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Letters to the Editor – Brief Communications

Abducens nerve palsy as a sign of pre-eclampsia with severe features

Dear Editor, we present a unique case of severe pre-eclampsia.

A 36-year old primigravid woman presented at a gestational age of 33 weeks and 2 days with abrupt reduced vision and headache. Past medical history was negative, except for migraine. At admission, blood pressure readings reached 169/100 mmHg. A urine dipstick indicated significant proteinuria, confirmed with a 24 h urine collection revealing 971 mg of protein. On clinical examination, hyperreflexia and unilateral abducens nerve palsy was apparent (Fig. 1).

The patient was admitted for fetal lung maturation and observation. Labetalol 600 mg daily and magnesium sulfate intravenously were administered, however diastolic hypertension persisted.

Due to the persisted severe hypertension, a caesarean section was performed 48 h after admission and with completion of the fetal lung maturation. The neonate was admitted to the neonatal unit since the birth weight was only 1470 g. Apgar of the neonate were 7/5/10, after 1, 5 and 10 min(s) respectively. No neonatal problems were observed during the 1-month hospitalization.

Magnesium sulfate was continued for 48 h after delivery. A cerebral MRI was performed to rule out ischemia or hemorrhage, no abnormalities were observed. Within 3 days after delivery, a reduction of the palsy and normalization blood pressure was detected. Full recovery of the palsy was observed within 5 months.

Only about nine cases have been published, displaying the rarity of this clinical feature [1–3]. Although this feature is uncommon, the obstetrician should be aware of the clinical significance. The true etiology of this sign is yet to be determined, but nerve compression due to intracranial hypertension and vasospasm of the nerve vessels due to hypertension, have been hypothesized [1]. Regardless, all neurologic symptoms, including palsy, should be regarded as a sign of severe pre-eclampsia [4]. Before 34 weeks of gestational age, British and American guidelines propose to first administer corticosteroids for fetal lung maturation for 48 h and then initiate delivery if severe hypertension remains or the clinical condition of the patient or fetus deteriorates [4,5]. In this case, a caesarean section was selected as the route of delivery, mainly due to the low estimated fetal weight. According to the guidelines, caesarean section should not be the *de facto* route of delivery and should be determined by other obstetrical, fetal or maternal factors [4,5].

Fig. 1. Unilateral abducens nerve palsy of the left eye when asked to look to the left direction.

Abducens nerve palsy should be regarded as a sign of pre-eclampsia with severe features and should lead to swift action. Although such a case is very rare, any obstetrician should recognize its importance.

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Twin pregnancy complicated by disseminated intravascular coagulation following single fetal demise

Dear Editor,

Intra uterine fetal demise (IUFD) complicated by DIC is a rare but known complication of singleton pregnancies [1]. Coagulation abnormalities are due to the gradual release of tissue factor from the placenta into the maternal circulation [1]. Unlike singleton pregnancies, twin pregnancies with single IUFD are rarely complicated by DIC [2,3].

A 26 year old woman was referred to our center at 21 weeks of her Dichorionic Diamniotic twin pregnancy due to hydrops fetalis of both twins with MCA PSV flow correlating with severe fetal

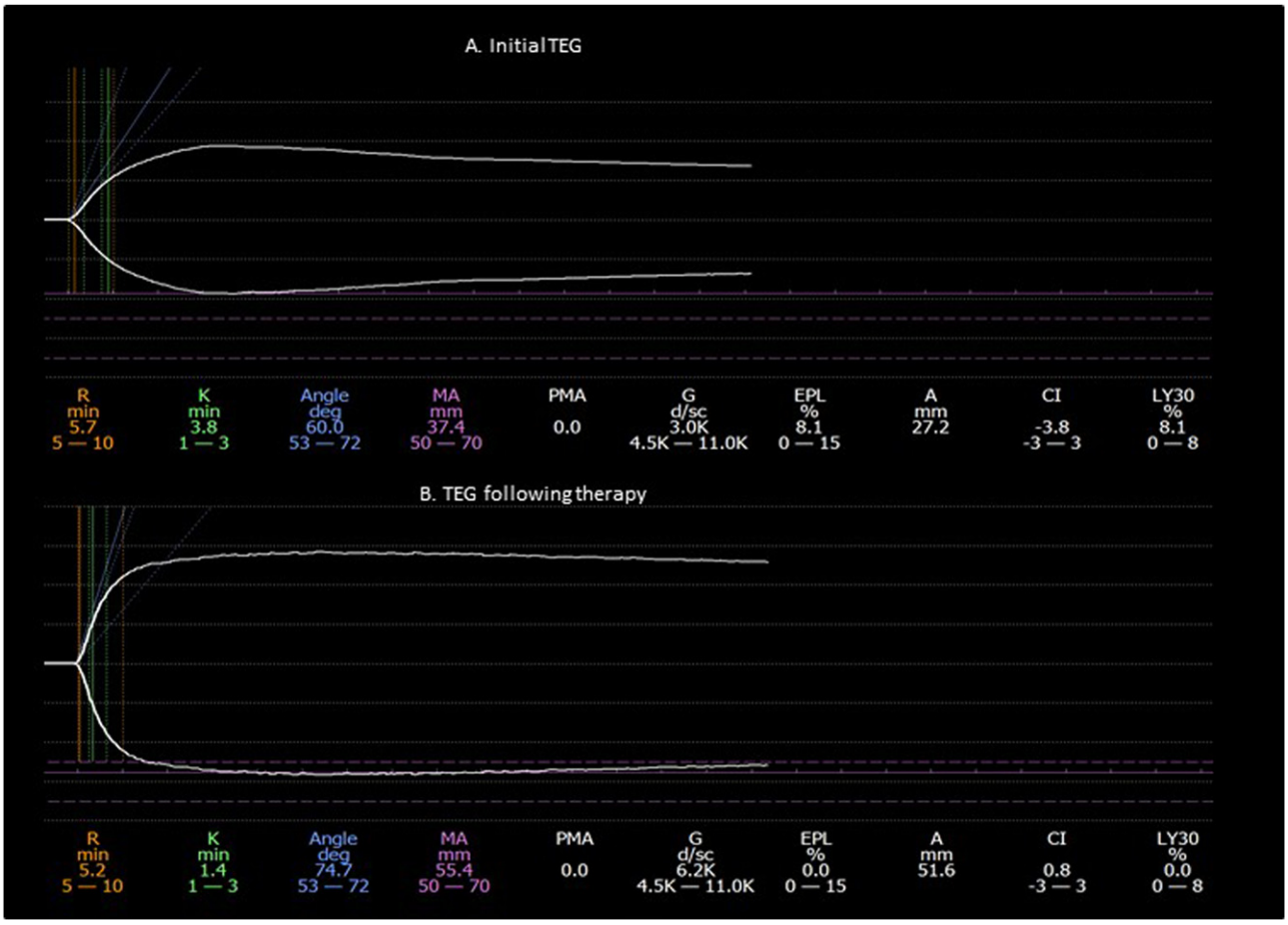

Fig. 1. A. Patient's initial Thromboelastogram, indiucating DIC, B. Patient's Thromboelastogram following therapy, represnting normal coagulation.

anemia was diagnosed. The pregnancy was treated at a fetal therapy center with intra uterine blood transfusion. This resulted in IUFD of twin A, twin B transfusion was successful and quick resolution of the hydrops and anemia was noted within weeks. The woman who suffered from mirror syndrome during fetal hydrops also quickly improved. Parvovirus infection was confirmed via maternal serology and PCR from amniocentesis of both fetuses.

She represented at 24 weeks with preterm premature rupture of the membranes (PPROM) of the demised twin A. She was treated with a 24 h course of betamethasone and a one week course of ampicillin and roxithromycin and the fetal routine sonographic biophysical profile and non-stress test was unremarkable with no signs of chorioamnionitis or placental abruption.

At 30 week and 5 day gestation routine blood tests showed dysfibrinogenemia of 105 mg/dL (normal levels in the third trimester of pregnancy are 373–619). Other coagulation functions (PT, PTT, INR) were normal. The patient was asymptomatic with normal vital signs and no vaginal or other bleeding. Further blood tests were indicative for disseminated intravascular coagulation (DIC) including D-Dimer levels (35,088 ng/mL, normal levels 130–1700) and Thromboelastogram (TEG) (Fig. 1).

Considering all the above findings and even though she was asymptomatic we decided to deliver the fetus. An emergency cesarean under general anesthesia performed due to breech presentation with the intention of minimalize the potential bleeding of regional anesthesia. The woman was treated with 10 units cryoprecipitate and the course of the surgery was normal with no excessive bleeding. Shortly (about 15 min.) after surgery repeat laboratory tests worsened with lower fibrinogen levels (114 mg/dL) and TEG. The patient developed early post-partum hemorrhage with vaginal bleeding estimated at 1000 ml. She was treated with fresh frozen plasma and packed cells for her coagulation disorder together with uterotonics. The following laboratory tests and post-operative course were normal. The baby was released from the Neonatal intensive care unit at age 2.5 months with no prematurity complications.

DIC is a frequent cause of maternal morbidity and mortality and is associated with up to 25% of maternal deaths [4]. Proper treatment of this hazardous complication relays on early recognition diagnosis. Yet, frequently the diagnosis of DIC is challenging and the laboratory abnormalities are precursory of the clinical symptoms. The rare case presented above demonstrates that DIC following single fetal demise do occur, and may be associated with bleeding tendency. Awareness to this association should lead to prompt respiratory and hemodynamic support along with administration of coagulation resuscitation products (FFP/cryoprecipitate).

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Patient consent form has been completed and signed by the patient.

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Uterus-like mass: A case report

Dear Editors,

We report a case of a uterus-like mass. A 28-year-old nulliparous patient presented with increasing dysmenorrhea after discontinuing use of her contraceptive pill. Clinical examination revealed pelvic pain mainly located in the right iliac fossa. A pelvic ultrasound displayed a suspicious uterine mass initially described as a pseudo-unicorn uterus with a rudimentary horn. Magnetic resonance imaging (MRI) revealed a roundel image within the right myometrium appearing to have a cleavage plane with the myometrium. This suggested a uterus-like mass, showing hypersignal T1 and hyposignal T2 with discreet hypersignal in the center linking to a hemorrhagic content without communication to the uterine cavity (Fig. 1).

Abdominal examination through a coelioscopy showed no abnormalities. Uterus size and aspect were normal aside from an arch on the right side under the broad ligament. Incision was performed and no area of a cleavage plane was identified. Brown liquid evacuated from the mass alluding to a hematoma. The mass was removed and sent for histological examination. Patient showed no recurrence of pelvic pain or dysmenorrhea during follow-up. Histopathological examination described an endometrium with rich cellular cytogenetic chorion and oval glands lined with a cylindrical coating associated with smooth muscle cells, confirming diagnosis of a uterus-like mass.

A uterus-like mass is a rare, benign tumoral pathology first described in 1981 by Cozzuto [1]. It is defined by an ovoid structure with a central cavity bordered by endometrium and surrounded by a thick wall of smooth cells imitating normal myometrium. Literature is scarce and masses are mainly found within the pelvic cavity, affecting many generations of women, such as young women with early onset puberty or menopausal women. A uterus-like mass can also be located outside the pelvic cavity in other organs (abdominal wall, small and large bowel, mesentery, pancreas, liver, appendix, spinal cord) [2,3].

Preoperative diagnosis is difficult due to lack of specificity of both clinical and paraclinical investigations and is made through observation of pelvic pain and dysmenorrhea. Pelvic imaging typically describes an ovoid lesion, well limited, and isointense compared to endometrium with a hypointense junction without communication to the uterine cavity [4]. The central cavity shows hypersignal T2. Only histopathological examination can confirm diagnosis. Uterine malformation is the principal differential diagnosis (pseudo-unicorn uterus, unicervical unicorn with rudimentary contralateral and blind horn) but also defines other benign or malignant tumors, such as a hamartoma, teratoma, fibroma, sarcoma, and endometrioid carcinoma. It is particularly difficult to differentiate cystic adenomyoma from a uterus-like mass. It is organized around a cystic hemorrhagic cavity but without the organoid organized structure specific to a uterus-like mass.

Surgical treatment is key for removal and diagnosis of the mass. Resection should be complete to prevent recurrence, and minimally invasive. If there is no cleavage plane, dissection can be difficult.

Histopathogenesis is poorly understood and no theory explains all diseases. The theory of congenital malformation has been described due to concomitant malformations of the genitourinary tract and would imply a fusion defect or a duplication of Müller's canals. The theory of metaplasia, which Cozzuto postulated, explains that secondary coelomic pluripotent mesenchymal cells, belonging to the secondary Müllerian system, differentiate into endometrial

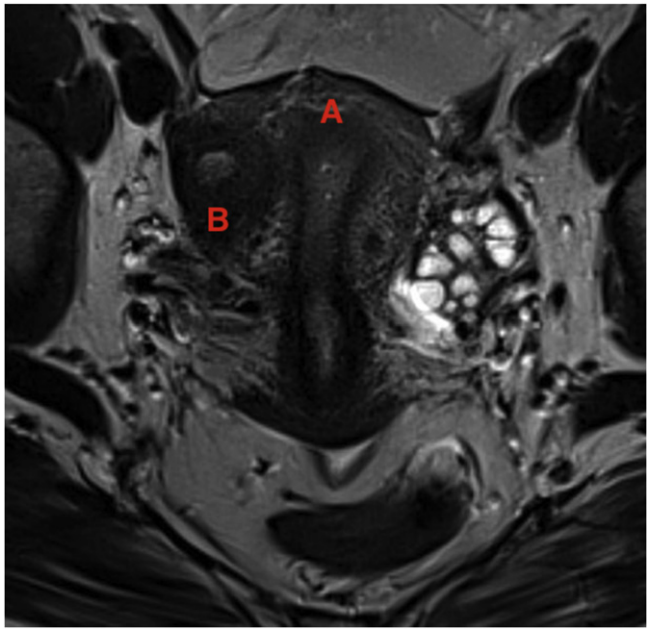

Fig. 1. MRI in frontal section in T2 sequence: A: uterus with hypersignal T2; B: right lateral-uterine mass not communicating with the endometrial cavity.