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Prenatal ultrasound diagnosis of congenital infantile fibrosarcoma and congenital hemangioma: A case report

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Abstract

BACKGROUND

Congenital infantile fibrosarcoma (CIF) and congenital hemangioma (CH) have similarities on prenatal ultrasound and are rare.

CASE SUMMARY

We report 3 cases of fetuses with superficial hypervascular tumors, confirmed by postnatal pathology as CIF (1 case) and CH (2 cases, including 1 in a twin fetus). In Case 1, a mass with rich blood supply in the fetal axilla was discovered by prenatal ultrasound at 28+0 weeks (wk) of gestation. The postpartum pathological diagnosis was CIF, the mass was surgically removed, and the prognosis of the child was good. In Case 2, at 23+1 wk of gestation, a mass was discovered at the base of the fetus's thigh on prenatal ultrasound. The postpartum pathological diagnosis was CH. After conservative treatment, the mass shrank significantly. Case 3 occurred in twin fetus. At 30+0 wk of gestation, prenatal ultrasound revealed a bulging mass with rich blood supply on the abdominal wall of one of the fetuses. Three weeks later, the affected fetus died, and the unaffected baby was successfully delivered by emergency cesarean section. The affected fetus was pathologically diagnosed with CH.

CONCLUSION

Prenatal ultrasound can provide accurate information, such as the location, size and blood supply of a surface mass in a fetus. We found similarities between CIF and CH in prenatal ultrasound findings. Although it is difficult to distinguish these conditions by prenatal ultrasound alone, for superficial hypervascular tumors that rapidly increase in size in a short period, close ultrasound monitoring of the fetus is required to quickly address possible adverse outcomes.

INTRODUCTION

Congenital infantile fibrosarcoma (CIF) and congenital hemangioma (CH) are two kinds of hypervascular tumors in newborns and infants that may involve the body surface. The incidence rate of soft tissue sarcomas in the first year of life was reported to be 16.0 per million[1], and one prospective study reported CH in 2/594 newborns (0.3%)[2]. Most current research focuses on infants and children[3,4].Fetal CH is relatively common in existing related reports, while there are few reports on CIF, another extremely rare fetal tumor. Through a literature review, only 6 cases with both prenatal ultrasonography and confirmation by immunochemistry and specific fusion genes after delivery were reported. Both CIF and CH can manifest as body surface masses with rich blood supplies on prenatal ultrasound, which brings challenges to prenatal diagnosis. Given the rarity of cases detected prenatally and confirmed as CIF or CH postnatally, this paper summarizes the cases of fetal CIF and CH discovered by prenatal ultrasound in our hospital. One case of CIF and two cases of CH, including one case in a twin fetus, provide experience in prenatal counseling and perinatal management for such tumors discovered prenatally.

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CASE PRESENTATION

Chief complaints

Case 1: A 31-year-old Chinese singleton primigravida (gravida 1 para 0) at 34+5 wk of gestation was admitted to our hospital due to abnormalities shown on fetal imaging examination 6 wk previously.

Case 2: A 29-year-old Chinese singleton primigravida (gravida 1 para 0) at 23+1 wk of gestation presented to our hospital for routine follow-up.

Case 3: A 32-year-old Chinese twin pregnant woman (gravida 2 para 1) at 30+0 wk of gestation was admitted to our hospital due to abnormalities shown on fetal imaging examination 2 wk previously.

History of present illness

Case 1: The patient was admitted to Xijing Hospital for routine follow-up at 28+0 wk of gestation. Fetal imaging examinations revealed a mass in the left axilla and the left chest wall of the fetus. The pregnant woman had been followed up by ultrasound and had not received any treatment.

Case 2: The patient had no history of present disease.

Case 3: The patient was admitted to Xijing Hospital for routine follow-up at 28+3 wk of gestation. Fetal imaging examination showed a mass protruding from the abdominal wall of one of the twins. The affected fetus was alive without pleural or peritoneal effusions. No abnormalities were found in the other twin fetus. The pregnant woman had been followed up by ultrasound and had not received any treatment.

History of past illness

Case 1: The woman had no history of past disease.

Case 2: The woman had no history of past disease.

Case 3: The woman delivered her first child vaginally 5 years ago.

Personal and family history

Cases 1-3: The personal and family histories of all patients were unremarkable.

Physical examination

Case 1: The pregnant woman's physical examination was normal. Physical examination of the newborn revealed a mass of approximately 7.0 × 5.0 cm from the left axilla to the left chest wall. The surface of the mass was purplish red and hyperemic, without ulceration or hemorrhage. The mass was tough and unsmooth, and the fetus had poor mobility and tenderness.

Cases 2: The pregnant woman's physical examination was normal. Physical examination of the newborn revealed a purplish red mass of approximately 7.0 × 6.0 cm at the root of the right thigh, with unclear borders, tough quality, and no rupture on the surface. The mobility of the right lower limb was poor.

Case 3: The pregnant woman's physical examination was normal. The affected fetus was stillborn with an enormous mass in the thoracic and abdominal wall. The vital signs of the other fetus were stable.

Laboratory examinations

Case 1: Laboratory examinations of the pregnant woman showed no abnormalities. The newborn's postpartum laboratory examination results for the platelet count, D-dimer level, prothrombin time and fibrinogen level, which were 52 × 10^9/L, 20360.00 ng/mL, 15.1 s and 0.33 g/L, respectively, were abnormal.

Case 2: The pregnant woman's laboratory examinations showed no abnormalities. The newborn's postpartum laboratory examination results for the hemoglobin concentration, platelet count and D-dimer and fibrinogen level, which were 85 g/L, 25 × 10^9/L, 78930.00 ng/mL and 0.84 g/L, respectively, were abnarmal.

Case 3: The pregnant woman's laboratory examinations showed no abnormalities.

Imaging examinations

Case 1: The prenatal ultrasound images at 34+5 wk of gestation showed a well-defined, heterogeneous soft tissue mass, 5.5 × 4.5 cm in size, from the left axilla to the left chest wall of the fetus. The mass mainly comprised isoechoic solid components. Color Doppler flow imaging (CDFI) revealed abundant blood flow within the mass, with a dendritic distribution (Figures 1A and 1B). Prenatal magnetic resonance imaging (MRI) revealed an irregular soft tissue mass in the fetal axilla on the T2 weighted image (Figure 1C). The size of the lesion was more extensive on postnatal ultrasound than on prenatal ultrasound, and the lesion had increased arterial blood flow. Pulse Doppler flow imaging showed low-resistance blood flow within the mass (resistance index, RI was 0.19) (Figure 2A and 2B). X-ray showed a vast basal arc-shaped soft tissue density shadow in the left axilla and lateral chest wall. The boundary with the chest wall tissue was unclear, and the adjacent ribs were compressed, deformed, and displaced inward (Figure 2C).

Case 2: A well-defined, heterogeneous mass (2.6 × 1.4 cm) was found at the root of the right thigh of the fetus on ultrasonography at 23+1 wk of gestation. CDFI showed sparse punctate blood flow around the mass (Figure 4).

Case 3: Prenatal ultrasound at 30+0 wk of gestation showed that in one of the twins, a heterogeneous mass (7.1 × 4.9 cm) protruded from the subcutaneous layer of the thoracic and abdominal wall to the body surface with a clear boundary, complete

capsule, smooth surface, and regular shape. The boundary between the mass and the abdominal wall was unclear. CDFI showed abundant blood flow within the mass, with a dendritic distribution. (Figure 5). Prenatal ultrasound at 33+0 wk of gestation showed that the affected fetus had no heartbeat, and the volume of the mass in the thoracic and abdominal wall had increased significantly, to 9.6 × 5.5 cm. Compared to the previous examination, the internal echoes of the mass were more chaotic. The mass had a clear boundary and regular shape, and no apparent defect was found in the abdominal wall. CDFI showed no blood flow in the mass. At the same time, the whole body of the fetus showed edema, and the thickness of the scalp was approximately 6 mm. Effusion was found in the pleural cavity and pericardial cavity of the affected fetus, accompanied by unclear amniotic fluid (Figure 6).

FINAL DIAGNOSIS

Case 1: Congenital infantile fibrosarcoma (CIF).

Case 2 and Case 3: Congenital hemangioma (CH).

TREATMENT

Case 1: The patient delivered a baby boy by cesarean section at 37+0 wk of gestation. Owing to the detection of thrombocytopenia and abnormal coagulation function, platelets, human fibrinogen and plasma were administered. After correcting coagulation dysfunction, surgical resection of the tumor was performed on Day 50.

Case 2: The patient delivered a baby boy at 40+0 wk of gestation. Owing to the detection of anemia, thrombocytopenia and abnormal coagulation function, cell suspensions and platelets were administered. Oral prednisone and propranolol were administered and were adjusted based on the changes in the CH. In addition, the tumor was closely monitored.

Case 3: After urgently promoting fetal lung maturity, the pregnant woman delivered two fetuses by cesarean section, one of which was stillborn and had a considerable mass in the thoracic and abdominal wall. The vital signs of the other fetus were stable.

OUTCOME AND FOLLOW-UP

Case 1: After the operation, the child recovered well, and the tumor had not recurred at 12 mo of follow-up.

Case 2: The mass on the right thigh became softer and smaller in size. At present, the child is two years old. The lesion on the right thigh has no noticeable bulge, and the movement of the right leg is unrestricted.

Case 3: The newborn was discharged after a period of observation.

A summary of the characteristics of the above three cases is shown in Table 1.

DISCUSSION

Congenital infantile fibrosarcoma (CIF) is rare, accounting for less than 1% of all childhood cancers; these tumors are mainly detected at birth (30%) or in the first year of life and are rarely detected prenatally[5]. Prenatal imaging is very useful for detecting abnormal fetal masses, but the final diagnosis relies on postpartum pathology confirmation. CIFs are usually located in subcutaneous tissues and develop in different parts of the body, most commonly in the extremities. Complete surgical resection is usually curative and sufficient in most cases[6]. Our findings were consistent with data in previous literature[7-11]: fibrosarcoma is a highly vascular tumor, which leads to difficulties in interpretation on prenatal ultrasound and can show rapid growth. We have summarized these studies (Table 2) and compared their results to the present study. Prenatal imaging features of CIF are nonspecific, and the diagnosis of CIF is challenging. On prenatal sonography, CIF appears as a large, expansive, solid soft tissue mass. It usually presents as a heterogeneous lesion with high growth potential

that tends to compress adjacent structures, and cystic echogenicity might be present within the mass. Central necrosis or hemorrhagic areas might be present depending on the rate of increase in the dimensions of the mass[12,13]. The blood supply arteries of CIFs are similar to typical tumor vessels, with irregular calibers and unorganized branching patterns[14]. The immunophenotype for CIF is nonspecific, with variable expression of desmin, SMA, CD34, and S100 protein. ETV6/NTRK3 gene fusion is vital in making the correct diagnosis. Complete surgical resection is still the primary treatment for CIF. CIF can ulcerate within the uterus and cause many complications, such as polyhydramnios, torsion, anemia, hemorrhagic shock, hemolysis, oppression of adjacent organs, and cardiac insufficiency, all of which may threaten the life of the fetus[9]. In view of the imaging features, location and rapid growth of CIFs, we believe that prenatal diagnosis of soft tissue tumors and suspected fibrosarcoma is possible. It is essential to continuously monitor fetal vital signs and tumor development by ultrasound. For children with concomitant coagulation dysfunction, surgical resection after the correction of coagulation dysfunction can achieve good results. Long-term follow-up is necessary before it can be safely assumed that the child has been cured.

Congenital hemangioma (CH) is the most common hypervascular benign tumor in fetuses. The prenatal imaging features of our patients were consistent with previous literature[3]. Ultrasonic features of CH included a high-vessel density, a high peak systolic velocity, and visible blood vessels. Brix performed Doppler ultrasound imaging of 3 fetuses with prenatally discovered CH, all of which showed abundant and rapid blood flow[15]. In Case 2 of our study, the lesion was small and did not have a rich blood supply at the time of prenatal ultrasound, but it grew rapidly during the monitoring process. Postnatal ultrasonography revealed a highly vascular soft tissue mass with smooth contours. In large lesions, complications such as thrombocytopenia, coagulation disorders (an increased prothrombin time, a decreased fibrinogen level, an increased D-dimer levels, *etc.*), high cardiac output, or bleeding are usually present[3]. In Case 2, coagulation dysfunction also occurred after birth, and good results were achieved after the correction of coagulation function and conservative medical treatment. Unlike

previous reports, Case 3 involved a twin fetus. The monitoring of the affected fetus and the protection of the healthy fetus are key issues. In Case 3, after the prenatal discovery of a hypervascular mass on the surface of one of the twins, 5 wk later, the volume of the mass had increased significantly, and intrauterine stillbirth and fetal edema occurred. We speculated that this might be due to the large size of the lesion and high flow in tumor vessels leading to heart failure, resulting in fetal death. A previous study also showed that CH is a high-flow vascular tumor that can cause high-output heart failure[16]. This might also be one of the causes of fetal death in utero. Although CHs are benign tumors, in Case 3, the CH progressed rapidly and led to an adverse outcome. Close ultrasonography follow-up was necessary, especially when the volume of the tumor increased rapidly and when CDFI demonstrated hypervascularization. In Case 3, after the affected fetus died in uterus, treatment to promote fetal lung maturation and emergency cesarean section were performed immediately, which ensured stability of the vital signs of the normal fetus and achieved good results. Based on our experience and previous studies, in the case of prenatal detection of a suspected fetal tumor, biweekly ultrasonography is recommended for the first four weeks after diagnosis to dynamically assess the likelihood of delivery and fetal cardiac status[15].

CONCLUSION

In summary, both CIF and CH may present as soft tissue masses with rich blood supplies on the fetal body surface prenatally. Once discovered, close dynamic ultrasonography is very important, especially for large lesions that grow rapidly. Prenatal ultrasound can provide accurate information such as the location, size, growth direction and blood supply of the tumor, which can provide useful information for perinatal management strategies. Although it is difficult to differentiate between CH and CIF with prenatal ultrasound, it can provide useful imaging information and is a convenient means of monitoring. Especially for lesions that occur prenatally in one twin, close ultrasonographic monitoring may allow early prediction of adverse

outcomes in the affected fetus. Once adverse outcomes occur, prompt obstetric intervention can ensure the successful delivery of the healthy fetus.

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