CASE REPORTS

Foetal vascular lesion–Case report

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Received: April 16, 2018 **Accepted:** May 21, 2018 **Online Published:** May 28, 2018
DOI: 10.5430/ijdi.v5n2p1 **URL:** https://doi.org/10.5430/ijdi.v5n2p1

ABSTRACT

Frequency and preciseness of prenatal detection of foetal tumours increases due to improvement of sophisticated imaging methods. As correct diagnosis impacts the course of care in utero, it is essentially to improve diagnostic workout in a case of detected foetal anomalies. Here we report the case of partly involuting congenital haemangioma of foetus, which antenataly caused foetal secondary cardiomegaly. Pregnant woman was referred to Riga Maternity Hospital with unexplained tumour on the surface of foetal head at 24+2 weeks of gestation. Ultrasound exam revealed tubular structure without solid components between calvarium and skin under the left ear with very rich vascularization. Magnetic Resonance Image demonstrated enhancing multi-cystic lobulated mass. Hypertrophic secondary cardiomegaly was present without any additional structural abnormality. The foetus remained stable until 36+4 weeks of gestation, when the size of tumour succeeded 85 mm $\times$ 46 mm. Haemangioma was confirmed after delivery as round raised and infiltrating vascular lesion. After birth MRI demonstrated its connection with a. carotis externa. Propranolol was recommended with continuing follow-up. At 2 years and 3 months of age the lesion decreased by size noticeably, but still persists. Accurate diagnosis lets obstetricians to optimize antenatal care by providing an opportunity for planning deliveries, preparing family and medical staff for appropriate postpartum therapy and management.

Key Words: Foetal vascular tumours, Haemangioma, Prenatal diagnosis

1. INTRODUCTION

The incidence of all congenital tumors was reported as the range of 1.7-13.5 per 100,000 live births, but data may be underestimated, as many of these cases may not be reported due to stillbirth or miscarriages.[1] Frequency and preciseness of prenatal detection of foetal tumours increases due to improvement of sophisticated imaging methods.[2] Ultrasound examination may even reveal their histological features. Foetal MRI can be used to examine lesions and differentiate abnormality. The differential diagnosis of vascular lesions, according to histology and history, usually includes tumours and vascular arteriovenous malformation.[3,4] The International Society for the Study of Vascular Anomalies (ISSVA) updated tumours classification and divided congenital haemangiomas into three main types: rapidly involuting congenital haemangioma (RICH), noninvoluting congenital haemangioma (NICH) and partly involuting CH PICH.[3,4] Vascular malformations that grouped into 4 types enlarge gradually after birth and it growth influence such factors as pregnancy, trauma, surgery and puberty.

As correct diagnosis impacts the course of care in utero, as well as during labour and postnatally, it is essentially to improve diagnostic workout in a case of detected foetal anomaly.

The objective of this article is to demonstrate the case of partly involuting congenital haemangioma of foetus, which antenataly caused the foetal secondary cardiomegaly.

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2. Case report

Pregnant 25 years old woman (G1 P0) was referred to Riga Maternity Hospital for the expert opinion with unexplained tumour on the head surface of the foetus, which was detected on ultrasound examination at 24+2 weeks of gestation. The vascular malformation has been growing fast, as it was not present on anomaly scan at 20+1 weeks of gestation. The first trimester genetic screening was negative.

Ultrasound exam revealed tubular structure (see Figure 1) without solid components between scull and skin under the left ear (31 mm × 29 mm) with very rich vascularization. Spectral and Colour Doppler revealed high blood flow velocities and low pulsatility within the lesion (PSV of 210 cm/s and pulsatility index [PI] of 0.50) suggesting the presence of arteriovenous shunts. The sick blood vessels were coming from brachial vessels (see Figure 2). Hypertrophic secondary cardiomegaly was present without any additional structural abnormality. Otherwise, foetus was compensated, no signs of hydrops were observed.

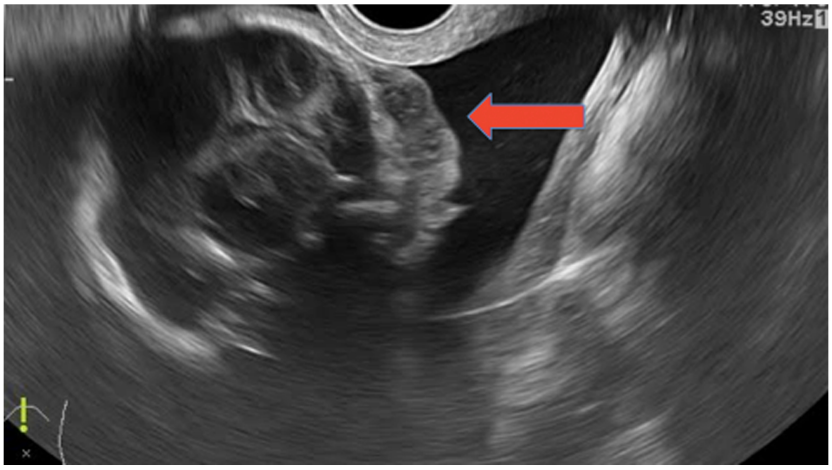

Figure 1. Gray scale ultrasound image at 24+2 weeks of gestation demonstrates tubular structure without solid components between scull and skin under the left ear (arrow)

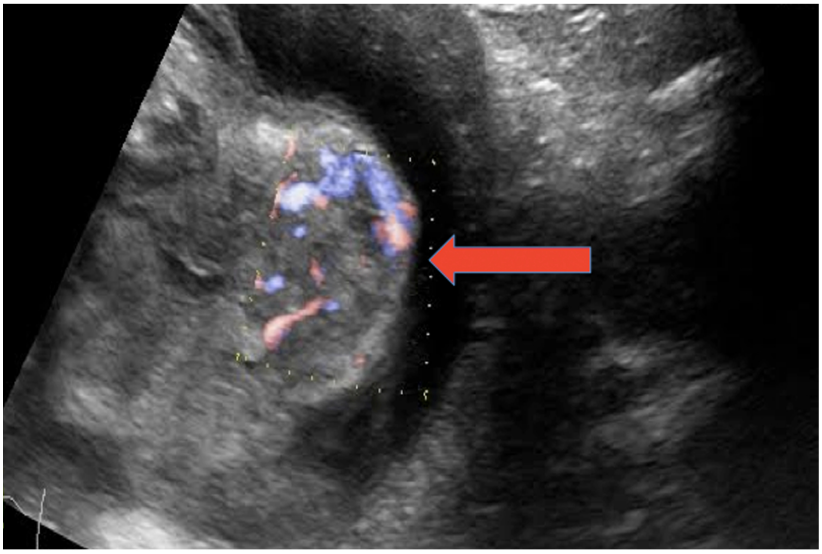

Figure 2. Colour Doppler image demonstrates typical high vascularity within the lesion (arrow)

Magnetic Resonance Image (MRI) demonstrated enhancing multi-cystic lobulated mass (see Figure 3) and allowed evaluate anatomically it effect on surrounding structures.

The family was counselled about prognoses of probably arteriovenous malformation with it possible fast enlargement and following cardiac impairment, when the foetus may die in utero. The different therapeutic intrauterine options such as fulguration of the vessels with Radiofrequency Ablation

ISSN 2331-5857 E-ISSN 2331-5865

(RFA), interstitial laser or alcohol were offered.[5,6] The family opted conservative management.

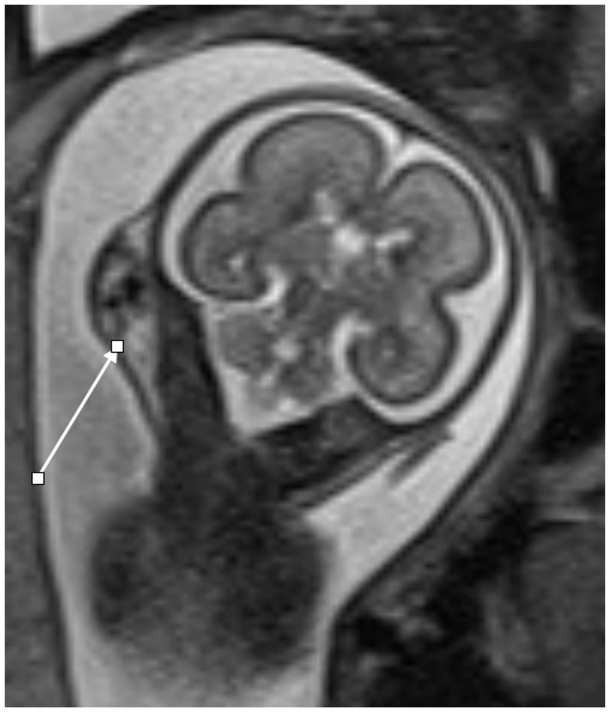

Figure 3. MRI demonstrating enhansing lobulated multicystic lesion with (T2 signal) hypodence areas (arrow)

The foetus remained stable until 36+4 weeks of gestation, when the size of tumour succeeded 85 mm × 46 mm. Contractions started shorty after (37+0 weeks) and emergent caesarean section was performed. The birth weight of girl was 2,860 grams and Apgar scores were 7/9 on the 1st and 5th minutes. Haemangioma was confirmed after delivery (see Figure 4) as round raised and infiltrating vascular lesion.

After birth MRI demonstrated its connection with a. carotis externa and not related to the other tissues and structures. Propranolol was recommended (2 mg/kg/per day) with continuing follow-up.

At 3 months of age the tumour became smaller in size (65 mm × 30 mm) and brighter (see Figure 5), responding to the therapy. That confirms the diagnosis of rapidly involuting congenital haemangioma. At 2 years and 3 months of age the lesion decreased by size noticeably, but still persists (see Figure 6), that turned the previous diagnosis into PICH. At the moment the family is satisfied with cosmetic effect of conservative treatment and do not consider surgical intervention.

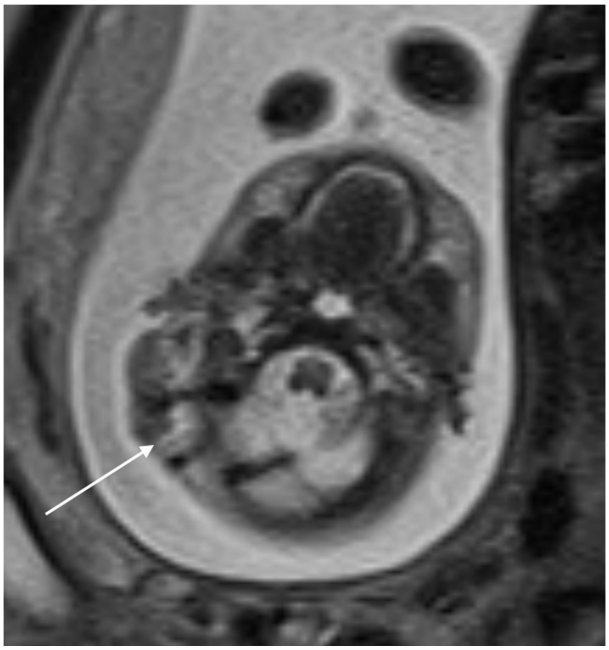

Figure 4. MRI demonstrating enhansing lobulated multicystic lesion with (T2 signal) hypodence areas (arrow)

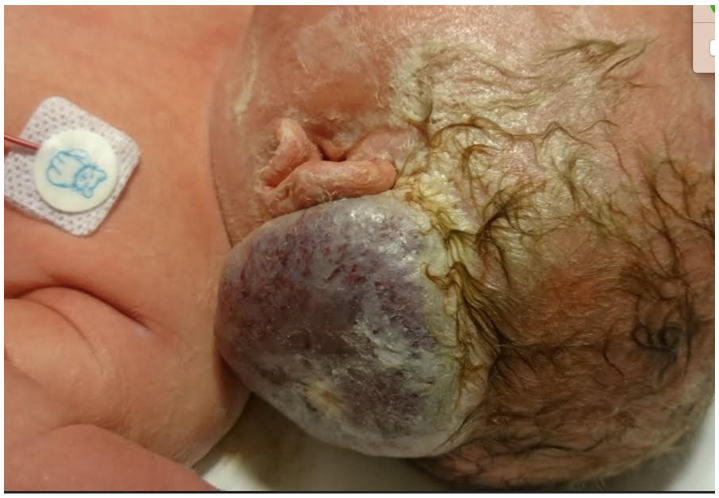

Figure 5. Image of the heamangioma after birth

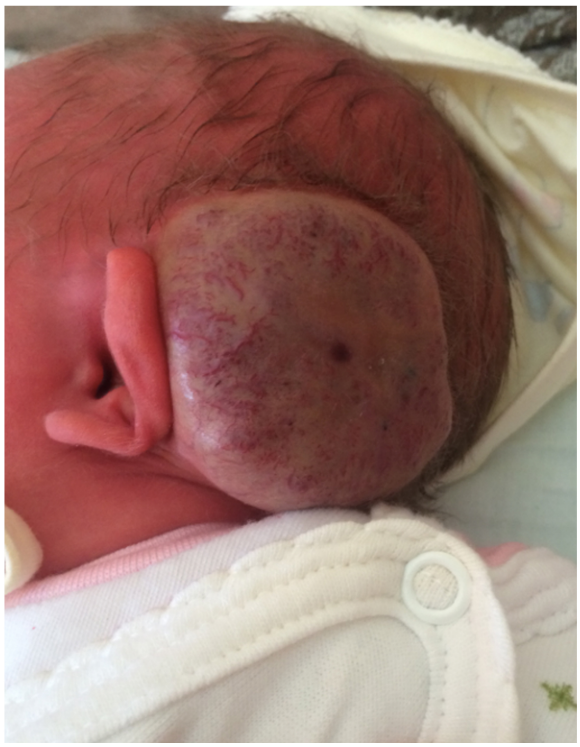

Figure 6. Image of the heamangioma at 3 months age

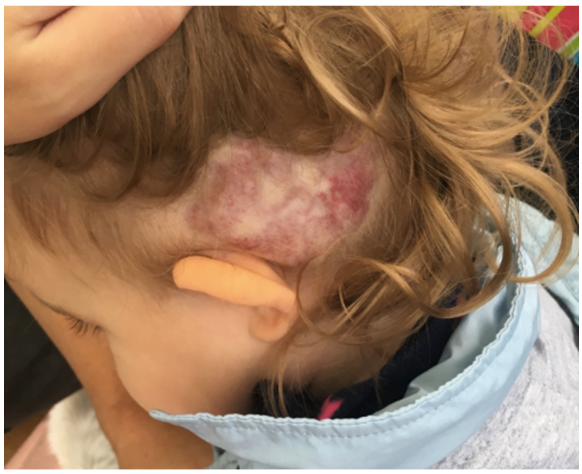

Figure 7. Image of the heamangioma at 2 years and 3 months age

3. Discussions

The clinical presentation of vascular lesions of the head and neck can range from a birthmark to severe disfigurement, functional impairment or relevant hemorrhage pre- or intranataly.

Mulliken and Glowacki in 1982 the first time published classification of congenital vascular tumours and malformations, but still interchangeable use of these terms puzzles obstetricians and pediatricians.[7]

Haemangioma is a benign tumour of endothelial cells with an incidence rate up to 12% among newborns. Sometimes CT or MRI or even invasive diagnostic procedures are necessary to confirm the diagnosis. Kasabach-Merritt phenomenon, characterized by serious bleeding due to severe consumption coagulopathy, should be kept in mind. In our case this abnormality was excluded as thrombocytopenia have not been confirmed.

Different therapeutic methods were historically used, including topical and systemic steroids as well other cytotoxic drugs with known side effects and quite low efficacy. The effectiveness of the beta-blocker propranolol for IH and CH was discovered in 2008.[8,9] Some cases, particularly on the face, require plastic surgery including laser treatment or resection, to achieve cosmetic appearance.[8,9]

In our case MRI was used for differential diagnosis of vascular lesion, respecting ALARA (as low as reasonably achievable) concept (T2 signal with 1.5 T). MRI is considered safe during pregnancy. According to the Food and Drug Administration the limits for radiofrequency (RF) exposure is 4 Wkg_1 for maternal whole-body exposure, independent of the applied magnetic field strength.[10] At the same time the Canadian Task Force on Preventive Health Care confirms safe use of MRI at less than 3 T (Tesla field strength) at the second half of pregnancy.[11] Even use of gadolinium-based contrasts should not be limited in diagnosing of serious maternal and fetal disease.[12] The last was not applied during the examination of described case.

4. Conclusion

Our case demonstrates how congenital haemangioma could influence and complicate prenatal period and even may cost someone life. Accurate diagnosis lets obstetricians to optimize antenatal care by providing an opportunity for planning deliveries, preparing family and medical staff for appropriate postpartum therapy and management.

Conflicts of Interest Disclosure

The authors have no conflicts of interest related to this publication.

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