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Metastatic testicular teratoma of the nasal cavity: a rare cause of severe intractable epistaxis

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Abstract
Malignant neoplasms of the nasal cavity and paranasal sinuses are uncommon. Choriocarcinoma is a highly malignant germ cell tumour occurring in the reproductive organs. Metastasis may be principally by the lymphatic route as in other germ cell tumours but choriocarcinoma is also known to spread haematogenously. We present a rare case of metastatic choriocarcinoma to the nasal cavity from testicular teratoma presenting with intractable epistaxis in a 32-year-old Caucasian male, who ultimately succumbed to this disease.

Key words: Nasopharyngeal neoplasms; Teratoma; Testicular neoplasms; Epistaxis; Neoplasm metastasis

Introduction

Malignant tumours of the nose and paranasal sinuses make up about 0.5 per cent of all tumours. The maxillary sinuses are most often involved, accounting for approximately 77 per cent of malignant sinus tumours (Sisson *et al.*, 1989). Of tumours metastatic to the paranasal sinuses, spread from renal cell carcinoma account for almost 50 per cent (Cinberg *et al.*, 1980). These metastatic tumours tend to be highly vascular and epistaxis is the most common presenting symptom. However, epistaxis is a less common presentation with other tumours metastasizing to the paranasal sinuses such as the ones arising from breast, lungs, testes and gastrointestinal tract.

Case report

A 32-year-old man was referred to the ENT Department with a history of intermittent epistaxis for the previous 10 days, worsening in severity and frequency. Initial examination revealed blood clots in both nostrils and active bleeding from the left nasal cavity. No postnasal bleeding was apparent. An anterior nasal pack was applied and the patient was admitted for bed rest and monitoring. Initial investigations including full blood count, differential count and clotting profile were normal. The epistaxis continued intermittently despite repeated packing, and so a decision was taken to examine the nose under general anaesthesia. This revealed an irregular fungating mass replacing the left middle turbinate. The lesion was biopsied and covered with calcium alginate haemostatic dressing, and postnasal and anterior packs were inserted.

Histopathology confirmed metastatic choriocarcinoma (Figure 1). Special stains showed the tumour cells were PAS positive but not diastase resistant. Immunohistochemistry was also consistent with a choriocarcinoma. The patient then admitted to the rapid appearance of an enlarging painless right testicular mass over the preceding three months. He also identified an erythematous skin lesion on the chest wall which had appeared over the same period. Intermittent diplopia was also reported. A repeat clinical examination revealed a firm right-sided testicular mass and an erythematous raised skin nodule measuring 4 mm in diameter in the right pectoral region. Computerized tomography (CT) showed a soft tissue mass in the left nasal cavity, displacing the septum to the right. Fluid was present in both maxillary antra. There was erosion of the medial wall of the left maxillary antrum, and the left middle turbinate was replaced by the mass (Figures 2 and 3). Enhancing lesions in the right parietal lobe and the cerebellum were demonstrated (Figure 4). Multiple pulmonary, mediastinal and liver metastases were also shown. His βhCG (beta-human chorionic gonadotropin) and αFP (alpha-fetoprotein) levels were 7.09×10^6 i.u./L and 15 K.U/L respectively (normal range –βhCG 0–4 i.u./L, αFP 0-8 K.U./L). He underwent a right orchidectomy and excision of the chest wall lesion. Histology confirmed a

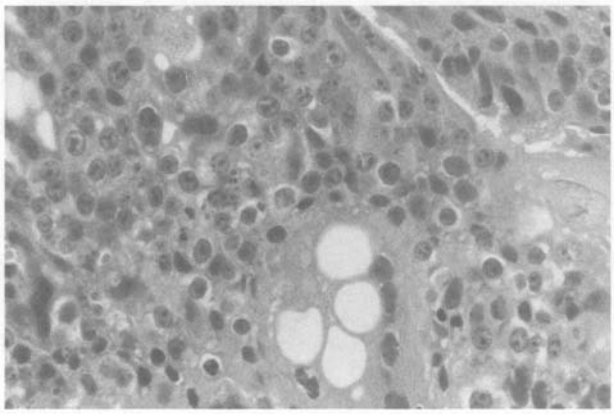

FIG. 1
Biopsy from left middle turbinate: Metastatic choriocarcinoma (H & E; × 40).

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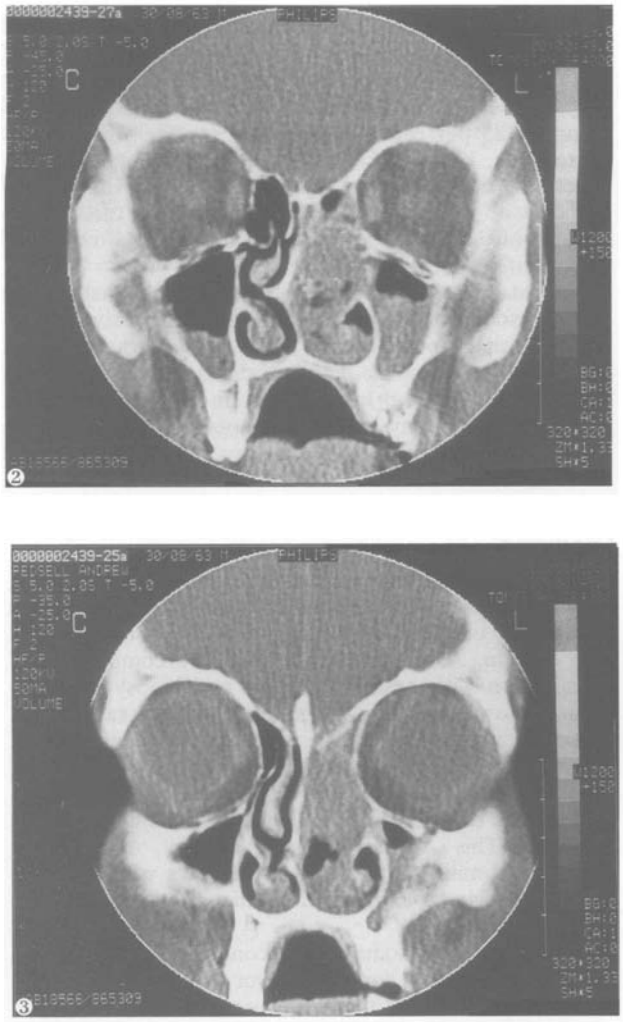

FIGS. 2 and 3

CT of the nose and paranasal sinuses: The left nasal cavity is expanded compared with the right and the nasal septum is deviated to the right. There is erosion of the medial wall of the left maxillary antrum and an air fluid level in both maxillary antra.

primary malignant testicular teratoma of intermediate type (Figure 5); with metastatic choriocarcinoma to the skin (Figure 6).

The patient was thus confirmed to have widely disseminated disease arising from malignant testicular teratoma of intermediate type (M.T.I.). He was appropriately staged and planned for urgent chemotherapy. Three days into treatment with the chemotherapy, he deteriorated with the rapid progression of coma and succumbed soon thereafter. It was suggested that he had haemorrhaged into the intracranial metastases following their response to chemotherapy.

Discussion

Primary malignant tumours of the nose and paranasal sinuses comprise three per cent of all malignancies of the upper respiratory and alimentary tracts and less than 0.5 per cent of all neoplasms (Sisson *et al.*, 1989). Sino-nasal malignancies in this area transgress natural anatomical boundaries and usually involve two or more anatomical sites by the time diagnosis is made. The maxillary sinus is most often involved, accounting for approximately 77 per

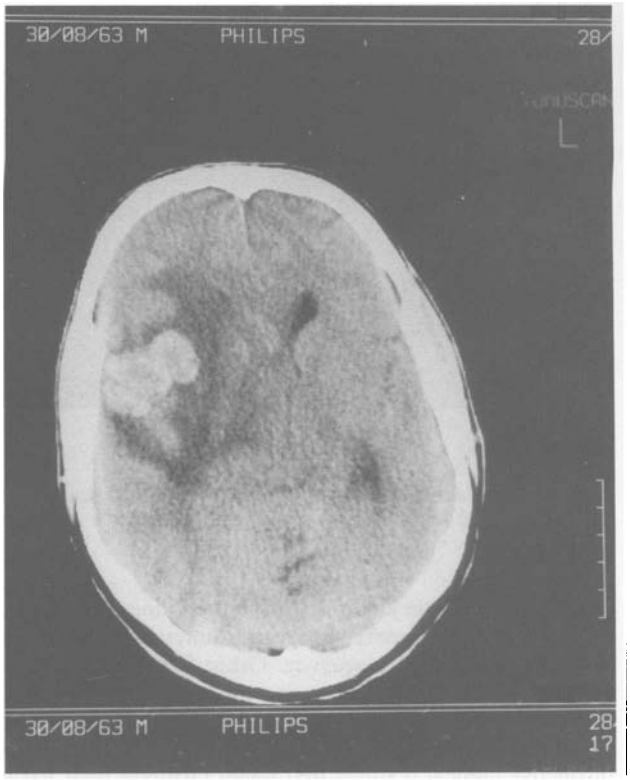

FIG. 4

CT brain: Enhancing lesions surrounded by oedema in right parietal lobe and both cerebellar hemispheres.

cent of all malignant sinus tumours. The ethmoid sinus is next most frequently involved, accounting for some 22 per cent. The sphenoid and frontal sinuses are involved in fewer than one per cent of the reported sinus malignancies (Renner *et al.*, 1984).

Squamous cell carcinoma is the most common malignancy involving the paranasal sinuses. This is usually as a primary neoplasm or as direct local extension from an adjacent primary site (Renner *et al.*, 1984). Adenocarcinoma, transitional cell carcinoma and adenoid cystic carcinoma are also found in the paranasal sinuses.

Metastatic tumours occur less frequently in the nose and paranasal sinuses. Fewer than 100 cases have been reported in the literature (Cinberg *et al.*, 1980). Primary

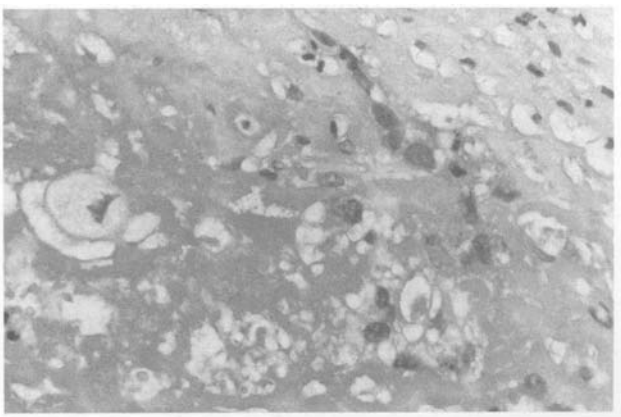

FIG. 5

Right testis: Malignant teratoma intermediate with areas of choriocarcinoma. There is vaso-invasion (H & E; × 40).

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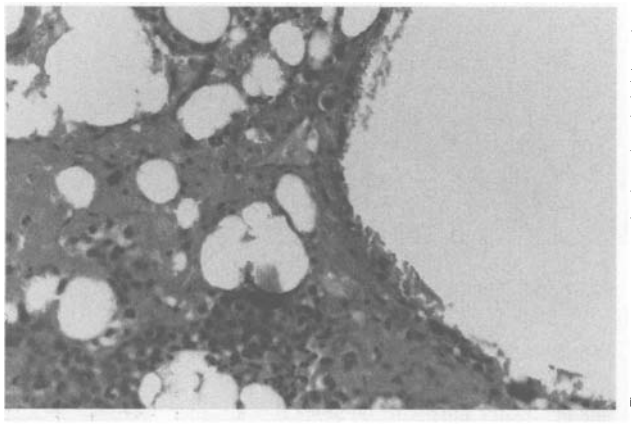

FIG. 6
Biopsy from skin: Metastatic choriocarcinoma similar in appearance to the biopsy from nose (H & E; × 40).

sites from which they may arise are the kidney, bronchus, gonads, female breast and gastrointestinal tract, in descending order of frequency. Other less common sites of origin include thyroid, pancreas, prostate, adrenals, liver, bone marrow and skin (Bernstein *et al.*, 1966).

Metastatic tumours to the paranasal sinuses have no distinctive clinical features that might facilitate their early diagnosis. The symptoms are identical to those caused by primary tumours in the same locations. These are nasal obstruction, epistaxis, pain, and facial asymmetry. Epistaxis is the presenting feature in 70 per cent of patients with nasal metastases secondary to renal cell carcinoma (Batsakis and McBurney, 1971; Renner *et al.*, 1984). Metastatic thyroid carcinoma presenting with severe recurrent epistaxis has also been reported (Barrs *et al.*, 1979; Cinberg and Terrife, 1980). Metastatic choriocarcinoma to the skin as the first indication of malignant disease in a young woman has also been documented (Cosnow and Fretzin, 1974).

Radiological investigation offers no special aid in as much as both primary and metastatic tumours can destroy bone and invade soft tissue. It is not until a biopsy is obtained that the true nature of the tumour may become evident. Even then the metastasis may not show any obvious microscopic features that would readily indicate its origin.

The pathogenesis of metastatic tumours has been discussed by Batson (1940). He emphasized the role of the vertebral venous plexus in tumour spread. This venous system consists of epidural and prevertebral veins with many intertwining vessels that communicate at every somite level with either the intercostal veins, the venae cavae, the azygous system or the pelvic veins. As these veins are without valves, increase of intrathoracic or intra-abdominal pressures could possibly drive the tumour cells into the vertebral venous plexus. Emboli could in this way reach the venous sinuses of the head potentially via the pterygoid plexus, the cavernous sinus and, by retrograde spread enter the paranasal sinuses. This would explain the propensity of renal, bronchial, breast and urogenital malignancies to metastasize to the paranasal sinuses. An alternative route of haematogenous spread has been discussed by Nahum and Bailey (1963). They suggested that spread occurs via the caval system, then through the pulmonary circulation, and thence from the heart to the sinuses through the arteries of the head and neck.

Whereas pure choriocarcinoma is the least common germ cell tumour, teratoma comprises 35 per cent of testicular tumours, while seminoma account for 40 per cent. Mixed germ cell tumours which behave like teratoma comprise 25 per cent of testicular tumours (Mostofi and Sesterhenn, 1994). Teratoma occurs at any age but more frequently during the first and second decades. In five to 10 per cent of patients with malignant testicular disease, the first manifestations are those resulting from metastatic deposits (Nespeca and Sass, 1980).

Metastatic spread is principally by the lymphatic route but choriocarcinoma is known for its aggressive blood-borne spread (Johnson, 1976). Metastases from teratoma spread to the paraortic lymph nodes and from thence to the mediastinum and lungs. As germ cells are totipotent elements, they may undergo trophoblastic or somatic differentiation either in the primary lesions or in the areas of metastases.

Conclusion

Intractable epistaxis is usually a condition involving hypertensive and elderly patients. However, when it occurs in a young otherwise healthy patient it must be viewed with caution. Assessment must include a complete ENT examination with nasal endoscopy and biopsy if appropriate, followed by thorough general medical examination. Particular attention should be given to the examination of the thyroid gland, breast, abdomen and testes. A chest X-ray, urine analysis and in appropriate instances, an intravenous urogram and computerized scanning may be required. This diagnostic process must exclude the presence of malignant disease in the sites which are the potential origin of malignancies which metastasize to the paranasal sinuses.

Treatment of teratoma usually consists of orchidectomy, followed by either surveillance or chemotherapy. The latter is indicated if there is vascular invasion in the primary tumour or there is metastatic disease confirmed by raised tumour markers (βhCG, αFP) in stage IM or by imaging in stage II–IV.

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