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Polymorphous low grade adenocarcinoma of the palate in a child

Y. W. Tsang M.B.,B.S., * Y. Tung F.R.C.R.,† J. K. C. Chan M.R.C.Path., F.C.A.P.* (Kowloon, Hong Kong)

Abstract
We report a case of polymorphous low grade adenocarcinoma involving the palate of a 12-year-old girl, the first example of this tumour occurring in the paediatric age group. The tumour displayed infiltrative growth, neural invasion, variegated histological patterns, and minimal cytological atypia. The patient remained disease-free four years after wide local excision of the tumour. The distinction of polymorphous low grade adenocarcinoma from pleomorphic adenoma and adenoid cystic carcinoma is also discussed.

Introduction

Salivary gland tumours are uncommon in the paediatric age group, with pleomorphic adenoma of the parotid gland accounting for half of these tumours (Baker and Malone, 1985; Dehner, 1987). Among the malignant salivary gland tumours in children, mucoepidermoid carcinoma is the commonest and most occur in the major glands.

We report the occurrence of a polymorphous low grade ade-

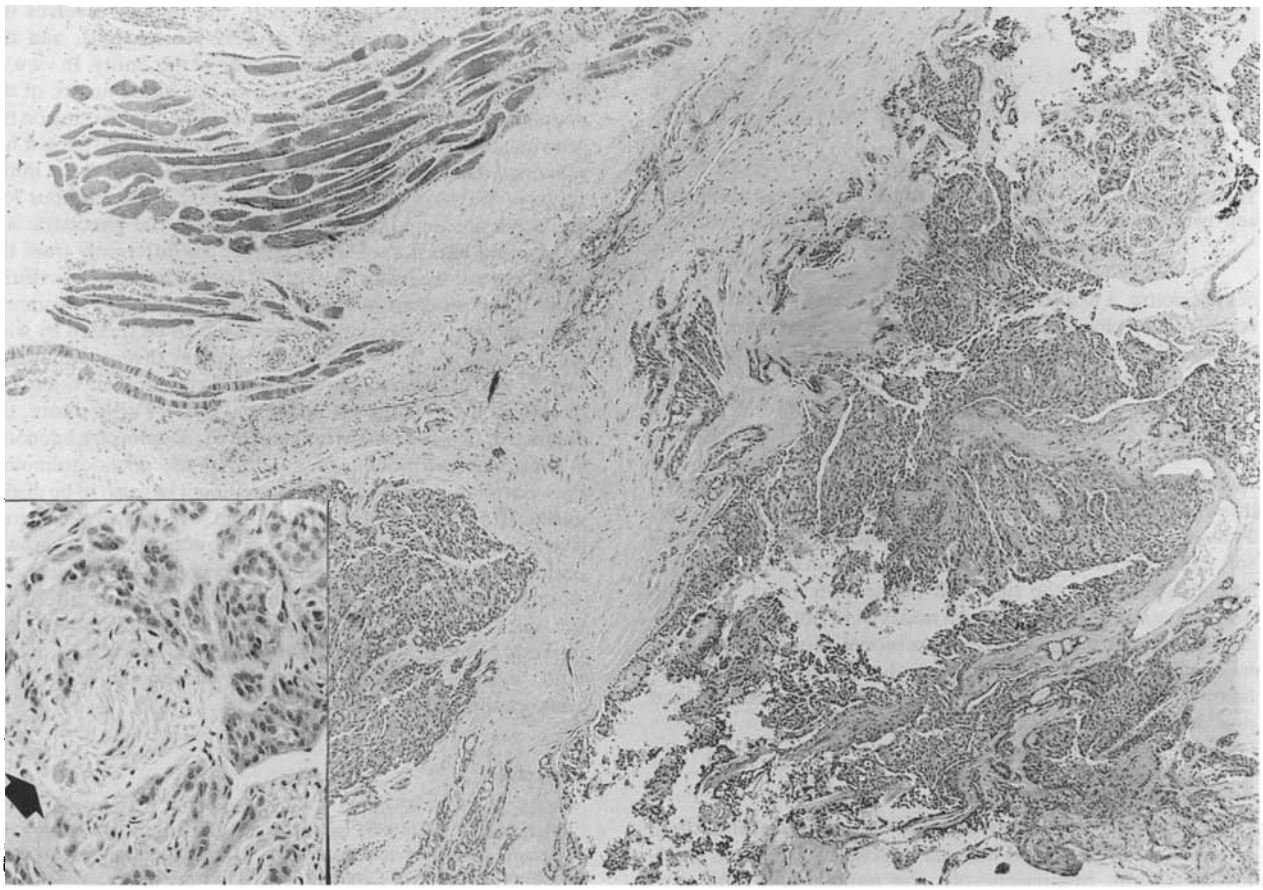

Fig. 1

The tumour infiltrates the skeletal muscle of the palate; it shows a variegated growth pattern. The tumour infiltrates around and into the nerve (arrow), as shown in the inset. H&E, ×50; inset, ×125.

*Institute of Pathology, and †Institute of Radiology and Oncology, Queen Elizabeth Hospital, Hong, Kong.
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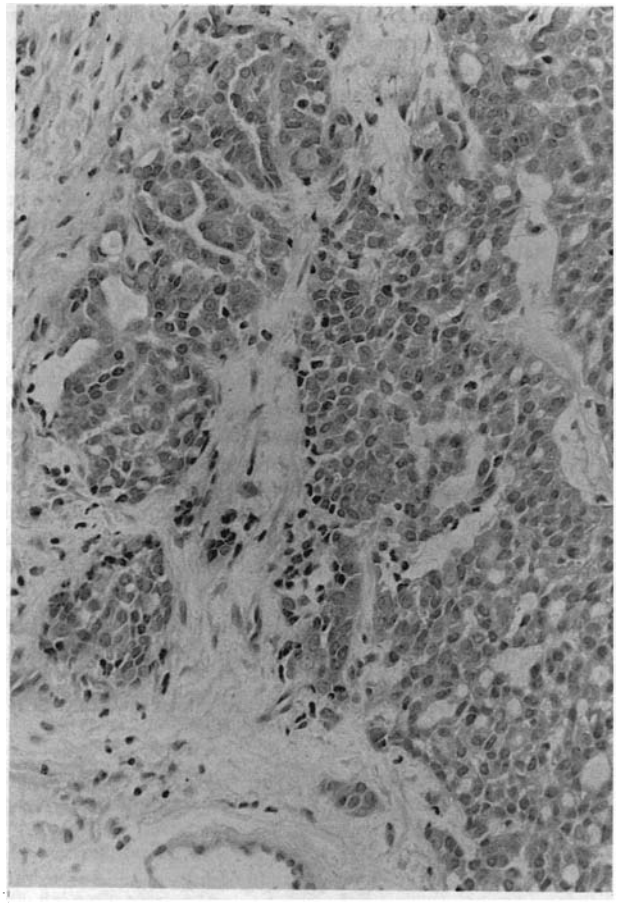

FIG. 2

Tumour cells possess bland nuclei with finely stippled chromatin and inconspicuous nucleoli. They form tubules and cribriform structures in this field. H&E, ×190.

nocarcinoma, a distinctive indolent carcinoma which has only recently been characterized (Evans and Batsakis, 1984; Aberle *et al.*, 1985; Anderson *et al.*, 1990) and which has hitherto been reported only in adults.

Case report

A 12-year-old girl who had open heart surgery at the age of three years for Fallot's tetralogy presented in March 1986 with a slowly progressive painless swelling at the right side of the soft palate for two years. Physical examination revealed a 2 cm smooth swelling at the right soft palate with intact overlying oral mucosa. The cervical lymph nodes were not palpable. Frozen section during subsequent excisional biopsy showed an infiltrative adenocarcinoma; wide local excision was performed. No adjuvant therapy was given and she has remained disease-free four years after surgery.

Pathological findings

The excision specimen consisted of a non-circumscribed mass with intact overlying mucosa, measuring 2 cm in its greatest dimension. Microscopically, the tumour had infiltrative borders, invading both the skeletal muscle of the palate and around and inside the nerves (Fig. 1). The growth pattern was highly variable, with solid, trabecular, cribriform, papillary and complex tubular structures. The stroma in between was sclerotic. The tumour cells were uniform, with oval nuclei, finely stippled chromatin and inconspicuous nucleoli. They were cuboidal, polygonal or columnar, and the cytoplasm was clear to eosinophilic (Fig. 2). In most areas, only a single population of cells was identified; in small foci, a minor population of myoepithelial cells was present beneath the glandular lining. Mitotic figures were not seen. The resection margins were not involved. The features were those of a polymorphous low grade adenocarcinoma of minor salivary gland origin.

Discussion

Polymorphous low grade adenocarcinoma is a distinct indolent neoplasm occurring mostly in minor salivary glands of the palate and oral cavity, and occasionally in other sites such as the nasopharynx (Wenig *et al.*, 1989). In major salivary glands, it occurs almost exclusively in the setting of carcinoma ex pleomorphic adenoma (Luna *et al.*, 1987). It has variously been termed 'lobular carcinoma', 'terminal duct carcinoma' and 'low grade papillary carcinoma' (Batsakis *et al.*, 1983; Freedman and Lumerman, 1983; Evans and Batsakis, 1984; Mills *et al.*, 1984). It is characterized histologically by diversity of pattern, cytologic blandness and an infiltrative growth pattern (Evans and Batsakis, 1984; Ellis and Gnepp, 1988). It is a low grade malignant tumour, as evidenced by the small but definite risk of local recurrence and metastasis to regional lymph nodes; distant metastasis has not been documented (Evans and Batsakis, 1984; Aberle *et al.*, 1985; Ellis and Gnepp, 1988).

In previous studies on polymorphous low grade adenocarcinoma, the patients' ages ranged from 23 to 79 years with peak incidence in the fifth and sixth decades (Luna *et al.*, 1987; Ellis and Gnepp, 1988; Anderson *et al.*, 1990). It has not been reported to occur in the paediatric age group in the English-language literature. In this report, the tumour involves the typical site (palatal mucosa) of a 12-year-old girl, and the microscopic findings are characteristic of this entity. In view of the completeness of excision, known low grade nature of the neoplasm and potential side-effects of radiation therapy on the growing bone, no adjuvant therapy was given. The patient has remained disease-free on follow-up four years after the initial surgery. We have thus documented that polymorphous low grade adenocarcinoma can also occur in the paediatric age group, and that it appears to behave no differently from the same tumour occurring in adults. It is most important to distinguish this tumour from a pleomorphic adenoma or monomorphic adenoma, because this tumour has to be treated by wide local excision with clear margins. The key distinguishing feature is the infiltrative growth, with perineural invasion frequently being identified (Luna *et al.*, 1987). Furthermore, the distinctive two-cell type arrangement of pleomorphic adenoma is uncommon in polymorphous low grade adenocarcinoma. Polymorphous low grade adenocarcinoma has also to be distinguished from adenoid cystic carcinoma, which is a more aggressive tumour. Though both tumours show an infiltrative growth pattern and neurotropism, polymorphous low grade adenocarcinoma is composed of cells with pale nuclei which differ from the dark basaloid cells of adenoid cystic carcinoma; the biphasic cytological pattern is also more a characteristic of adenoid cystic carcinoma than polymorphous low grade adenocarcinoma.

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Address for correspondence:
Dr Y. W. Tsang,
Institute of Pathology,
Queen Elizabeth Hospital,
Wylie Road,
Kowloon,
Hong Kong.

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