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Ameloblastoma presenting as nasal obstruction
Case reports

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Introduction

Ameloblastoma is described as a neoplasm of enamel organ-type tissue which does not undergo differentiation to the point of enamel formation (Shafer *et al.*, 1983). It is a rare tumour accounting for 1 to 2.3 per cent of all tumours and cysts of the jaws and originates in the mandible in 80 per cent of cases with only 20 per cent occurring in the maxilla (Small and Waldron 1955). Involvement of the nose by ameloblastoma at initial presentation appears to be recorded in only three previous single case reports, although there are additional reports of nasal involvement from recurrent maxillary ameloblastoma. We present two cases in which the presenting complaint was due to nasal involvement. We discuss the pathology and treatment, and review the relevant literature.

Case 1

A 76-year-old lady presented in June 1985 with a 10-month history of left nasal obstruction, purulent blood-stained catarrh and intermittent swelling of her left maxilla. She had been treated for acute sinusitis previously and recently had had a left upper molar extracted as it had become loose. Examination revealed a complete posterior left-sided nasal obstruction. There was fullness of both the infraorbital and gingivobuccal sulci on the left. A defect had persisted in the alveolus at the site of her dental extraction which was filled with a pink fleshy tumour. Sensation over the face and alveolus was normal. X-rays of the sinus revealed opacification of the left frontal and maxillary sinuses associated with destruction of both the lateral and medial walls of the maxillary antrum. There was also increased soft-tissue opacification of the nasal cavity. Computerised tomogram scanning confirmed a large soft tissue density lesion expanding into the left maxillary antrum,with destruction of the lateral postero-lateral and anterior bony walls of the antrum (Fig. 1). A trans-nasal biopsy was performed. At this time the lateral nasal wall was found to be deficient and the entire left nasal passage was filled from the inferior turbinate upwards with polypoidal tissue. The infero-lateral aspect of the maxillary antrum was also deficient at the point of continuity with the gingivobuccal swelling; however, the orbital floor appeared to be intact. A good clearance of the polypoid tissue was obtained from both the maxillary sinus and the nasal cavity. This caused the release of pus that had accumulated superiorly. The macroscopic appearance was of a pale polypoid swelling with vascular foci, more solid than an inflammatory polyp. Histology revealed the typical appearance of an ameloblastoma with a mixture of follicular and plexiform patterns (Fig 2). Islands of columnar cells with small oval nuclei and a paucity of mitoses grew in sheets and cords on a richly vascular framework. In areas the polarisation of nuclei towards the stellate reticulum was well demonstrated (Fig 3). There was some squamous differentiation but keratin formation, cellular degeneration and cholesterol cleft formation (as one would expect in a craniopharyngioma) were absent. There was no evidence of bony invasion in the tissue submitted for histological examination. In view of the desire to avoid cosmetic deformity, a left radical antrostomy was performed. A standard left canine fossa approach was extended to include the tooth socket which was excised with a surrounding ellipse of mucosa. Full macroscopic clearance was obtained from the floor and medial wall of the antrum and part of the inferior region of the medial wall was removed to create a large antrostomy. The tumour separated easily from the surrounding structures and the area of the tooth socket was cleared with excision of the left posterior alveolus. A new upper denture was fitted and she remains well and free from recurrence 18 months later.

Case 2

A 63-year- old man presented to his GP in June 1985 with a large fleshy polyp obstructing the right nostril of one month's duration. He had been undergoing investigations to exclude an intracranial cause of long-standing headaches. A CT scan had been normal and the impression was that these were anxiety related. When he was seen at the ENT clinic the nasal polyp had an appearance suggestive of a melanoma. X-rays and PA tomograms of the sinuses showed increased opacity of the right antrum and nasal cavity with destruction of the medial wall of the antrum (Fig. 4). A biopsy was taken of the nasal part of this mass which appeared to arise from the lateral wall of the nose. Histologically the lesion proved to be an ameloblastoma. A CT scan displayed the lesion occupying the right maxillary antrum with destruction of the medial, posterior and lateral walls. Again a radical antrostomy approach was made with an extended canine fossa incision through which the thinned anterior wall was easily entered. Further removal of anterior and lateral walls produced adequate access to remove the tumour along with the medial wall of the antrum in continuity with the inferior turbinate, A transantral ethmoidectomy was performed and tumour clearance was obtained up to the level of the cribriform plate. It was noted that the floor and medial walls of the orbit were intact. The postero-lateral wall of the antrum was found to be breached and a biopsy was taken from the overlying buccal fat. This biopsy was free from tumour but the antral mass was confirmed histologically to be an ameloblastoma, with the same features as detailed for Case 1 but with a greater tendency to squamous differentiation. The bony

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FIG. 1

CT scan revealing soft tissue density mass filling the left maxillary antrum with destruction of the lateral, posterolateral and anterior walls of the antrum Case 1.

fragments were all free from tumour invasion on histological examination. Apart from a small oro-antral fistula which required resuturing, his recovery was satisfactory and he remains free from tumour although his initial headaches have persisted.

Discussion

Our two cases illustrate many of the salient points appropriate to the tumour which is slow-growing and locally destructive but rarely, if ever, metastasises. The presentation may follow local swelling or involvement of adjacent structures. In the maxilla the tumour tends to expand into the sinus and presents as a large tumour either with swelling of the cheek, gingiva or hard palate and may occasionally involve the orbit (Shaw and Katsakis, 1973). Recent dental extraction is often reported which is almost certainly a consequence of the tumour rather than of aetiological significance. These tumours have a long natural history and in the largest series, that of Small and Waldron (1955), the mean length of time the tumour was considered present before diagnosis was five years. Case 1 reported definite symptoms for at least 10 months before pres-



FIG. 2

Plexiform and follicular areas in an ameloblastoma. The epithelium shows central transition to a spindle morphology resembling the stellate reticulum Case 1.
Haematoxylin and eosin×50.

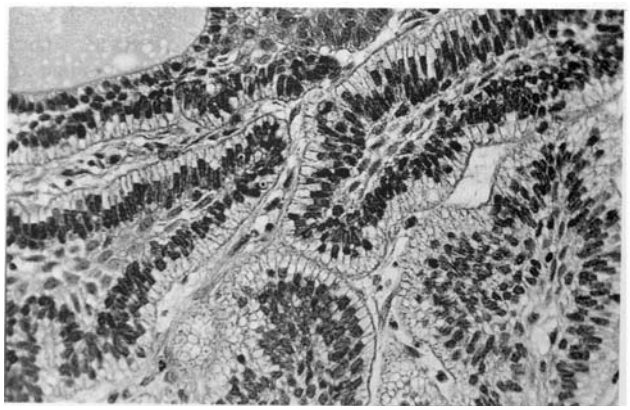


FIG. 3

The strands of epithelium show polarisation of the nuclei of columnar cells towards the stellate reticulum Case 1.
Haematoxylin and eosin×320.

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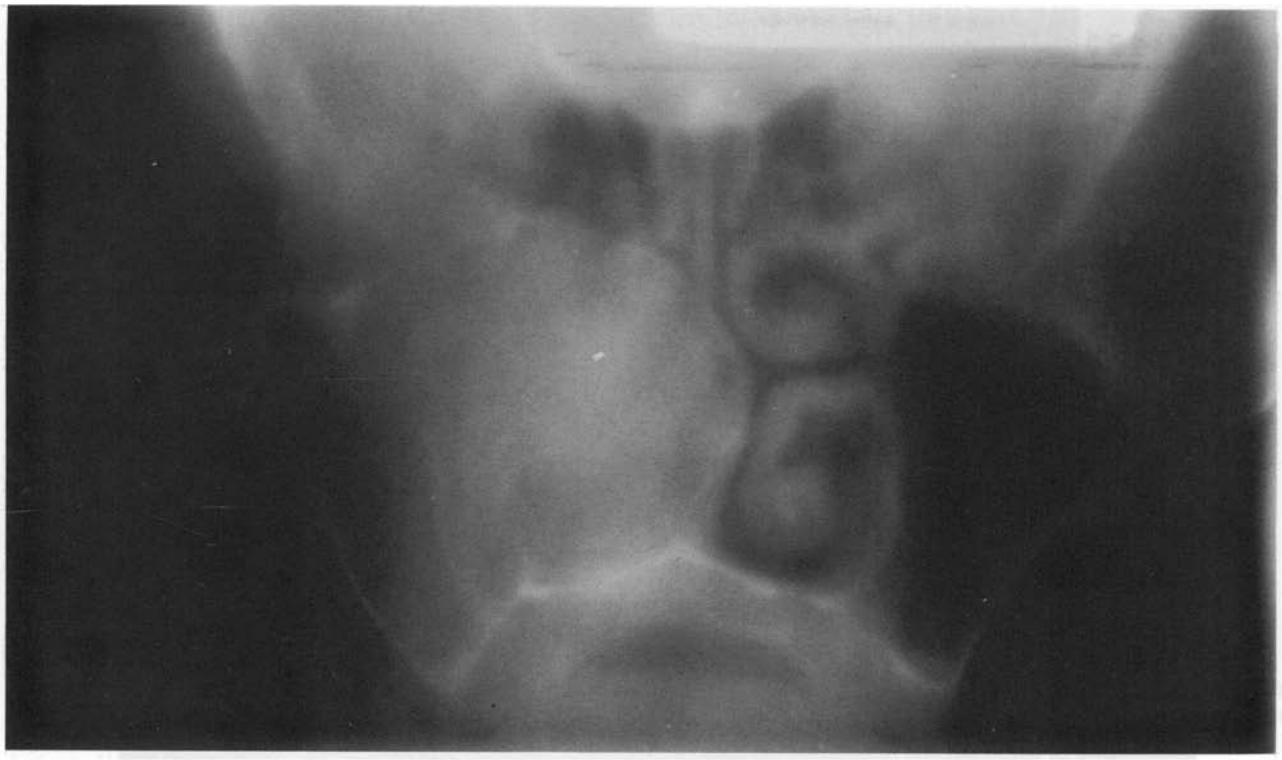


FIG. 4

Plain PA tomogram of Case 2 showing opacity of the right antrum with erosion of the medial wall and extension displacing the nasal septum.

entation and possibly symptoms had been present for much longer. In Case 2, the long history of headache may have been related but despite complete tumour removal this symptom persisted.

A review of the literature has not identified any previous cases presenting with nasal obstruction alone, as described in Case 2. Single case reports exist of this rare tumour presenting as a nasal mass but, as in Case 1, this has been in association with a bloody nasal discharge or frank epistaxis, which arouses the suspicion that the lesion is neoplastic rather than a simple inflammatory polyp (Chaudhuri, 1974; Weissman and Wetli, 1977; Wenig *et al.*, 1985). Further reports of maxillary ameloblastoma leading to nasal obstruction have been in cases with recurrence after failed primary treatment.

Since an ameloblastoma presenting as a nasal polyp is unusual, the pathologist encounters an unfamiliar cellular pattern; the more so since this particular lesion is diagnosed more frequently by oral pathologists and less often in a general diagnostic department. The highly cellular neoplasm,particularly one of a follicular variety, may mimic an adenocarcinoma, a lesion found (albeit rarely) with appreciable frequency within the nasal cavity (Sanchez-Casis *et al.*, 1971). Careful examination reveals that the apparent follicular lumina comprise in fact the oedematous but vascularised cores supporting the cellular cords. Thus there is no luminal secretion. Similarly mitoses are infrequent in comparison with adenocarcinoma. The typical localisation of nuclei at the apices of the cells and the maturation of columnar to stellate forms, confirms the diagnosis. The correct diagnosis is an important one, the ameloblastoma being an essentially benign lesion despite its cellularity and requiring less radical treatment than the rare but invasive carcinoma of the nasal cavity or accessory sinus.

A different problem of histological distinction relates to the resemblance of an ameloblastoma to a craniopharyngioma. The greater frequency of squamous differentiation, keratin release and cellular response thereto and cholesterol crystal deposition in the latter, aids in the correct interpretation. Similarly the age of the patient, 50 per cent of craniopharyngiomas present before puberty, and additional clinical information such as erosion of the ethmoid plate and continuity with a third ventricular lesion are supportive factors.

The preferred treatment of ameloblastoma was at one time conservative and curettage was advocated but recurrence rates as high as 100 per cent (Sedhev *et al.*, 1974) led to rejection of this approach. Radiotherapy has been tried but produced recurrence rates ranging from 42 per cent (Becker and Pertl, 1967) to 72 per cent (Pantzapoulos and Nomikos, 1966) and, therefore, more radical surgery is now advocated although the definition of what is considered radical excision has varied. With a policy of radical surgery, Becker and Pertl, 1967) claim a cure rate of 95 per cent.

Partial maxillectomy is now considered to be the treatment of choice for maxillary ameloblastoma (Sedhev *et al.*, 1974; Tsaknis and Nelsen, 1980), the latter series achieving 9 - 18 year disease-free survival in all five patients.

For the occasional patient who is not a suitable candidate for surgery, temporary resolution has been reported following radiotherapy despite the poor long term results (Hair, 1963; Shaw and Katsakis, 1973).

In Case 1 the excision of the involved tooth root and adjacent alveolus along with excision of parts of the medial and lateral walls of the antrum was felt to be adequate. In the second case, the tumour appeared to originate around the middle meatus and to extend into the nasal cavity and maxillary antrum. Radiologically the palate was not involved. The nasal wall of the antrum was excised as well as parts of the postero-lateral wall and alveolus. In neither case was there subsequent histological evidence of bone involvement. We felt that an adequate and complete excision had been achieved in both cases and intend to follow them up indefinitely with the option of a formal

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maxillectomy should there be recurrence. So far there has been no evidence of recurrence in either case.

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