CASE REPORT

Left Lower Limb Arteriovenous Malformation at Term Gestation: A Case Report

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Abstract

Background: Vascular malformations are relatively rare disorders of the vascular system and its maldevelopment can present at birth.

Case description: A 21-year-old primigravida presented at 38^{+4} weeks of gestation in the latent phase of labor. The patient was a known case of lower limb congenital hemangioma/arteriovenous malformation. The patient claimed to having been born with the deformity at birth, which presented as a small swelling over the lateral aspect of her knee. The patient had procured a single magnetic resonance imaging report that she had undergone 7 years back, which stated diffuse subcutaneous or intramuscular short T1 inversion recovery (STIR) hyperintense lesions in the left thigh and leg as described with phleboliths in the lower third of the thigh and to include the possibilities of venous malformations and venolymphatic malformations. There was anticipated difficulty in delivery owing to poor limb development, limb deformity, and inability to flex the hip and knee, and probable cephalopelvic disproportion was present. Patient spontaneously progressed and underwent full-term vaginal delivery and right mediolateral episiotomy to deliver a live baby girl of birth weight 2.443 kg and APGAR score of 8/10 and 9/10 at 1 minute and 5 minutes, respectively.

Conclusion: The disorders of the vascular system especially of the lower limb and pelvic girdle cannot be ignored as it can pose many complications during delivery.

Keywords: Arteriovenous malformation, Crepe bandage, Delivery, Neonate, Rare anomalies, Sclerotherapy, Treatment, Vaginal birth.

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Background

Vascular malformations are relatively rare disorders of the vascular system and its maldevelopment can present at birth.[1] This occurs in approximately 0.3% to 0.5% of the total population.[1] Pregnancy has been considered a risk factor in the development of arteriovenous (AV) malformation hemorrhage, especially in conditions with hemangioma or AV malformations.[2]

Children with AV malformations can experience an impairment in the overall bone growth, and this occurs with the coexistence of other vascular malformations.[3] Hypotrophy is the common manifestation in vascular bone syndromes that are usually complex and progressive over time.[3] The usual mode of therapy is conservative management.[3] The complications to be anticipated in an affected lower limb are pelvic tilting and scoliosis, as literature review mentions a discrepancy of 10 cm between the limbs and orthopedic interventions may worsen the existing condition.[3]

Case Description

A 21-year-old primigravida presented at 38^{+4} weeks of gestation in the latent phase of labor. The patient was a known case of lower limb congenital hemangioma/AV malformation. The patient claimed to having been born with the deformity at birth, which presented as a small swelling over the lateral aspect of her knee. The patient was able to carry out daily household activities and was able to participate in school activities, but did so with slight tilt compensation and shifting her weight predominantly to her apparently normal right leg. Over time, the patient and her parents noticed lesions that formed subcutaneous swellings and projections that deformed the overall contour of her leg. She was not evaluated for the same. Regular antenatal visits were conducted at the patient's hometown, and she presented to our center at 36 weeks of pregnancy (Fig. 1). During 17^{+5} weeks of pregnancy, her hemoglobin was 9.8 g% and she was treated with oral hematinics. Her anemic status improved thereafter (repeat Hb, 10.4). At 35^{+5} weeks of gestation, her routine scan showed features of fetal growth restriction, corresponding to 33 weeks with normal Doppler study. Incidentally her platelet count was noticed to be 1.2 lakhs/mm^3. In view of left lower limb hemangioma/AV malformation, a surgery opinion was taken. Crepe bandage application and conservative modalities were advised. A plan for sclerotherapy after delivery was proposed. On preparation for her safe confinement at term, routine investigations were done. Left lower limb venous Doppler showed no evidence of deep vein thrombosis. A soft tissue scan showed features of infiltrating subcutaneous and intramuscular hemangioma and

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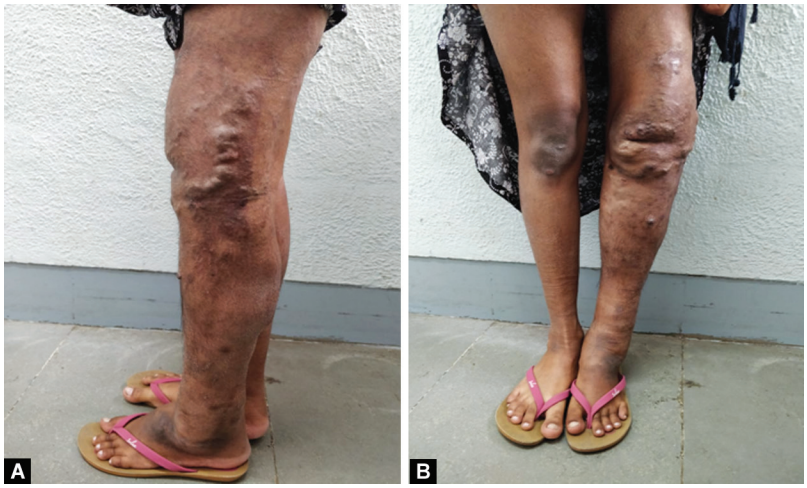

Figs 1A and B: Clinical presentation at term

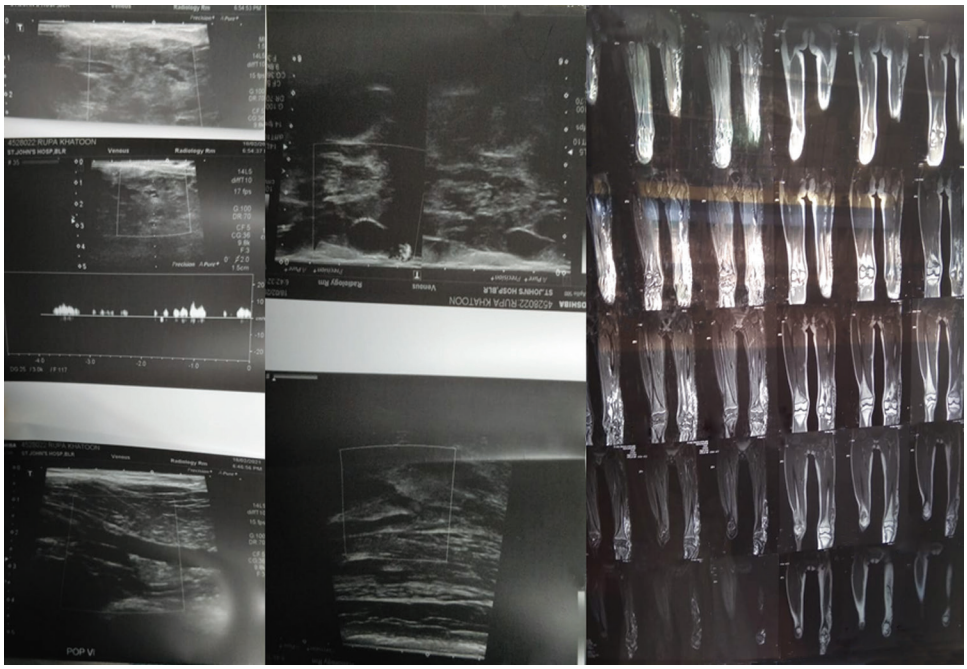

Fig. 2: Scan and MRI of the patient

AV malformation, and a magnetic resonance imaging (MRI) was performed for correlation. The patient had procured a single MRI report that she had undergone 7 years back, which stated diffuse subcutaneous or intramuscular STIR hyperintense lesions in the left thigh and leg as described with phleboliths in the lower third of the thigh and to include the possibilities of venous malformations and venolymphatic malformations (Fig. 2). On admission, the patient was advised to maintain daily fetal movement count, and nonstress test was found to be reactive. There was anticipated difficulty in delivery owing to poor limb development, limb deformity, inability to flex the hip and knee, and cephalopelvic disproportion. The patient spontaneously progressed and underwent full-term vaginal delivery and right mediolateral episiotomy to deliver a live girl baby of birth weight 2.443 kg and APGAR score of 8/10 and 9/10 at 1 minute and 5 minutes, respectively. Post delivery the uterus was intermittently relaxing, and the patient was managed with one dose of injection carboprost 250 mcg I.M., 600 mcg per rectal and one dose of injection methyl ergometrine I.M. A loop of cord was present around the neck, which was relieved after delivery of the head, and the baby cried after stimulation. The baby had audible grunting with a respiratory rate of 62 cycles/minute and was shifted to the neonatal intensive care unit for monitoring. The baby was found to have symmetrical Fetal growth restriction (IUGR) but had passed the congenital congestive heart disease screening test. The baby received an intermittent single light phototherapy due to a total bilirubin of 14.32. Injection enoxaparin 0.4 mL subcutaneous dose along with bilateral crepe bandage was opined by vascular surgery that was taken as a cross-reference. Postnatal period was uneventful. Crepe bandage was applied on both lower limbs, and

the patient received injection Clexane, ecosprin 75 mg tablets daily once, and antibiotics for 5 days. The patient was stable and discharged with an intended plan of performing an MRI of the left lower limb, followed by sclerotherapy on a later date.

Discussion

Vascular malformations are quiet discrete topics that pose lacunae and join the vast groups of poorly understood set of health conditions that cause many medical dilemma, both diagnostically and therapeutically.[4] Recent stand on treatment plans suggests endovascular therapy such as embolization and sclerotherapy, which are becoming more popular in recent times.[4] Our subject, however, preferred to be managed conservatively during pregnancy as there are no set guidelines on the effects of scleroscant on fetus and the changes it can impart to the ever-so-changing maternal hemodynamic state. As literature now considers these modalities as the first-line treatment, we have considered sclerotherapy for our patient after the puerperium of 6 weeks.[4–6]

Conclusion

The disorders of the vascular system especially of the lower limb and pelvic girdle cannot be ignored as it can pose many complications during delivery. Prompt and vigilant antenatal monitoring and antepartum fetal surveillance are a necessity in the management of such rare disorders in pregnancy. A multidisciplinary approach, watchful anticipation of complications (deep vein thrombosis) and prompt anticoagulant therapy can prolong the surgical intervention in late pregnancy. This can buy time to accustom the mother and neonate in the postnatal period thereby decreasing the added stress and complication of surgical interventions.

Clinical Significance

This report is one of the rare presentations of AV malformations in pregnancy which has been described in the past 5 years. The management of the patient with simple yet effective conservative modalities has resulted in an uneventful pregnancy, which can be considered for future treatment protocols.

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