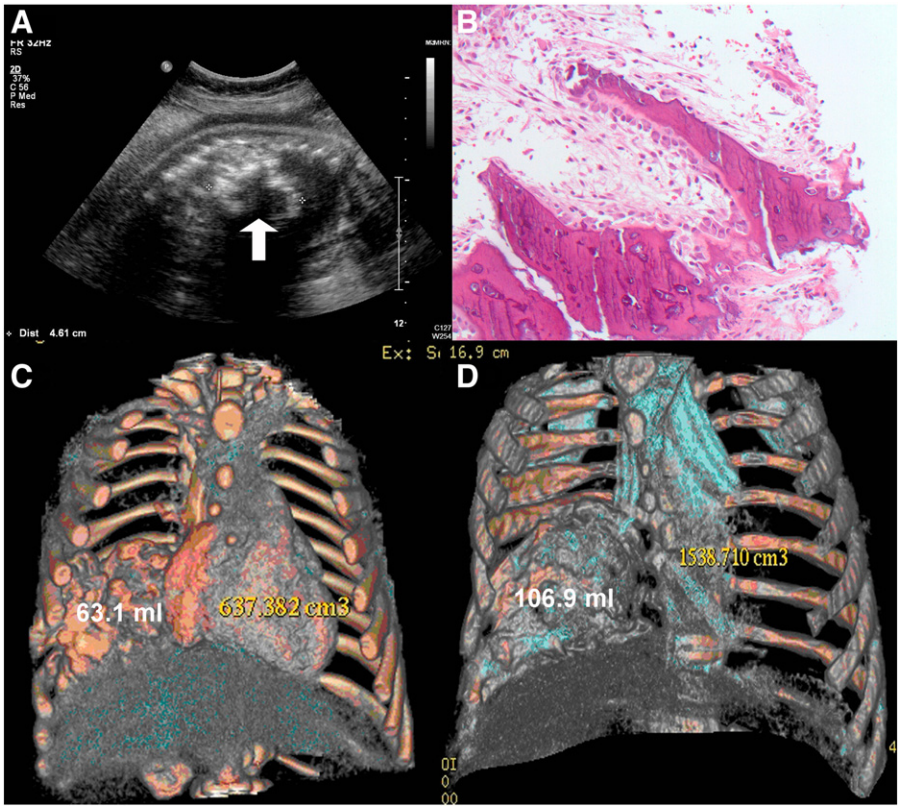

Fig. 1 A, Antenatal ultrasound at the 24th week of gestation demonstrating the intrathoracic mass in the right chest (white arrow). B, Histology of needle biopsy of MHCW. Three dimensional reconstructions of chest CT images at birth (C) and 6 months (D) of age. White numbers: tumor volume in milliliters. Yellow numbers: thoracic volume in milliliters.

Moulding and erosion of these ribs were also noticed. Three smaller lesions from the ninth, 11th and 12th ribs were also noted. The image was compatible with the diagnosis of MHCW (Fig. 1C and D). Urine vanilylmandellic acid was normal. An MIBG scan of the tumor was negative. On day 7 of life, the infant underwent a CT-guided biopsy of the lesion. Histology of the specimen showed epithelioid cell infiltrates with condensed chromatin and abundant eosinophilic cytoplasm. This was accompanied by prominent myxoid stroma rich in hyalinized collagenous mesh and scattered trabeculae of bone. Multinucleated giant cells also accumulated at the sites of heavy calcification. The microscopic findings were consistent with mesenchymal hamartoma of chest wall (Fig. 1B).

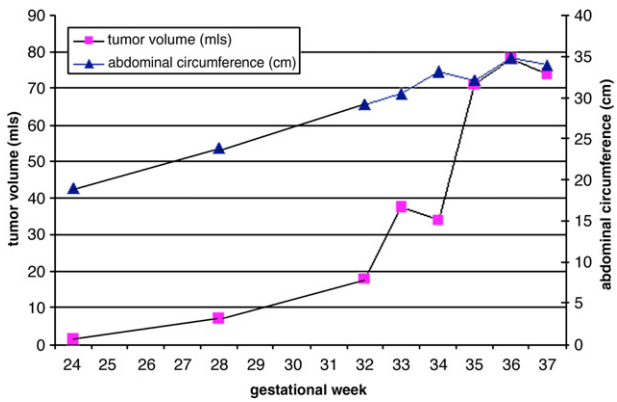

Fig. 2 The serial antenatal ultrasound measurements of the fetal abdominal circumference and the MHCW volume.

Computed tomographic scan was repeated at 6 months of age (Fig. 1D). Compared to the postnatal CT, although the tumor volume per se increased by 69% (from 63.1 to 106.9 mL) during the first 6 months of life, the thoracic volume, spleen volume and body weight increased by 141%, 450% and 105%, respectively, in the same period. As a proportion to the references (ie, thoracic volume, spleen volume and body weight), the tumor size decreased by 30%, 69%, and 17% respectively (Table 1). Up to the time of this report (9 months), the patient has no respiratory symptom. We are currently continuing the conservative management.

2. Discussion

We have documented a case of antenatally detected mesenchymal hamartoma of the chest which showed radiological evidence of postnatal involution in the first 6 months of life.

Chest wall hamartomas are extremely rare. Its incidence is estimated to be 1 in 3000 among primary bone tumors [4] or