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Spontaneous acute subdural haemorrhage, cerebral and pulmonary metastases in a complete mole

Agrawal A, Agrawal C S, Kumar A, Kumar M, Yadav R

ABSTRACT

Complete and partial moles remit spontaneously in most cases, following evacuation of the uterine cavity. However, either persistent trophoblastic disease or a frank trophoblastic tumour can follow a complete hydatidiform mole. To our knowledge, acute subdural haematoma, as a complication of cerebral metastases, following treatment for hydatidiform mole has not been reported. We describe a 29-year-old woman who presented with spontaneous acute subdural haemorrhage and pulmonary metastases, eight months after evacuation of a complete hydatidiform mole, with a fatal outcome.

Keywords: cerebral metastases complication, gestational trophoblastic disease, hydatidiform mole, subdural haematoma

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INTRODUCTION

Gestational trophoblastic tumours (GTT) are unique in oncology in that they follow either a normal or abnormal pregnancy. The most common antecedent pregnancy to GTT is a complete or partial hydatidiform mole. Both complete and partial moles remit spontaneously in most cases, following evacuation of the uterine cavity. However, either persistent trophoblastic disease or a frank trophoblastic tumour can follow a complete or partial hydatidiform mole, with an incidence of approximately 8% and 0.5%, resepectively.[1] To our knowledge, acute subdural haematoma as a complication of cerebral metastases following treatment for hydatidiform mole, has not yet been reported. We describe a young woman who presented with spontaneous acute subdural haemorrhage and pulmonary metastases, eight months after evacuation of a complete hydatiform mole, with a fatal outcome.

CASE REPORT

A 29-year-old, right-handed housewife presented with a history of sudden loss of consciousness of six hours' duration, preceded by sudden onset of headache associated with two episodes of vomiting. She also had left-sided hemiplegia. There was no history of seizure or fever. She had undergone hysterectomy with bilateral salpingo-oophorectomy for complete mole two months previously at six month's gestation (Fig. 1). She was also under treatment for malignant pleural effusion for the past two weeks (Figs. 2 & 3).

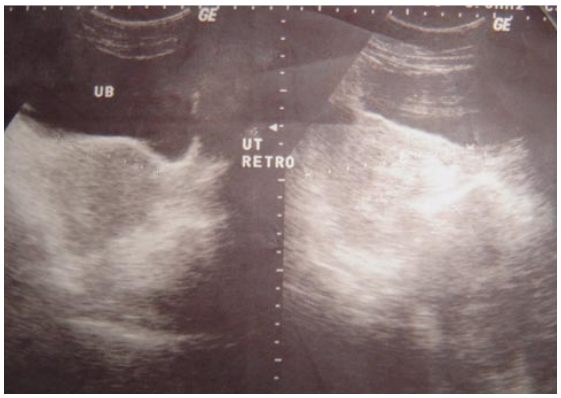

Fig. 1 US image of the pelvis shows a snowstorm appearance, suggestive of hydatidiform mole.

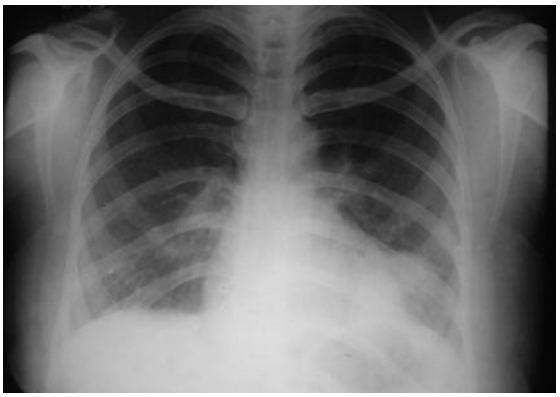

Fig. 2 Chest radiograph shows a mass lesion involving the left lower lobe, with evidence of pleural effusion.

Department of Surgery, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Wardha 442005, Maharashtra, India

Agrawal A, MCh
Associate Professor in Neurosurgery

Agrawal CS, MS
Professor

Kumar A, MS
Professor

Kumar M, MS
Associate Professor

Yadav R, MS
Senior Resident

Correspondence to:
Dr Amit Agrawal
Tel: (91) 7152 395 6552 ext 2047
Fax: (91) 7152 224 5318
Email: dramitagrawal@gmail.com

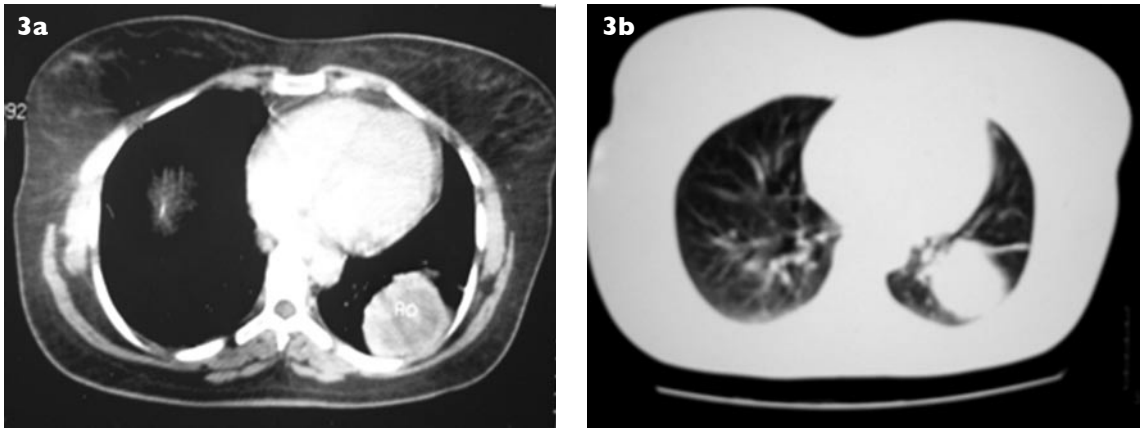

Fig. 3 Enhanced axial CT images of the chest taken in (a) mediastinal, and (b) lung windows show a well-defined mass lesion involving the left lower lobe.

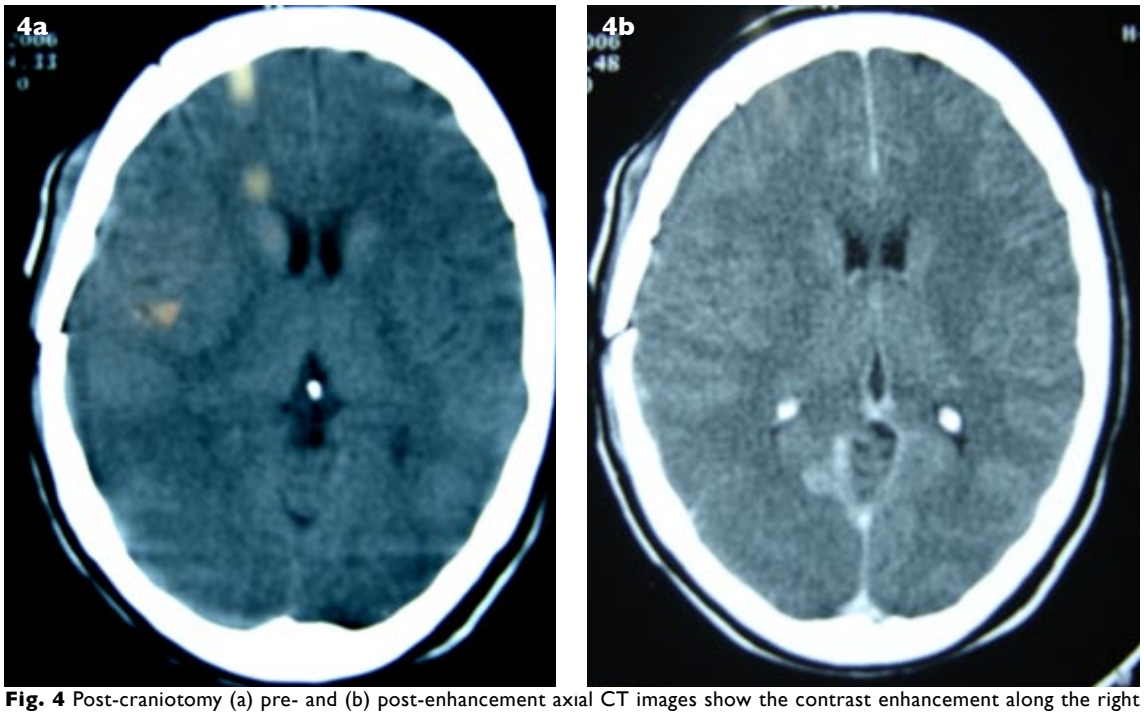

Fig. 4 Post-craniotomy (a) pre- and (b) post-enhancement axial CT images show the contrast enhancement along the right side of the tentorial hiatus with a nodule.

Her general examination was unremarkable. She had decreased air entry on the left side of the chest with a dull percussion note. Neurologically, she had an altered sensorium with GCS of E1V5M2 associated with left hemiparesis of grade I/V and was localising on the right side. Pupils were dilated; the right pupil was fixed, suggestive of uncal herniation, but the left pupil reacted normally. Her CT of the brain showed an acute subdural haematoma in the right fronto-temporo-parietal region with a significant mass effect and midline shift (1.8 cm). Complete blood examination and coagulation profile were within normal limits. She underwent emergency evacuation of the subdural haematoma. There was a thick subdural clot with mucinous material. The brain was tense and pulsatile, and dura could be closed with pericranial graft. Postoperative beta-hCG was very high (3,260 mIU/dL). Contrast CT perfomed for brain metastases showed diffuse meningeal metastases along the tentorial hiatus and tentorium cerebelli (Figs. 4 & 5). She was gradually recovering in her sensorium; on the third day, she could open her eyes, and speak coherently. However, on the seventh day, she lapsed into an altered sensorium and did not recover.

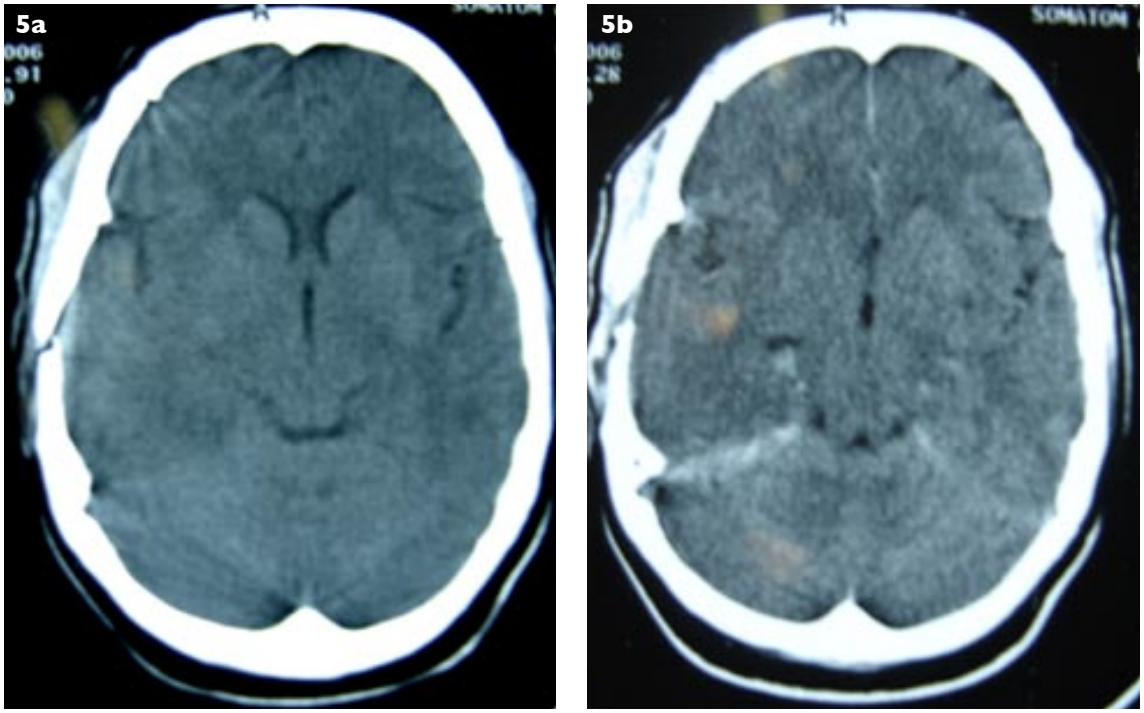

Fig. 5 Post-craniotomy (a) pre- and (b) post-enhancement axial CT images show contrast enhancement along the petrous attachment of side of tentorial cerebelli.

DISCUSSION

Hydatidiform mole is classified into partial and complete subtypes, according to histopathological and genetic criteria.[2] GTT, particularly choriocarcinoma, while rare in developed countries, is still a major problem in developing countries, especially in Southeast Asia, and can present with metastases at various sites, including unusual sites.[3] GTT patients presenting within a few months of delivery have widespread pulmonary and, not uncommonly, cerebral metastases. Lung metastases, presenting as pneumothorax and haemothorax, have been reported in the literature. Presentation may occur several years after pregnancy, usually with persistent or irregular uterine bleeding.[1,4-7] In this case, the patient had haemoptysis and subsequently developed pleural effusion, which was refractory to treatment, and became symptomatic for cerebral lesions. Metastatic lesions after hysterectomy for hydatidiform mole should be suspected when there is persistently high beta-hCG levels, as was seen in this case.[8] The use of a chest radiograph was considered adequate for the detection of lung metastases, but lesions can be missed on conventional chest radiographs. CT of the chest is recommended if the chest radiograph is negative, especially if the clinical findings point to a diagnosis of malignant GTT.[9] Metastases from this unusual tumour stimulate virtually no stromal reaction and is therefore essentially a mixture of haemorrhage and necrosis with tumour cells scattered within the mass. Tumour cells can be scanty and present problems of pathological interpretation, as in this case where we could not demonstrate the tumour cells.[3]

Established classifications divide GTT, including choriocarcinoma, into cases with "high" and "low" risks to facilitate the management.[8] Recently, a new staging system has been proposed for GTT, to ensure uniformity in staging as well as to facilitate comparison of treatment outcomes. The scoring, and therefore treatment, according to high risk or low risk depends on various risk factors, including the number of metastases.[9] Without respect to histology in "high-risk" cases, including cerebral and pulmonary metastases, polychemotherapy with good response has been reported.[8] However, the key factors for successful outcome seem to be early diagnosis and vigorous therapy.[10] Although the development of effective chemotherapy has resulted in an improved survival of patients with GTT tumour, lung lobectomy is an important adjunct treatment in a selected subset of patients with pulmonary metastases.[11] Poor prognosis for gestational trophoblastic disease can also be primarily treated by multi-agent chemotherapy, with adjunctive surgery and radiation therapy in selected patients. Unsuccessful chemotherapy prior to treatment, a prolonged interval from the antecedent pregnancy to treatment, and liver metastases portended a worse prognosis in patients with widespread

disease.[12] Irradiation has a distinct therapeutic role in the treatment of central nervous system metastases. There is evidence that the irradiated brain tends to resist recurrent disease, even in patients whose outcomes are fatal.[13] However, in the present case, the disease was at an advanced stage and could not be controlled. In future, we hope that better treatment modalities and chemotherapeutic agents will help to better control the disease and improve patient survival.

REFERENCES

1. Tidy JA, Rustin GJ, Newlands ES, et al. Presentation and management of choriocarcinoma after nonmolar pregnancy. Br J Obstet Gynaecol 1995; 102:715-9.
2. Cheung AN, Khoo US, Lai CY, et al. Metastatic trophoblastic disease after an initial diagnosis of partial hydatidiform mole: genotyping and chromosome in situ hybridization analysis. Cancer 2004; 100:1411-7.
3. Prabhu MM, Baxi J, Malhothra A, Setty M, Sharma P. A perplexing case of gastrointestinal haemorrhage. Singapore Med J 2006; 47:159-62.
4. Sudduth CD, Strange C, Campbell BA, Sahn SA. Metastatic choriocarcinoma of the lung presenting as hemothorax. Chest 1991; 99:527-8.
5. Ouellette D, Inculet R. Unsuspected metastatic choriocarcinoma presenting as unilateral spontaneous pneumothorax. Ann Thorac Surg 1992; 53:144-5.
6. Santhosh-Kumar CR, Vijayaraghavan R, Harakati MS, Ajarim DS. Spontaneous pneumothorax in metastatic choriocarcinoma. Respir Med 1991; 85:81-3.
7. Altiparmak MR, Pamuk ON, Pamuk GE, Ozbay G. Membranous glomerulonephritis in a patient with choriocarcinoma: case report. South Med J 2003; 96:198-200.
8. Honig A, Rieger L, Kristen P, et al. A case of metastasizing invasive hydatidiform mole. Is less--less good? Review of the literature with regard to adequate treatment. Eur J Gynaecol Oncol. 2005; 26:158-62.
9. Moodley M, Moodley J. Evaluation of chest X-ray findings to determine metastatic gestational trophoblastic disease according to the proposed new staging system: a case series. J Obstet Gynaecol. 2004; 24:287-8.
10. Weed JC, Hammond CB. Cerebral metastatic choriocarcinoma: intensive therapy and prognosis. Obstet Gynecol. 1980; 55:89-94.
11. Ren T, Xiang Y, Yang XY, Wan XR, Zhang ZY. [Evaluation of surgical resection of pulmonary metastasis of trophoblastic tumour]. Zhongguo Yi Xue Ke Xue Yuan Xue Bao. 2003; 25:418-21. Chinese.
12. Surwit EA, Hammond CB. Treatment of metastatic trophoblastic disease with poor prognosis. Obstet Gynecol. 1980; 55:565-70.
13. Yordan EL, Schlaerth J, Gaddis O, Morrow CP. Radiation therapy in the management of gestational choriocarcinoma metastatic to the central nervous system. Obstet Gynecol. 1987; 69:627-30.