The Journal of Laryngology and Otology
October 1992, Vol. **106**, pp. 917–922

Metastatic choriocarcinoma of the maxilla: an unusual cause of severe intractable epistaxis

BHARATH SINGH, M.Med. (Otol), (Durban, South Africa)

Abstract
A fifth case of metastatic choriocarcinoma to the maxilla presenting an intractable epistaxis in a 24-year-old African female, is described.

Introduction

Although there are numerous known causes of epistaxis, in the majority of patients the cause is not found. Therefore, any patient who presents with epistaxis, but with a normal coagulation, is said to have idiopathic epistaxis.

The younger patients with idiopathic epistaxis usually present with a minor bleed which is easily controlled with either anterior nasal pack or cautery, whilst the elderly and the hypertensive patients present with intractable epistaxis which is not controlled with anterior packing, and often these patients require either anterior-posterior nasal packing or arterial ligation.

If a young non-hypertensive patient presents with epistaxis, then the diagnosis of idiopathic epistaxis must be reviewed.

Case report

A 24-year-old African female was referred from one of the peripheral hospitals on 24.9.87, with a two-day history of severe epistaxis. On presentation, she was not bleeding actively. Blood clots were sucked from the nostrils and anterior nasal packs were placed. Full blood count revealed a haemoglobin of 9.2 g/dl., platelet count of 359,000 and white cell count of 7,000 per ml. The prothrombin index and partial thromboplastin time were within normal limits. The epistaxis continued intermittently for two weeks and four units of packed cells were transfused during this time. Finally, the bleeding stopped and she was discharged on 9.10.87.

Five weeks later, on 16.11.87, she was readmitted but to the medical wards, with a two-day history of bleeding from the nose, mouth and vagina and swelling of the left cheek. A provisional diagnosis of either Burkitt's lymphoma or leukaemia was made. Haematological investigations were all within normal limits. The next day a large necrotic bleeding ulcer was noted on the palate and an urgent ENT consultation was requested. On examination, an ulcerative tumour was found to involve the left side of the palate and alveolus of the maxilla (Fig. 1); there was also swelling of the left cheek and a mass in the left nostril. A trucut biopsy was taken through the mouth and urgent histology was requested.

A CT scan of the sinuses (Fig. 2) revealed opacification of the left maxillary sinus with destruction of the medial, lateral

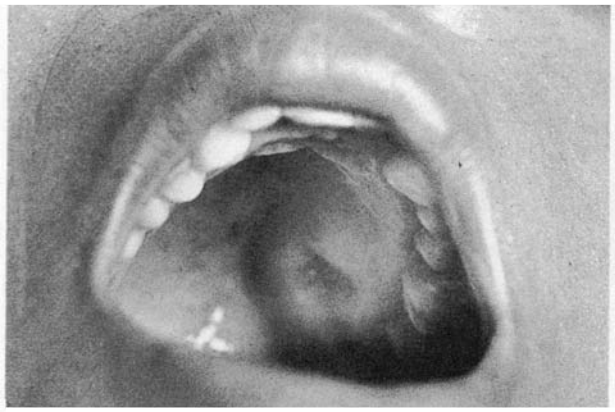

FIG. 1
Tumour of the palate with central area of ulceration.

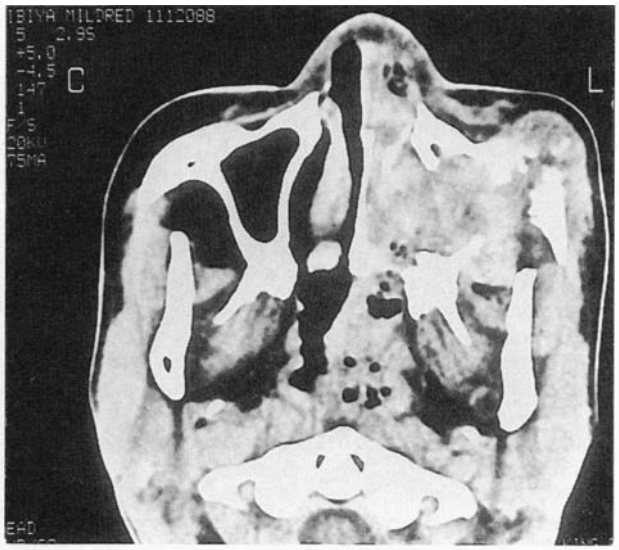

FIG. 2
CT showing tumour of the left maxillary sinus with destruction of medial, lateral and anterior walls.

Accepted for publication: 8 July 1992.

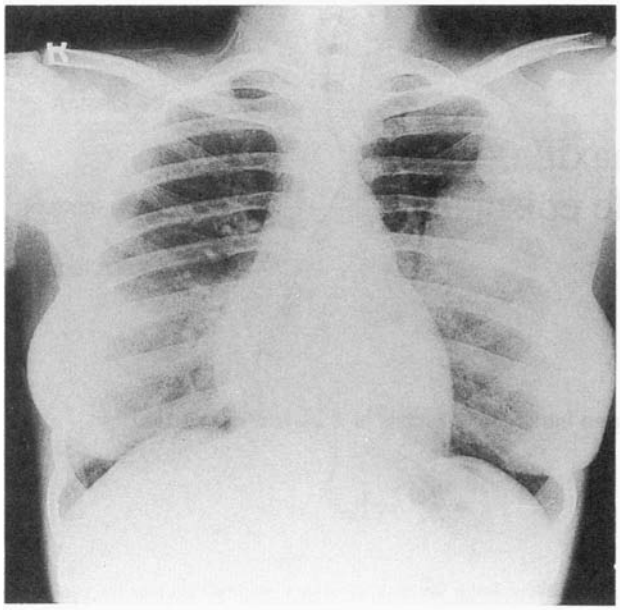

FIG. 3
Chest radiograph displaying metastatic choriocarcinoma in the left upper lobe.

and anterior walls, and extension of the tumour into the nasal cavity and soft tissue of the face. Chest radiograph showed two rounded opacities in the left upper lobe (Fig. 3).

The provisional report from the pathologist was that this was a high-grade malignant tumour. As the tumour was bleeding continuously and briskly, it was decided to irradiate the maxilla with a single dose of 10 Gy. Despite this, the bleeding continued and it was thought, at this stage, that this might be a lymphoma, but this was excluded on aspiration cytology of the bone marrow.

On 30.11.87 the patient began bleeding from a nodule on the left parieto occipital region of the scalp. This was biopsied and irradiated with 10 Gy. The histology of the nodule was that of metastatic choriocarcinoma (Fig. 4-6b). This diagnosis was confirmed by finding a high titre of beta-human chorionic gonadotropin (B-HCG) of 124 000 IU/ml in the serum. Subsequently the histology of the first biopsy from oral cavity was reviewed and was consistent with choriocarcinoma (Fig. 7). The gynaecological history revealed that she had two children. Her last delivery was in December, 1986, and her periods were normal until August, 1987. Thereafter her periods stopped, then she had vaginal bleeding in November.

Vaginal examination revealed a normal vagina, 3cm long cervix, normal size uterus and absence of adnexial or rectal masses. These findings were confirmed on ultra-sonography.

Angiography revealed tumour circulation in the left hemipelvis. The radiologist was not sure whether this was a primary within the uterus or a secondary in the pelvis.

Chemotherapy, consisting of vincristine, cisplatin, methotrexate and etoposide, was commenced on 5.12.87. This was repeated at three-weekly intervals. After six courses of chemotherapy, the oral lesion and scalp lesion had completely disappeared.

On 17.4.88, a tender swelling appeared over the left cheek. This settled on antral wash-out and antibiotics. Cytology of the fluid failed to show any malignant cells.

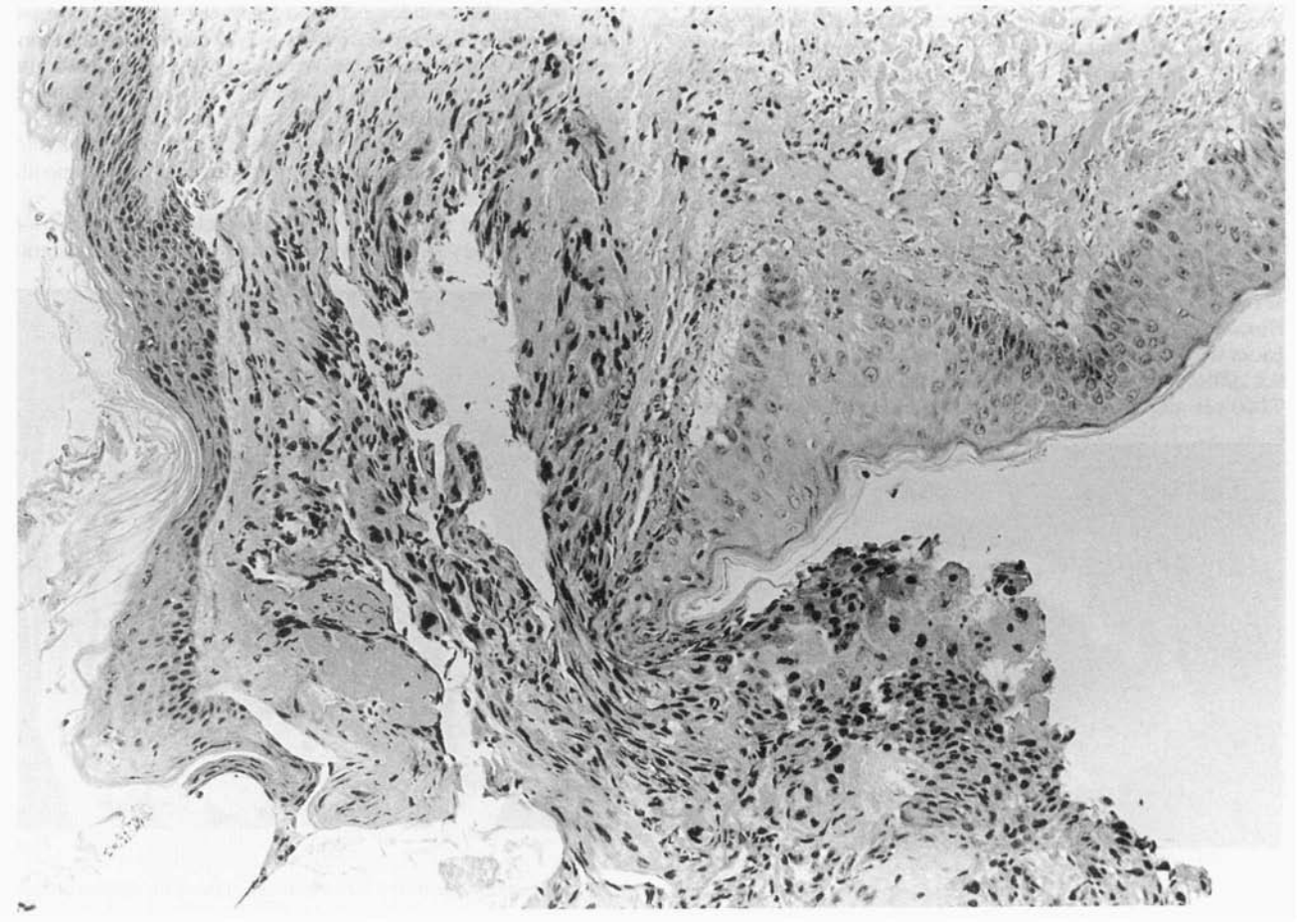

FIG. 4
Metastatic choriocarcinoma of the scalp. H&E — low power. Strongly positive with Human Chorionic Gonadotrophin (HCG) and less strongly positive with Human Placental Lactogen (HPL).

https://doi.org/10.1017/S0022215100121280 Published online by Cambridge University Press

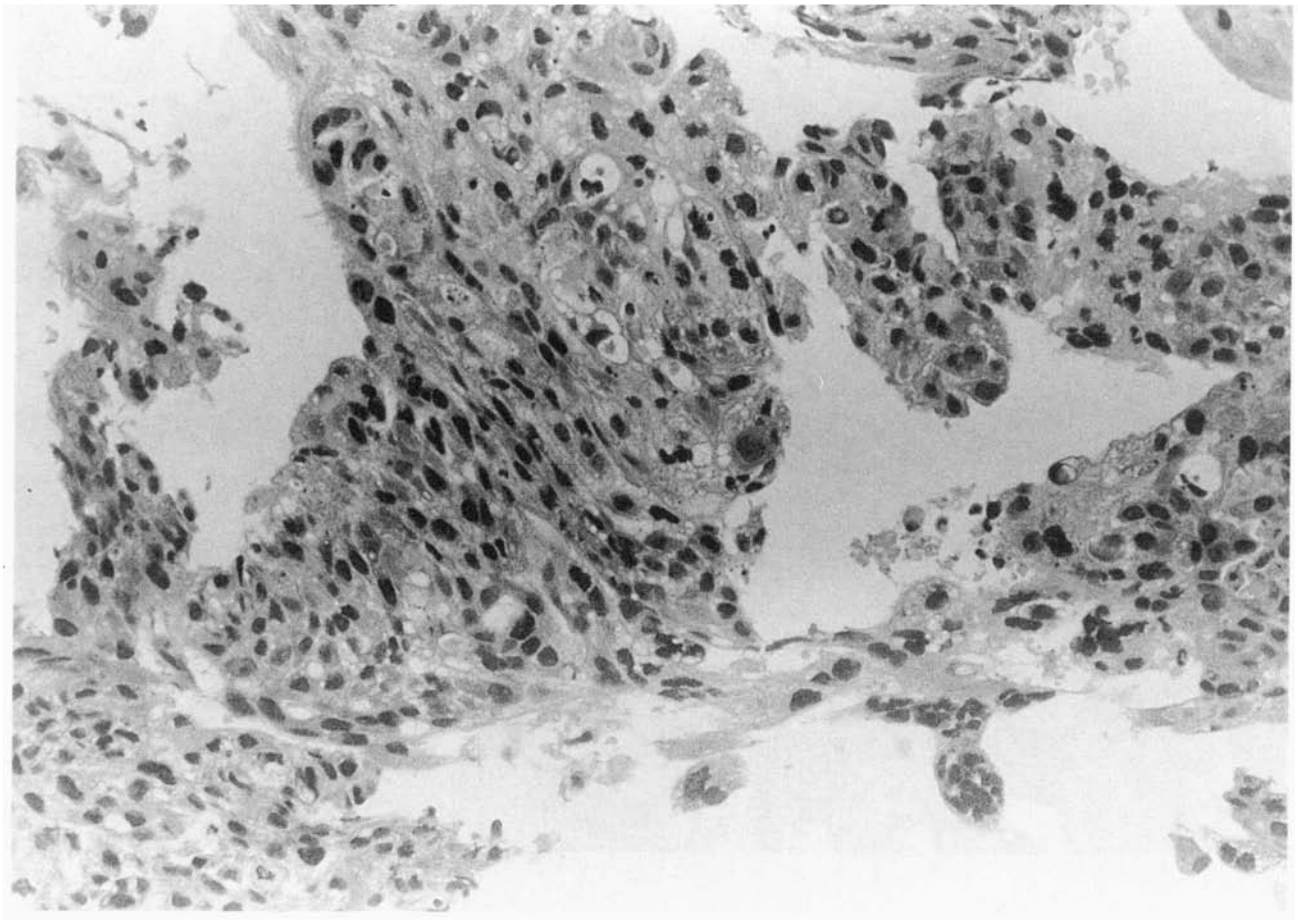

FIG. 5

Metastatic choriocarcinoma of the scalp showing plexiform pattern of syncytiotrophoblastic and cytotrophoblastic elements (H&E — high power). Strongly positive with HCG moderately positive with HPL.

On 4.7.88 the swelling recurred. The maxillary sinus was opened and necrotic debris was evacuated. The mucosal lining of the maxillary sinus was biopsied and histological analysis failed to show any evidence of malignancy.

On 28.7.88, the pulmonary lesions had disappeared. After 11 courses of chemotherapy, the B-HCG decreased to 10 IU/ml. The patient was discharged and asked to return after three weeks but she defaulted and only returned 13 months later, with recurrence of the tumour in the maxilla and severe epistaxis. She was transfused 11 units of packed cells and two units of whole blood before being transferred from the peripheral hospital. The B-HCG was 204 300 IU/ml. Cytotoxic treatment was commenced and the epistaxis subsided. Chest radiograph revealed recurrence of the secondaries.

A CT scan of the maxilla, abdomen and pelvis on 30.10.89 showed recurrence of tumour in the maxilla, slightly bulky and irregular uterus, and a low density area over the liver and right kidney. Ultrasound of the abdomen excluded a liver secondary but confirmed a 3.5cm diameter secondary deposit on the right kidney.

On 23.2.90 pelvic angiography was repeated and it showed complete absence of tumour circulation.

After completing 14 courses of chemotherapy, she was discharged and asked to return in two weeks, but again, she defaulted. She was readmitted three months later (19.9.90) with a left-sided convulsion and hemiparesis.

CT scan of the brain showed multiple intracranial secondaries (Fig. 8).

Chemotherapy was commenced but unfortunately this did not help. She died on 26.9.90.

Discussion

Choriocarcinoma is a malignant tumour of the fetal trophoblast and is composed of two cell types, the syncytiotrophoblast and cytotrophoblast.

In the female it is closely associated with pregnancy; about 57 per cent follow molar pregnancy, 26 per cent normal pregnancy and 17 per cent either ectopic pregnancy or abortion (Buckley, 1984). The most striking feature of choriocarcinoma is the diverse clinical features, brought on by haematological spread to multiple sites very early in the course of the disease. Although vaginal bleeding is the commonest presenting symptom, often the first sign of the disease is its secondary manifestation.

There have been only four reported cases of metastases to the maxilla, one each by Ramanathan *et al.* (1968), Bakeen *et al.* (1976), Nespeca and Sass (1980) and Schaffner *et al.* (1982). The patient presented here differs from those reported in the literature in that she is the only patient to have extensive bony destruction of the maxilla and to present with severe intractable epistaxis. Another unique feature about this patient is the metastatic scalp lesion. There are two other reported cases in the literature (Cosnow and Fretzin, 1974; Catania, 1953). Intractable epistaxis is a disease of the hypertensive and elderly patient but when it occurs in a young healthy patient, it must be viewed

https://doi.org/10.1017/S0022215100121280 Published online by Cambridge University Press

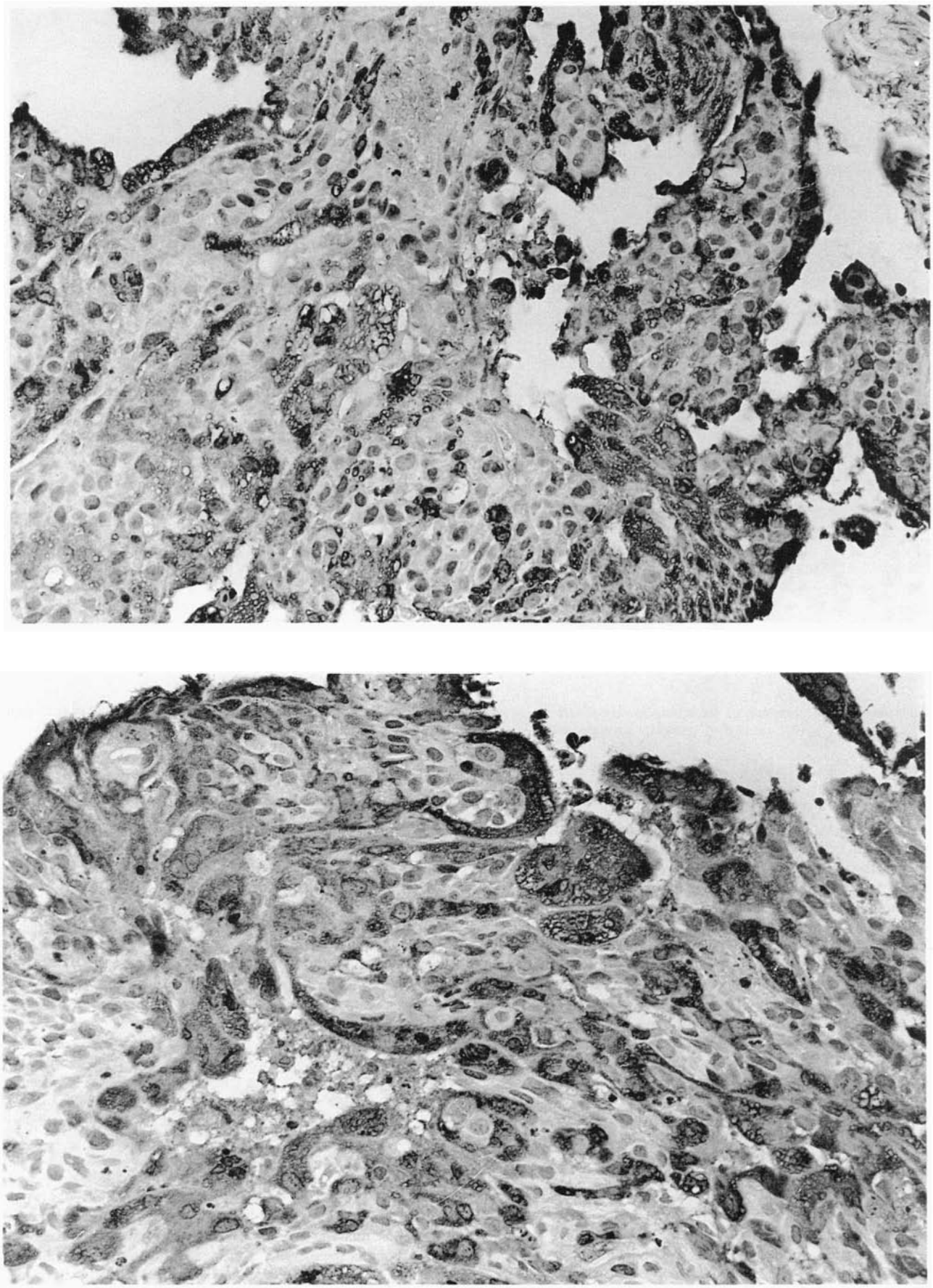

FIG. 6a&b

a. Choriocarcinoma composed of sheets of cytotrophoblasts and syncytiotrophoblasts. Positive reaction to HPL. ×400.
b. Choriocarcinoma showing syncytiotrophoblasts reacting to HCG. ×400.

https://doi.org/10.1017/S0022215100121280 Published online by Cambridge University Press

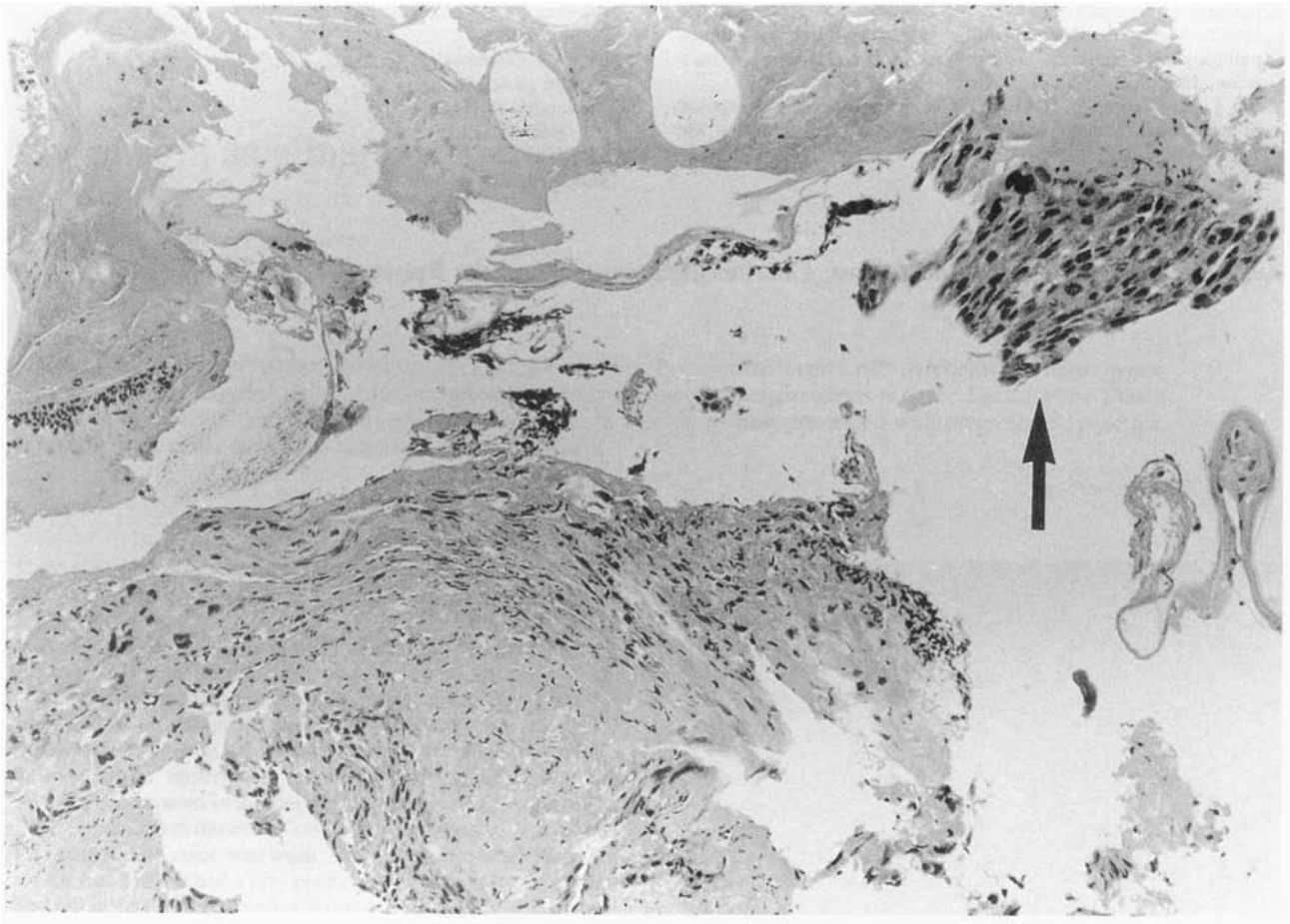

FIG. 7
Metastatic choriocarcinoma of the maxilla (H&E — low power).

with a great deal of suspicion. In this patient, the tumour went unnoticed during the first admission because no radiological investigations were requested. Plain radiographs of the paranasal sinuses are not routinely done in patients with epistaxis because tumours have been found to be a rare cause of epistaxis; Juselius (1974) found tumours in 0.3 per cent of his patients; Hara (1962) 0.7 per cent and Wang and Vogel (1981) 1.8 per cent.

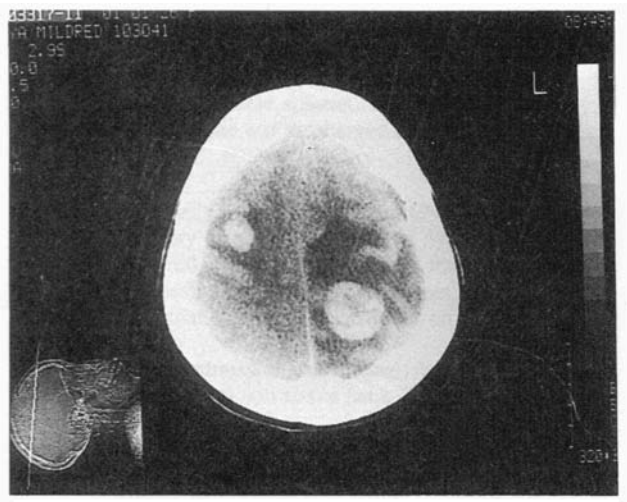

FIG. 8
CT scan of the brain displaying multiple intracranial metastases.

Conclusion

Idiopathic epistaxis is a disease of the hypertensive and elderly patients. If it occurs in a young, healthy individual, then it is no longer considered idiopathic and causes, such as bleeding disorders and tumours must be excluded. Therefore, the very basic investigation in this type of patient, besides coagulation profile must include radiographs of the paranasal sinuses and chest.

References

Bakeen, G., Hiyarat, A. M., Al-Ubaidy, S. S. (1976) Chorioepithelioma presenting as a bleeding gingival mass. *Oral Surgery, Oral Medicine, Oral Pathology,* **41:** 467–471.

Buckley, J. D. (1984) The epidemiology of molar pregnancy and choriocarcinoma. *Clinical Obstetrics and Gynaecology,* **27:** 153–159.

Catania, A. F. (1953) Three non-vital teeth associated with chorioepithelioma: Report of Case. *Journal of Oral Surgery,* **11:** 324–330.

Cosnow, I., Fretzin, F. (1974) Choriocarcinoma metastatic to skin. *Archives of Dermatology,* **109:** 551–553.

Juselius, H. (1974) Epistaxis a clinical study of 1724 patients. *Journal of Laryngology and Otology,* **88:** 317–327.

Hara, H. J. (1962) Severe epistaxis. *Archives of Otolaryngology,* **75:** 258–269.

Nespeca, J. A., Sass, J. K. (1980) Choriocarcinoma metastatic to maxillary gingiva. *Journal of Oral Surgery,* **38:** 534–537.

Ramanathan, K., Eravelly, J., Ken, T. P. (1968) Metastatic choriocarcinoma involving the maxilla. *Oral Surgery, Oral Medicine, Oral Pathology,* **26:** 688–693.

https://doi.org/10.1017/S0022215100121280 Published online by Cambridge University Press

Schaffner, D. L., McKinney, R. V., Braxton, M., Hollyfield, N. (1982) Metastatic disease of the jaws simulating periodontal pathological conditions. *Journal of American Dental Association,* **105:** 809–812.
Wang. L., and Vogel, D. H. (1981) Posterior epistaxis: Comparison of treatment. *Otolaryngology — Head and Neck Surgery,* **89:** 1001–1006.

Address for correspondence:
B. Singh,
Dept. of Otorhinolaryngology,
Faculty of Medicine,
University of Natal,
P.O. Box 17039,
CONGELLA. 4013
South Africa.

Key words: Maxillary sinus neoplasms; Chorioncarcinoma, metastasis; Epistaxis.