RECURRENT HYDATIDIFORM MOLE COMPLICATED BY TOXIC GOITRE.

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INTRODUCTION

Hyperthyroidism complicates few pregnancies, in many cases due to Graves' disease .[1,2] Gestational trophoblastic disease is a rare cause of hyperthyroidism in which high levels of hCG causes activation of the thyrotrophin receptor to stimulate supraphysiological secretion of thyroid hormone with or without thyroid gland enlargement[1].

Molar pregnancies are usually not recurrent,[3] however, women with a previous hydatidiform mole (HM) are at higher risk of having a second mole than women from the general population.[3,4] After a prior molar pregnancy, the risk of having a second one is 540 times that of the general population,[5] however familial molar pregnancies are exceedingly rare.[6]

Here we present a case of recurrent HM complicated by a toxic goiter in a patient with family history of molar pregnancy

CASE REPORT

Mrs ZB is a 37-year-old woman who was admitted to the gynecologic ward of the University of Maiduguri Teaching Hospital, with excessive vomiting and irregular bleeding per vaginum following amenorrhea of 13 weeks. There was no passage of vesicles, no cough. She was a fifth gravida with history of two previous molar pregnancies. There was a family history of molar gestation in two of her siblings but no family history of thyroid or autoimmune disorders. There was no history of consanguineous marriage in her or in her siblings. There was also no history of neck radiation, neck pain, fever, ophthalmopathy or goiter.

Her general physical examination revealed no abnormality except for a neck swelling. Her pulse was 108 and regular, blood pressure was 130/60 mmHg and the axillary temperature was 36.7^{0}C. Her blood group was 'B' Rhesus positive.

Thyroid examination revealed a diffused enlargement of the thyroid gland. She has mild proximal muscle weakness involving the shoulder girdle and her palms were warm. No tremors, ophthalmopathy, or peripheral stigmata of Grave's disease.

On pelvic examination, the uterus was doughy and 18 weeks' gestational size and the cervical Os was closed. There was minimal bleeding through the cervix. There was no adnexal mass.

Laboratory studies revealed normal complete blood cell count and electrolytes. Her urine pregnancy test in serial dilution was positive up to 1: 1000 dilutions. β-human chorionic gonadotrophin (β-hCG) level was elevated at 159,845 IU/L and subsequent thyroid biochemistry revealed hyperthyroidism: serum thyroid

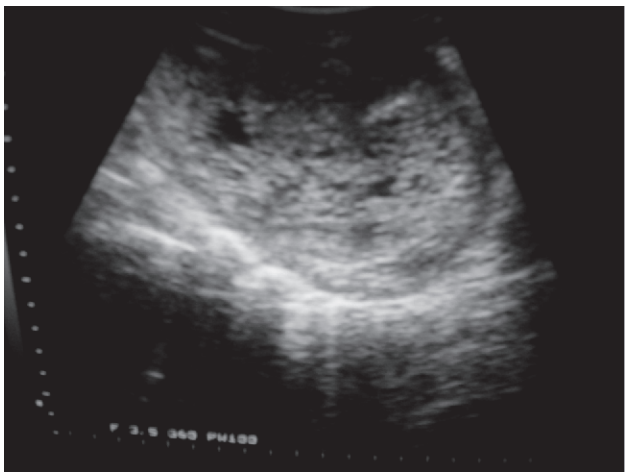

Figure 1: *Longitudinal ultrasound image of the uterus showing an intrauterine mass with heterogeneous echopattern (snowstorm) suggestive of molar gestation.*

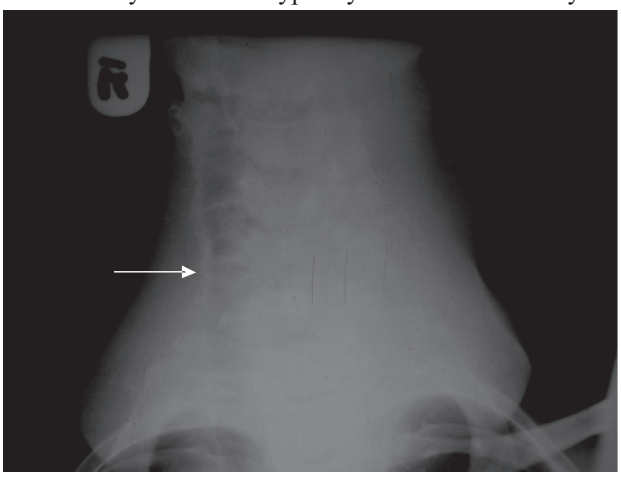

Figure 2: *Anterior-posterior radiograph of soft tissue neck demonstrating huge anterior neck swelling involving the root of the neck, more towards the left with tracheal deviation to the right (arrows).*

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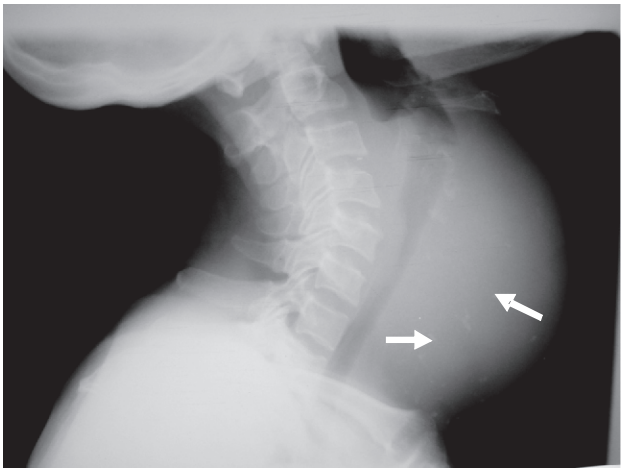

Figure 3: *Lateral soft tissue radiograph of the neck demonstrating huge anterior neck swelling with tracheal compression. Flecks of calcifications are noted within the soft tissue mass (arrows).*

stimulating hormone (TSH) suppressed at <0.05 mU/L (Normal 0.24.5), with a total thyroxine (T4) of 316 nmol/L (Normal 71148), and Free triiodothyronine (fT3) of 17.8 nmol/L (Normal 2.66.2).

Ultrasound examination was in keeping with molar pregnancy because of the uniform snowstorm appearance in the uterine cavity and absence of fetal parts (Fig 1), no adnexal mass was seen. Soft tissue radiographs of the neck (Fig. 2&3) showed a uniform soft tissue mass involving the anterior neck, with compression and displacement of the trachea at that level. Punctate calcific opacities were present within the mass. There was no retrosternal extension.

A provisional diagnosis of complete hydatidiform mole with toxic goiter was made and patient was admitted, stabilized, and treated with suction evacuation. Histology of the products confirmed hydatidiform mole. On follow up, the pregnancy test in serial dilution became negative within 8 weeks of evacuation. She had sub-total thyroidectomy after she was rendered euthyroid and repeat thyroid function tests done after the surgery were normal.

DISCUSSIONS

Hydatidiform moles (HM) are abnormal pregnancies characterized histologically by aberrant changes within the placenta.[6] The typical clinical presentation of molar pregnancies has changed with the advent of high-resolution ultrasonography. Most moles are now diagnosed in the first trimester before the onset of the classic signs and symptoms of anaemia, hyperemesis gravidarum, preeclampsia, and uterine sizes in excess of that predicted for gestational age. The most common symptom of a HM is vaginal bleeding [1-5] due to molar tissue separating from the decidua, causing bleeding. This symptom occurs in 97% of cases.[2] The uterus may also become distended by large amounts of blood, and dark fluid may leak into the vagina. This patient present with irregular vaginal bleeding and had a large for date uterine size.

Recurrent HM is a rare phenomenon and has been reported in only 0.62.6% of cases. [3] It has significant clinico-pathological implications, which include risk of malignant sequelae and subsequent poor reproductive performance. In a study carried out by Moglabey et al,[3] women with recurrent HM were homozygous for a rare autosomal recessive gene. This case is reported because of two rare clinical situations; recurrence of HM in a patient with family history of molar gestation and the development of hyperthyroidism. There is a need to know the prognosis, regarding malignant transformation and future obstetric outcome in patients with such a bachground.

A familial HM syndrome has been described in the literature. [3,7] A higher incidence of subsequent molar pregnancies has been reported to occur with extensive intermarriages, suggesting a defect in HLA histocompatibility.[7-9] Since there was no history of consanguineous marriage in our patient, could this be another familial hydatidiform mole syndrome?

Ultrasonography is the standard for identifying molar pregnancies. The classic image, shows a complex intrauterine mass containing many small cysts (snowstorm pattern).[8,9] Ultrasonography confirmed the diagnosis in this patient.

This patient had toxic goiter most likely because the elevated ß-hCG have greater ability to bind to TSH receptors leading to hyperstimulation of the thyroid gland. Her symptoms resolved after suction evacuation and normalization of ß-hCG. She had thyroidectomy after stabilization and her thyroid function normalized.

Lastly, the future obstetric management of this woman needs to be discussed. In women with molar pregnancy, the recommendation for any subsequent pregnancy is to have a pelvic ultrasonogram during the first trimester to confirm normal gestational development. This option alone is might not be enough for our patient, who had two molar pregnancies already. The possibility of intracytoplasmic sperm injection (ICSI) can be considered in this case, as it has been reported to decrease the chances of molar pregnancy.[10] However, it remains to be seen how much benefit can be obtained by this intervention, as the pathogenesis of familial recurrent mole may be different from a sporadic molar event.[9]

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