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Ossifying pleomorphic adenoma of the maxillary antrum

K. C. Lee, M.B., B.S.,* J. K. C. Chan, M.R.C.Path.,* Y. W. Chong, M. B., B.S.† (Hong Kong)

Abstract
An unusual case of pleomorphic adenoma with exuberant bone formation, occurring in the maxillary antrum of a 21-year-old male and showing repeated recurrence, is reported. In contrast to the endochondral ossification in the previous reported cases of pleomorphic adenoma, direct deposition of osteoid by metaplastic myoepithelial cells is suggested to explain the bone formation. The difficulties of distinguishing this tumour from osteosarcoma during intraoperative diagnosis are discussed.

Introduction

Pleomorphic adenoma of the upper respiratory tract occurs almost exclusively in the nasal cavity, and very few cases have been reported to arise in the paranasal sinuses (McDonald and Havens, 1948; Russell, 1955; Rafla, 1969; Spiro *et al.*, 1973; Compagno and Wong, 1977; Hyams *et al.*, 1988). We report a unique case occurring in the maxillary sinus and showing extensive bone formation. The latter feature is very uncommon in pleomorphic adenomas, and posed problems in intraoperative diagnosis because it mimicked an osteosarcoma.

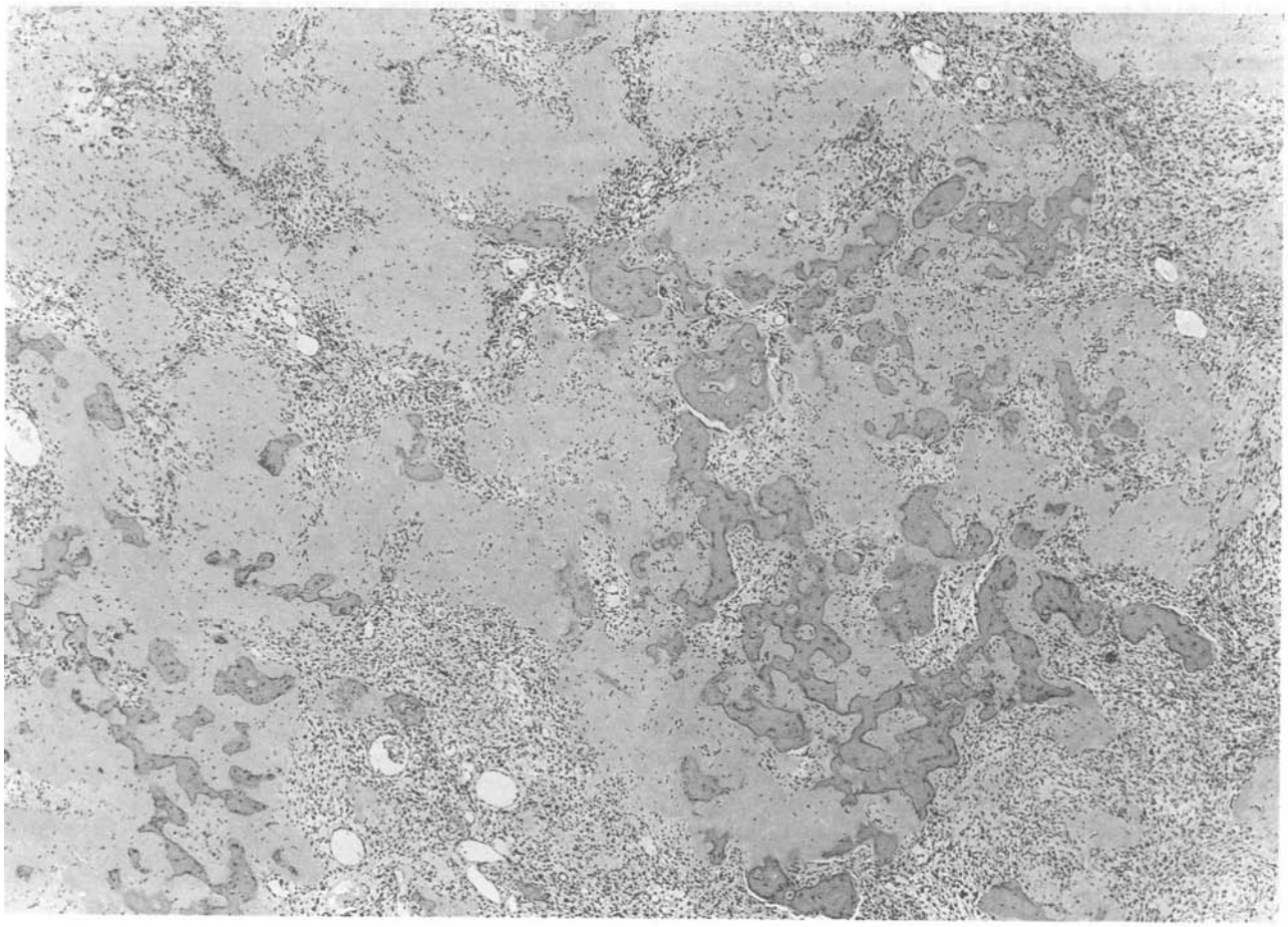

Fig. 1
Low power view showing extensive bone and osteoid deposition. Note the high cellularity in the background, mimicking osteosarcoma. H&E ×150.

From: Institute of Pathology, Queen Elizabeth Hospital* and United Christian Hospital,† Hong Kong.
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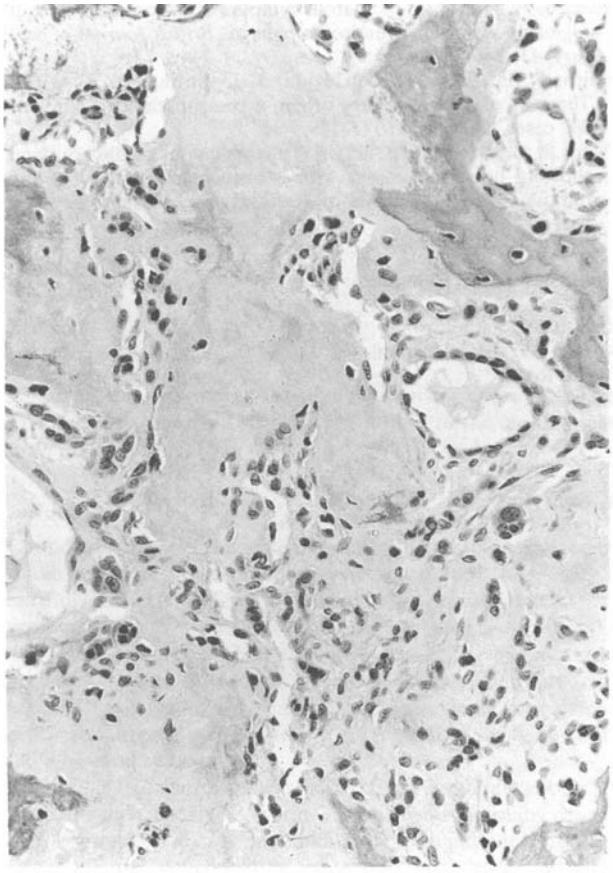

FIG. 2

Higher magnification showing close association of myoepithelial-like cells with the osteoid. Note a small glandular structure in the right field providing clue to the diagnosis of pleomorphic adenoma. H&E ×250.

Case report

A 21-year-old Chinese male first presented in 1979 with a tumour in the left maxillary sinus. Maxillectomy was performed, and the histological diagnosis was a pleomorphic adenoma. Unfortunately, details of the operative findings and comments on resection margin were not available. The lesion recurred in 1988 in the same site with involvement of the infratemporal region, ethmoid and sphenoid sinuses, up to the dural base and nasal fossa. The tumour had a well defined plane except in the left orbit where it appeared to infiltrate the orbital fat and rectus muscle; 44 g of tumour were removed by piecemeal resection and blunt dissection. The lesion recurred again in 1989 with bulky local disease, and 75 g of tumour tissue were removed in pieces. The patient has remained recurrence-free after the third operation.

Pathological findings

The tumour exhibited the same histological appearances in the original resection and in the recurrences, and maintaining the pushing borders. The original tumour was well circumscribed and spherical. It was covered by an intact repiratory mucosa. It consisted of small glandular structures lined by epithelium of two-cell type, that is, an inner layer of ductal epithelium and an outer layer of myoepithelium. The outer layer merged into a fibrocellular stroma which contained abundant anastomosing osteoid and bony trabeculae (Fig. 1). The bone was present both in the peripheral portion and the centre of the tumour, and was therefore unlikely to represent merely host reaction to the tumour. In between the bony trabeculae, isolated myoepithelial-like cells were present, often closely apposed to their surfaces (Fig. 2). There was no intervening cartilage. Immunostaining confirmed the biphasic cellular population, with carcinoembryonic antigen-positivity in the luminal surface of the glands, and actin-positivity in the myoepithelial elements. Cytokeratin staining was demonstrable in both elements (Fig. 3).

Discussion

Pleomorphic adenoma is well known for its diverse histological patterns and the frequent presence of chondromyxoid matrix. However, bone formation is very rare and is inconspicuous if present. In the previously reported cases, the bone was formed within areas of metaplastic cartilage, that is, via the process of endochondral ossification (Yates and Paget, 1952). The present case is distinctive in that ossification is so exuberant that it dominates the histological picture in many parts of the tumour. Furthermore, the bone is consistently present even in the recurrences providing further support that it is an integral component of the pleomorphic adenoma. The adenomatous nature of the tumour is not readily appreciated in many foci. The bony matrix in this case appears to be deposited directly by the metaplastic myoepithelial cells rather than through endochondral ossification in contrast to the reported cases. This ossifying pleomorphic adenoma is further unusual in taking origin in a paranasal sinus, a site rarely reported to be involved by this tumour. Since the tumour maintained pushing growth fronts and did not exhibit cellular pleomorphism, we do not consider the possibility of malignant transformation in the pleomorphic adenoma to be likely.

One of the recurrent specimens was received for intraoperative diagnosis. The close association of the osteoid and bone with a highly cellular background raised the concern for osteo-

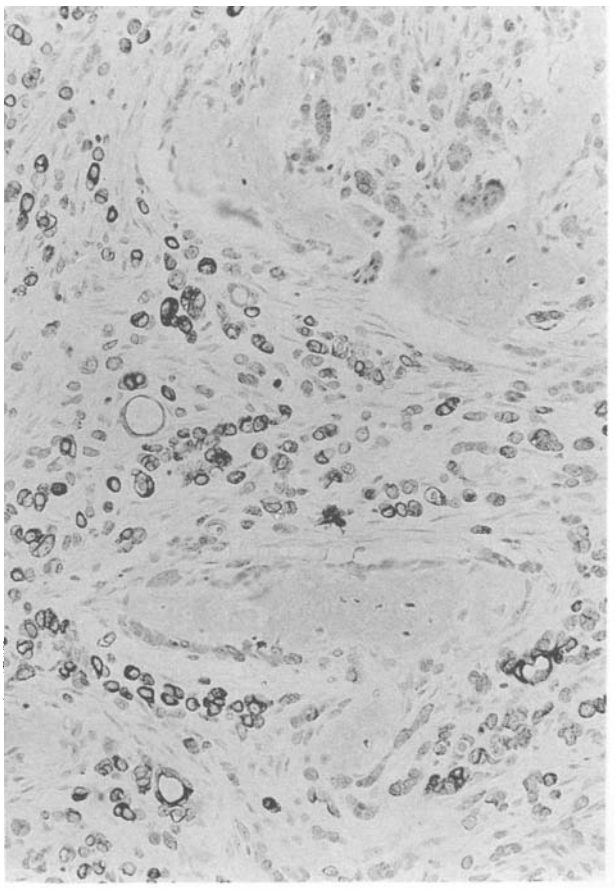

FIG. 3

Numerous cytokeratin-positive cells are found between the bony trabeculae. Peroxidase-anti-peroxidase method for cytokeratin. ×250.

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sarcoma. The recognition of focal ductal structures and the overall circumscription of the tumour helped to clarify the diagnosis of pleomorphic adenoma with exuberant ossification.

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Address for correspondence:
Dr. K. C. Lee,
Institute of Pathology,
Queen Elizabeth Hospital,
Wylie Road,
Kowloon,
Hong Kong.

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