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CASE REPORTS

Large chorioangioma: Antenatal color-flow Doppler ultrasonic imaging and its correlation with postpartum pathology

Experience of two cases

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With the advent of ultrasonography in prenatal diagnosis, a large chorioangioma over 5 cm in diameter can be easily detected without escaping the obstetrician's notice by conventional scanning (1, 2). However, to our knowledge, there have only been few reports (3, 4) in the literature of attempts to apply Doppler velocimetry and color flow imaging in demonstrating chorioangioma's echogenecity and vascular dynamic, and to determine the clinical efficiency in diagnosis. We present here the color-flow ultrasonograms of two cases of chorioangioma identified before delivery with live births. We describe the correlation of ultrasonic imaging and pathologic confirmation, and discuss the role of color Doppler ultrasound in antenatal assessment.

Case 1

A 24-year-old woman, gravida 1 para 0, had an uneventful pregnancy until 29 weeks of gestation when a large placental mass was detected by routine ultrasonic scanning at a local obstetrician's clinic where she was then referred to our hospital. The ultrasonic assessment of fetal anatomy, amniotic volume, and growth showed normal development compatible with gestational age but breech in presentation. All gray scale and color Doppler examinations were performed with an ALOKA SSD-680 EX ultrasound machine (ALOKA Co. Ltd., Tokyo) and a 3.5 MHz curvilinear transducer on optimizing settings. The placenta was located on the fundal area and was normal in appearance. A well-defined mass mixed with internal echogenic and echolucent textures was attached to the outer third of placenta. It protruded into the amniotic cavity and measured up to 12×8.2 cm. Color flow mapping (minimum detectable flow velocity ±1.6 mm/sec, automatic change of pulse repetition frequency with a range of 0.5–25 KHz, maximum acoustic intensity I-m 110 W/cm^2) with appropriate gain setting and signal filtering low to 50 Hz showed an absence of significant vascular flow in the central part of the mass but Doppler velocimetry revealed a continuous venous flow at the periphery (Fig. 1A). The systolic/diastolic ratio of the umbilical artery was 2.3 in value. During the following period, the patient returned to the high risk unit weekly or fortnightly, and ultrasonic scanning showed that the appearance and size of the tumor, which was interpreted as chorioangioma, remained unchanged.

In her 35th week, after a mild discharge of vaginal fluid for 4 days, the patient visited the emergency delivery room with the complaint of sudden onset of water gushing from the vagina. On examination, meconium-stained amniotic fluid and a footling type of breech presentation were noted, and the external fetal monitor revealed a normal heart pattern with no decelerations. A female baby stained with thick meconium was delivered by emergency cesarean section, Apgar scores of 5 and 7 were assigned at one and five minutes and birth weight was 2300 g. The neonate's laboratory studies disclosed hematological leukocytosis (WBC 22×10^3/μL), mild anemia (Hb 12.9g/dL), thrombocytopenia (platelets 6.1×10^4/μL), and an increase in C-reactive protein (136.2 mg%). The infant was treated for congenital

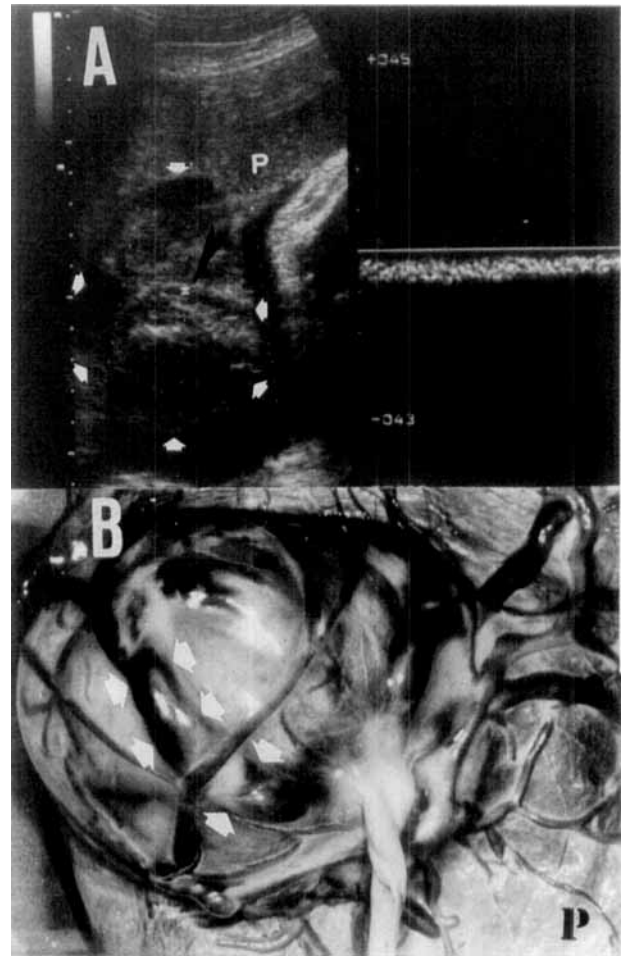

Fig. 1. A. Venous flow (arrowhead, scanning plane tangent to the mass) at the periphery of the mass (arrows), consistent with B. A chorionic vein (arrows) which transverses along the fetal surface of the chorioangioma. P, normal part of the placenta.

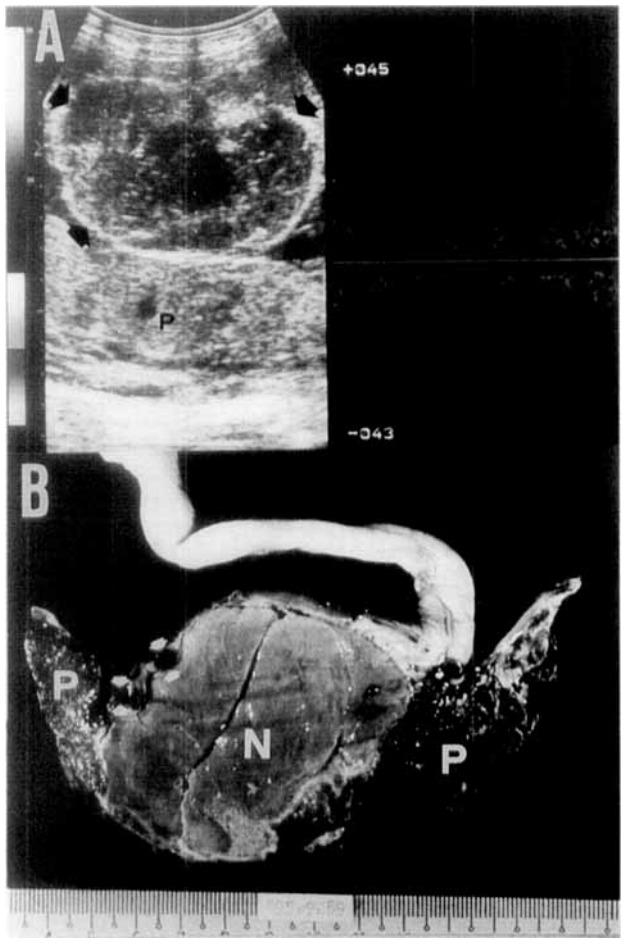

Fig. 2. A. Ultrasonic appearance of the chorioangioma (arrows), no flow demonstrated when color flow mapping superimposed. B. Section showing necrosis over a large area, no arteriovenous shunt in the mass, and chorionic vessels (arrows) that cross over the surface. P, normal part of the placenta. N, necrosis.

infection with ampicillin and gentamicin and was discharged from hospital 8 days later.

The placenta measured 22×18×10 cm and weighed 1030 g with a centric insertion of the cord. There was one round yellowish-gray well-encapsulated protruding mass measuring 15.5×9×8 cm, mediolateral to the cord insertion and embedded within the placenta. Chorionic vessels were visible beneath the amnion overlying the mass (Fig. 1B), and the arteries, which appeared smaller, crossed over and separated from the veins, which appeared larger. The cut sections showed numerous tiny anastomosing vascular channels lined by prominent endothelial cells, and necrosis and hemorrhage over a large area. The pathologic diagnosis was benign chorioangioma. There was no evidence of macro- or microscopic arteriovenous malformation, communicating artery or vein in the mass.

Case 2

A 23-year-old, gravida 2 para 1, patient visited our hospital at 35 weeks' gestation because she had had a large placental tumor for 5 weeks and was referred for survey. Ultrasonic examination confirmed that the fetal size and date were compatible, the fetus was normal looking, and the amniotic volume was adequate. However, the presentation was breech. The placenta was located anteriorly. Similar to Case 1, scanning revealed a round mass measuring 8.9×6.1 cm and protruding into the amniotic cavity (Fig. 2A). Color flow mapping and velocimetry, when superimposed, demonstrated an absence of vascularization in the central part of the tumor.

The fetus was carried to term at 38^+ weeks without complications and was delivered smoothly by cesarean section due to malpresentation. The newborn female had Apgar scores of 5 and 8 at one and five minutes, weighed 3100 g, and was moderately stained with meconium.

The placenta weighed 830 g and measured 26×13×6 cm. A well-circumscribed mass measuring 7.5×7.5×5 cm was embedded in the center and occupied the thickness of the placental plate. One prominent venous branch which was crossed over by small arterial branches of the umbilical vessels transversed along the fetal surface without entering the mass (Fig. 2B). Microarchitecture revealed a chorioangioma with diffuse, tiny anastomosing channels lined with endothelial cells in the subcapsular layer, tumor necrosis over a large area interlaced with hemorrhages and calcification, and an absence of arteriovenous connection.

Discussion

Placental chorioangioma may be associated with various fetal complications such as hydramnios, congestive heart failure, hydrops, anemia, prematurity, and growth retardation when the size exceeds 5 cm in diameter (1–5). Since the first ultrasonic diagnosis by Asokan et al. (5) in 1978, several reports have addressed its echopattern, relation with placental anatomy and association with fetal jeopardy. This has made intrauterine assessment and follow-up before delivery possible. Those reports of usual ultrasonic findings, including intraplacental subchorionic location, well-defined circumscription, complex echogenicity, single or multiple tumors, and protrusion into the amniotic cavity, are easily interpreted but there is still a lack of image specificity for certain diagnosis (2, 4). To advance ultrasonic technology's ability to assess chorioangioma, the Doppler velocimetry and color flow mapping were therefore introduced in an effort to demonstrate the hemodynamic status of the lesion, which is speculated to have internal vascular flows. The possibility of this technique being helpful needs to be proven.

There are two studies in which Doppler velocimetry and/or color flow mapping were used to check the chorioangioma (3, 4). In 1986, Grundy et al. (3) presented a woman who had severe hydramnios and a large chorioangioma with vascular channels in the tumor confirmed by Doppler studies at 32 weeks' gestation. There was an identifiable heart rate in the vascular spaces of the tumor, the same as the pulse rate in the umbilical cord. This is the first study to show hemodynamic flow in the chorioangioma. The infant was prematurely terminated at 33^+ weeks of gestation due to poor variability of fetal heart rate monitoring. The authors hypothesized that the hydramnios with a large chorioangioma resulted in excessive transfer of fluid into the amniotic cavity because of functional intraplacental insufficiency and fetal cardiac failure. In 1993, Hirata et al. (4) reported a woman who had a large complex chorioangioma and suffered from premature labor associated with impending fetal hydrops at 28 weeks' gestation. Doppler studies and color flow mapping, when superimposed, revealed pulsatile blood flow through the mass. The fetus was delivered because of tocolysis failure 20 days after initial diagnosis. The authors speculated that the nonimmune hydrops was secondary to either shunting through the large arteriovenous malformation or microangiopathic hemolytic anemia resulting in fetal congestive heart failure. Both reports showed evidence of significant blood flow detected by Doppler velocimetry and/or color flow mapping in the tumor and postulated that an arteriovenous shunt existed and resulted in maternal and fetal

complications. But, in their postpartum pathology, they did not disclose clearly whether the placental chorioangioma had either communicating vessels or arteriovenous malformation or not. Santolaya-Forgas in the text once showed an impressive figure of the chorioangioma, in the early stage of gestation, with internal hypervascularization recognized when color imaging was superimposed (6). Nevertheless, the author did not have the detailed information to know the following changes of the mass during pregnancy and the fetomaternal outcome.

In our study, two cases with large chorioangioma that were found at 30 and 35 weeks' gestation, respectively, did well during the antenatal follow-up period and both had good neonatal outcome. We used Doppler velocimetry and color flow mapping to evaluate the internal texture and flow dynamics of the tumor. However, an absence of flow was demonstrated. These findings were also compatible with pathologic examination, which disclosed numerous microscopic vascular endothelial linings with large areas of necrosis and calcification and no evidence of macro or microscopic shunting. The venous flow antenatally detected by ultrasound at the periphery of the mass corresponded to the tracing of the chorionic vein without direct connection with the artery overlying the chorioangioma. Based on the above observations, we speculate that the following possible pathophysiologic mechanisms may occur. First, spontaneous thrombosis and infarction of the feeding vessels of the tumor may occur in any stage of gestation and can lead to necrosis, hemorrhage and subsequent calcification, which block the preexisting arteriovenous shunt and then abolish the potential risk of fetal complications. In both of our study cases, visits to our clinic occurred too late to show the shunting. Second, a chorioangioma *per se* has neither clinically significant blood flow nor an identifiable anatomical arteriovenous shunt but is mainly composed of solid vascular endothelial neoplasm and may act as a unit of functional, not morphologic, shunt. Tumor necrosis associated with hemorrhage unavoidably occurs when it grows large. Actually, there have been few reports (7, 8) in which the authors could demonstrate any anatomical arteriovenous shunts on pathology.

Most placental chorioangioma have no clinical significance (1) and even large ones (>5 cm) may be associated with an uncomplicated pregnancy and a good neonatal outcome (2). The use of color-flow Doppler ultrasound in antenatal assessment of large chorioangioma may be helpful if the placental tumor is found early before thrombosis and infarction occur or anatomical arteriovenous shunts exist, but its use is of little help if the tumor is thought to be a functional shunt. As for those with fetal and/or maternal complications, according to the Grundy's and Hirata's reports, it may play a role in demonstrating the blood flow inside (3, 4). As for those without complications with where little blood flow can be found, as evidenced by our examination, it is of limited use. The efficiency of color-flow Doppler examination in diagnosis and evaluation of the chorioangioma remains equivocal, it may vary with the tumor size and age, and it still needs considerable study for further clarification.

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Primary mixed trabecular and insular carcinoid tumor of the ovary: a case report

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Primary carcinoid tumors of the ovary generally are divided into four categories: insular or islet carcinoid (midgut type) (1); trabecular carcinoid (foregut or hindgut type) (2); strumal carcinoid (struma ovarii and carcinoid or carcinoid tumor combined with thyroid tissue) (3); and mucinous (goblet cell) carcinoid (4).

Although many cases of primary ovarian carcinoid have been reported, only few cases of primary ovarian carcinoid composed of both trabecular and insular patterns have been described (1, 2, 5, 6, 7, 8).

The clinicopathologic features, treatment and follow-up

Abbreviations: 5-HIAA: 5-hydroxyindole acetic acid; NSE: neuron-specific enolase; hCG: human chorionic gonadotropin.