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Cardiac Arrest during Delivery Revealing a Pheochromocytoma: A Case Report

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Abstract **Case Report**

Pheochromocytoma, a rare catecholamine-secreting tumor during pregnancy, poses significant risks but is curable with timely identification and management. We present a case of a 43 year old nulliparous woman who experienced respiratory distress and cardiorespiratory arrest after delivering a stillborn at 38 weeks gestation. Initially diagnosed with amniotic embolism, her condition worsened, revealing hypertensive crises, hypoxia, and alveolar hemorrhage. Suspicion of pheochromocytoma arose, confirmed by positive urinary catecholamines and imaging. The patient underwent left adrenalectomy, and pathology confirmed a benign pheochromocytoma. The complex diagnostic and therapeutic challenges of managing pheochromocytoma during pregnancy are discussed, emphasizing the need for multidisciplinary care and tailored interventions for optimal maternal and fetal outcomes.

Keywords: Pheochromocytoma, pregnancy, hypoxia, diagnostic.

INTRODUCTION

Pheochromocytoma is a catecholamine-secreting tumour that is rare during pregnancy, with the prevalence of 1 in 54 000 pregnancies [1]. A pheochromocytoma diagnosed in pregnancy is a rare condition [2]. Published case reports describe pheochromocytomas mimicking severe pre eclampsia or causing hypertensive emergencies during delivery [2] However, pheochromocytoma is curable by removing the tumour. The ability to accurately identify pheochromocytoma during the antenatal period with timely and appropriate management reduces maternal mortality and fetal loss [1, 3, 4]. In this report, we present a case of pheochromocytoma diagnosed incidentally after respiratory distress that was complicated by cardiorespiratory arrest after vaginal delivery of a full term pregnancy.

OBSERVATION

43 year old patient, nulliparous, with no particular pathological history, her pregnancy was carried to 38 weeks amenorrhoea and she was admitted to the maternity hospital for the onset of obstetrical labour with the notion of a decrease in active foetal movements. Her obstetrical history included: the firs pregnancy one year ago complicated by gestational hypertension at 24 weeks amenorrhea and vaginal delivery of a stillborn, the second pregnancy was complicated by gestational diabetes on insulin and gestational hypertension discovered at 30 weeks amenorrhea treated with alpha methyl dopa 1500 mg/d and nicardipine 100 mg/d. Clinical examination on admission revealed systolic blood pressure of 140mmhg, diastolic blood pressure of 85mmhg and heart rate of 95 beats/min. The patient had no neurosensory signs, oedema syndrome or proteinuria. Obstetrical examination revealed a two finger dilated cervix, cephalic presentation, intact membranes, positive uterine contractions and negative fetal heart sounds. Obstetrical ultrasound showed normal biometry with no signs of in utero growth retardation and negative cardiac activity. Initial laboratory tests were normal. Four hours later, the patient delivered a macerated stillborn weighing 2900 grams by vaginal delivery without episiotomy. Artificial delivery and uterine revision were performed with antibiotic prophylaxis based on 2 grams of amoxicillin. Placental examination was normal. Immediately postpartum, the patient presented with rapidly worsening respiratory distress, consciousness disorders and cardio respiratory arrest. Basic cardiopulmonary resuscitation was performed with suctioning, recovery of a cardiac rhythm after 3min of cardiac massage, 3mg intraveinous epinephrine and orotracheal intubation. The patient was apyretic, blood pressure 70/40 mm Hg, heart rate 130 beats/min, oxygen saturation70% under 100% of fraction

of inspired oxygen, with bilateral diffuse crackling rales on cardiopulmonary auscultation and hematic secretions on tracheal aspirations. Neurologically, the patient was sedated and showed no signs of convulsions. Obstetrically, the uterine globe was good without bleeding. The patient was put on norepinephrine 0.7g/kg/min, hydrocortisone hemisuccinate 200 mg and Lasilix 120 mg/d. Chest X-ray showed diffuse alveolar interstitial syndrome. Gasometry showed lactic and respiratory acidosis with a partial pressure of oxygen /fraction of inspired oxygen ratio of 50, suggesting acute respiratory distress syndrome. The diagnosis of amniotic embolism could not be ruled out. However, echocardiography revealed no dilatation of the right cavities or signs of pulmonary hypertension with preserved systolic function and absence of intra-cavitary amniotic fluid.

Biological and infectious workup revealed: hemoglobin 8.3g/l, hyperleukocytosis 28,000, platelets 580,000, CRP 54, lactatemia 1.7, prothrombin time 90%, troponin 0.01ng/ml, fibrinogen 3g. Liver, kidney and thyroid function tests were normal. Urine cytobactriological study: leukocyturia 63 elements/mm. Distal bronchial sampling was negative; multiplex PCR negative. In view of the clinical, biological and radiological signs, the patient was put on antibiotic therapy based on Rocephine 2g/d, Ciprofloxacin 400 mg/d, Metronidazole 1g/d and transfused with 2 red blood concentrate, then progressively weaned off vasoconstrictive drugs after control of mean arterial pressure. The evolution was marked by the appearance on the second postpartum day of hypertensive crisis at 200-220 mmhg systolic arterial pressure and 100-120 mmhg diastolic arterial pressure with episodes of tachycardia at 130 beats/min and paroxysmal sweating. The diagnosis of pheochromocytoma was strongly suspected. Adrenal ultrasound was normal. The 24 hour urinary catecholamine metabolite assay was performed. The patient was put on a beta blocker, Loxen 6mg/h with electric syringe and alpha methyl dopa. On the respiratory level, the patient remained hypoxic, with oxygen saturation at 61-90% and oxygen partial pressure at 75, with alveolar hemorrhage, and was tracheotomized on day 3 and transfused with 2 packed red blood cells. Neurologically, she was conscious on cessation of sedation and had no sensory or motor deficits. A thoracic angioscan showed micronodules with hemorrhagic alveolar fillings and no signs of pulmonary embolism.

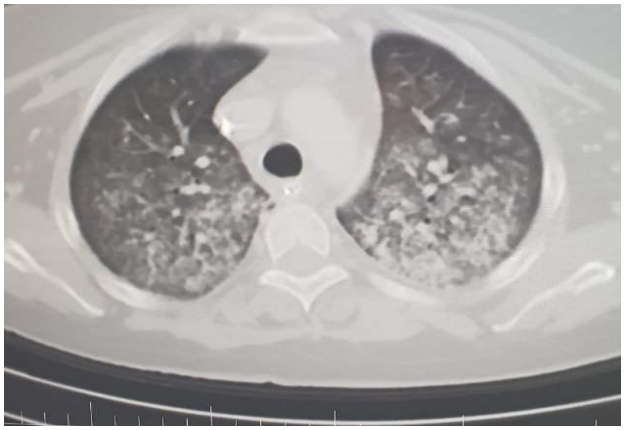

Figure 1: Cross section scanner thoracic showing hemorrhagic alveolar

An abdominal slice incidentally revealed a well limited mass in the outer arm of the left adrenal gland, with regular tissue contours and homogeneous enhancement, measuring 59*59 mm.

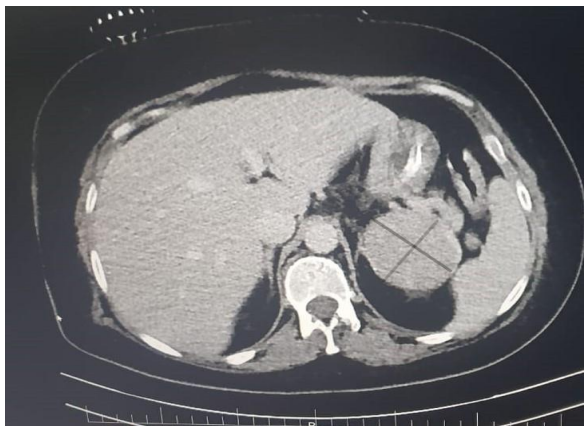

Figure 2: Cross-section of abdominal CT scan showing adrenal pheochromocytoma measuring 59*59mm

Urinary catecholamines were positive, with metanephrines at 1.43 µmol/24h (normal value: 0.2-1µmol/24), normetanephrines increased to 3.26 µmol/24h (normal value: 0.4 - 2.1µmol/24h). The diagnosis of pheochromocytoma was accepted. The patient underwent adjustment of antihypertensive treatment and progressive respiratory weaning, with good progression, before being transferred to the surgical oncology department. A left adrenalectomy removing the tumor was performed without complications. Pathological examination confirmed a benign pheochromocytoma. The evolution was favorable.

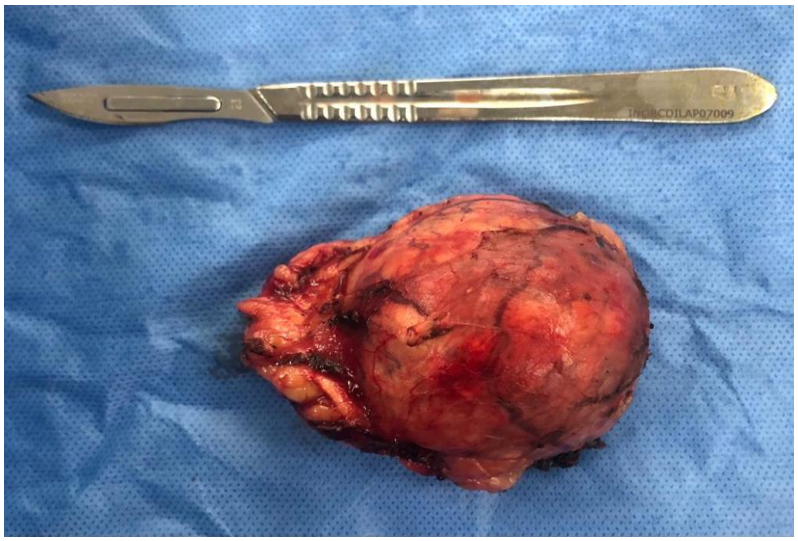

Figure 3: A section of the dissected pheochromocytoma

DISCUSSION

Pheochromocytoma is a chromaffin cell tumor producing excess catecholamines and located in the adrenal medulla in 85 % of cases [5]. Pheochromocytoma are malignant in 10% of cases [5]. The crisis during pregnancy can be triggered by mechanical pressure on the adrenal gland from a growing uterus, uterine contractions, fetal movement, the process of delivery and general anesthesia [6]. Its ambiguous presentation is often mistaken for pre eclampsia, although it may imitate other problems during pregnancy. Several symptoms are associated with pheochromocytoma, none of which is specific, even though arterial hypertension is constant throughout the course of the disease. The classic triad of "headache, palpitations, sweating" is found in 90 % of pheochromocytoma [7]. The diagnosis should be made in the presence of resistant hypertension even in the absence of vasomotor. Manifestations are variable, ranging from severe hypertension to circulatory failure and shock. Myocardial ischemia or cardiomyopathy, pulmonary edema, encephalopathy and multiorgan failure have all been reported [6, 8, 9]. Classically, plasma and urine determination of catecholamines (adrenaline, noradrenaline, and dopamine) and their urinary metabolites [metanephrine, normetanephrine, and vanylmandelic acid] is used to confirm the diagnosis [10]. Normal pregnancy does not alter urinary metanephrine values. The diagnosis of pheochromocytoma requires a combination of biochemical and anatomical confirmation. Localisation of tumor can be accomplished by either Computed tomography or magnetic resonance imaging of the abdomen [11, 12]. Sensitivities are comparable, although magnetic resonance imaging is preferable in pregnancy because of minimal radiation exposure [11, 12]. Pheochromocytoma once diagnosed, becomes an indication for surgery, however if associated with pregnancy, the timing of surgery remains debatable. Factors which have a bearing on management are the gestation period, preoperative medical optimization. Maternal and fetal outcomes are seen to be favorable if surgical intervention is done before the 24th week of gestation as pointed out by Kalra *et al*., [13]. Beyond this period, the uterus size may act as a deterrent to proper tumor assessment and removal. So, it becomes imperative to wait till fetal maturity for proper disease control and cesarean delivery followed by tumor removal. Cesarean section is the favored mode of delivery though some controversies do exist as successful vaginal deliveries have also been reported. The proponents of cesarean delivery claim that the uterine contraction and fetal movements aggravate the disease by release of catecholamines during vaginal delivery [13, 14]. However, after 24 weeks of gestation, the patient can be treated with the appropriate α adrenergic blockade until the fetus is viable, when the tumor can be removed after an elective cesarean section [15, 16].

CONCLUSION

Pheochromocytoma is a rare but importante cause of hypertension in pregnancy. This case report underscores the rarity and diagnostic challenges

associated with pheochromocytoma during pregnancy and postpartum. The subsequent identification of pheochromocytoma, though initially suspected, was confirmed through a meticulous diagnostic process involving urinary catecholamines, imaging, and clinical correlation. Ultimately, this case report contributes valuable insights to the medical community, reinforcing the significance of early identification, prompt management, and collaborative decision-making in optimizing outcomes for pregnant individuals with pheochromocytoma.

INFORMED CONSENT: Clear consent was obtained from the patient before publication of this observation.

ETHICAL APPROVAL: As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

Declarations: Authors have declared that no competing interests exist.

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