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Cardiac arrest in infant due to giant fetus-in-fetu

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A 7-month-old boy was referred to our institution following a cardiac arrest which was treated by timely cardiopulmonary resuscitation at another hospital. Previously, he had a progressive cough with a low-grade fever despite the use of antibiotics for 4 days. He was transferred to our institution for further investigation. He had no other associated diseases, nor use of medication. His mother did not report any abnormal findings from ultrasonography during prenatal examinations. Auscultation revealed shallow respiration with moist rales in the left lung. The echocardiogram revealed a 3.8 mm atrial septal defect with the heart shifted to the right. CT scan indicated a giant tumour in the posterior mediastinum (figure 1A, B). Serum alpha-fetoprotein (AFP) and β -chorionic gonadotropin (HCG) were negative. To exclude malignant conditions where chemotherapy would be warranted, a mini-thoracotomy for biopsy was performed. However, the biopsy indicated tissues of fibre, fat and bone without evidence of malignancy. A definite pathological diagnosis could not be determined due to limited specimens.

Subsequently, an emergency resection of the mediastinal tumour to alleviate the compression was performed. Through a left thoracotomy, a giant 18×14 cm tumour, extending to the right thoracic cavity was grossly identified in the posterosuperior mediastinum. The tumour was connected to the host twin with three main vessels originating from the thoracic aorta. Gross anatomy of the resected tumour revealed an axial skeleton with symmetric arrangement of tissues resembling muscles and

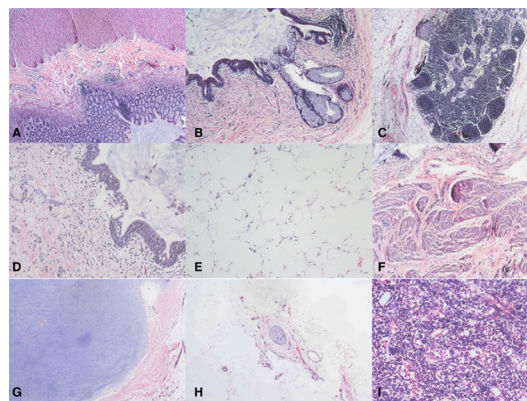


Figure 2 Pathological findings of resected tumour. (A) Intestinal tissue with glands (H&E staining, 40×). (B) Digestive glands (H&E staining, 100×). (C) Lymph nodes (H&E staining, 100×). (D) Bronchial tissue (H&E staining, 200×). (E) Adipose tissue (H&E staining, 100×). (F) Skeletal muscles (H&E staining, 100×). (G) Cartilage (H&E staining, 40×). (H) Neural fibrils (H&E staining, 40×). (I) Bone marrow (H&E staining, 200×).

intestines. (figure 1C, D). Pathological findings revealed well-differentiated tissues of fat, bronchus, cartilage, bone marrow, peripheral nerves, skeletal muscle and intestines from all three germ layers (figure 2). The diagnosis of fetus-in-fetu (FIF) was eventually established based on the presence of an axial skeleton with a symmetric arrangement of well-differentiated organ tissues. Eventually, the patient was discharged with a nearly normal chest CT scan.

DISCUSSION

FIF is a rare congenital anomaly with an overall incidence of 1 in 500 000 live births.¹ FIF is thought to be a consequence of an abnormal mono-chorionic diamniotic pregnancy where the parasitic twin is located within the body of its host twin.² FIF is commonly found in the abdomen, and this was the first case with mediastinal FIF leading to cardiac arrest. The patient's cardiac arrest was possibly caused by the overwhelming vagus reflex due to the significant mediastinal shift, traction or compression by the FIF. The main challenge in this case was how to differentiate FIF from a highly differentiated teratoma. This patient was ultimately diagnosed with FIF due to the presence of an axial skeleton with a symmetric arrangement of well-differentiated organs.² FIF is a benign condition where complete resection is promising and curative. However, malignant transformation of FIF has also been reported.³ Therefore, a regular follow-up with serum AFP and β -HCG is recommended, especially in cases with immature tissue components. Surgeons should have a high index of suspicion for FIF if a

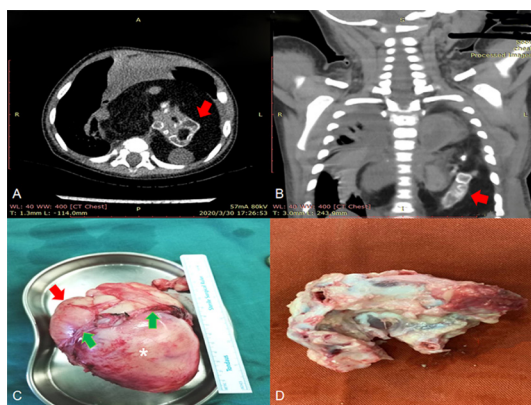


Figure 1 Chest CT and gross anatomy of resected mass. (A) and (B) indicate a giant tumour in the posterior mediastinum with spinal column structures (red arrow), fat and cystic densities. (C) Gross anatomy of resected tumour is anencephalic and acardiac with structures that resemble a diaphragm (green arrow) and lungs (red arrow). (D) An axial skeleton is found inside the tumour (asterisk in C) with symmetrical arrangement of other organs.



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mass containing an axial skeleton with well-differentiated organs is encountered.

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