

Hodgkin's disease secondary to recurrent adenoid cystic carcinoma of the parotid gland

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Abstract

A case of Hodgkin's disease presenting as a second primary malignancy in a patient admitted eight years previously for total parotidectomy for an adenoid cystic carcinoma is reported.

Introduction

The occurrence of metachronous malignant neoplasms is a relatively frequent event, (Biggar *et al.*, 1983; Gluckman and Crissman, 1983; Licciardello *et al.*, 1989). Such tumours can develop independently, in the same organ affected by the primary neoplasia, in different areas of the same organ or in different organs. Although they often present the same histological features as the primary tumour, they can be of different histological type.

Metachronous malignant tumours appear to be more frequent in individuals who smoke, drink (Dana Flanders and Rothman, 1982; De Stefani, *et al.*, 1987; Merletti *et al.*, 1989) or received radiotherapy or chemotherapy for the primary tumour (Lawson and Som, 1975; Jenkins *et al.*, 1976; Penn, 1976; Cooper *et al.*, 1989). Hormonal, immunological, viral and diatthetic factors have also been implicated as predisposing agents (Dellon *et al.*, 1975; Katz, 1983; Schantz *et al.*, 1987).

Statistical data confirm that males are at higher risk than



FIG. 1

Picture showing the operative field after total parotidectomy with facial nerve preservation.

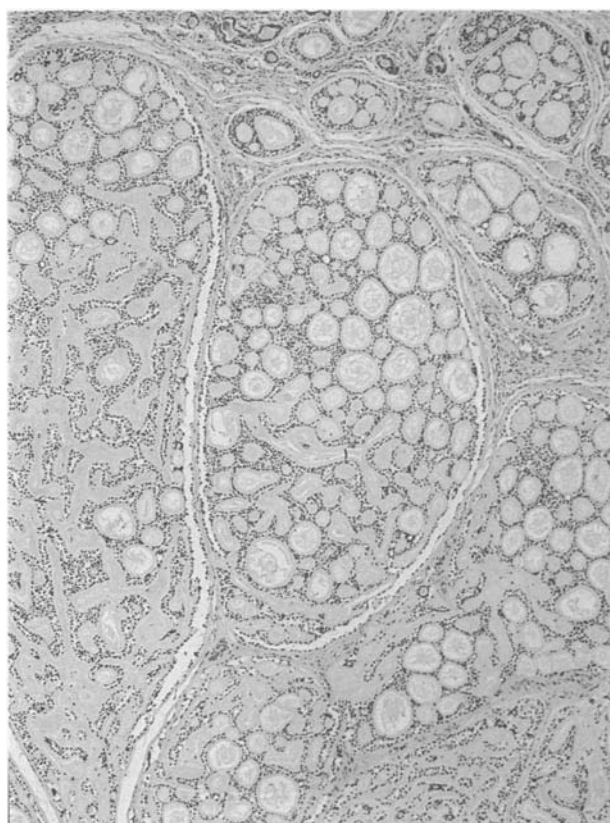


FIG. 2

Adenoid cystic carcinoma of the parotid gland, cribriform pattern. H&E. ($\times 36$).

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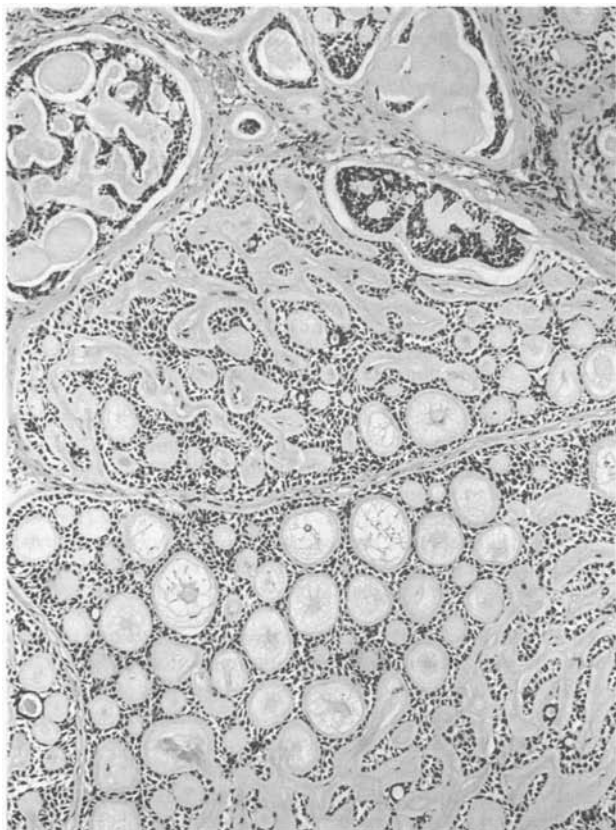


FIG. 3

Recurrence of the adenoid cystic carcinoma. The cylindrical spaces are occupied by mucoid material. H&E. ($\times 75$).

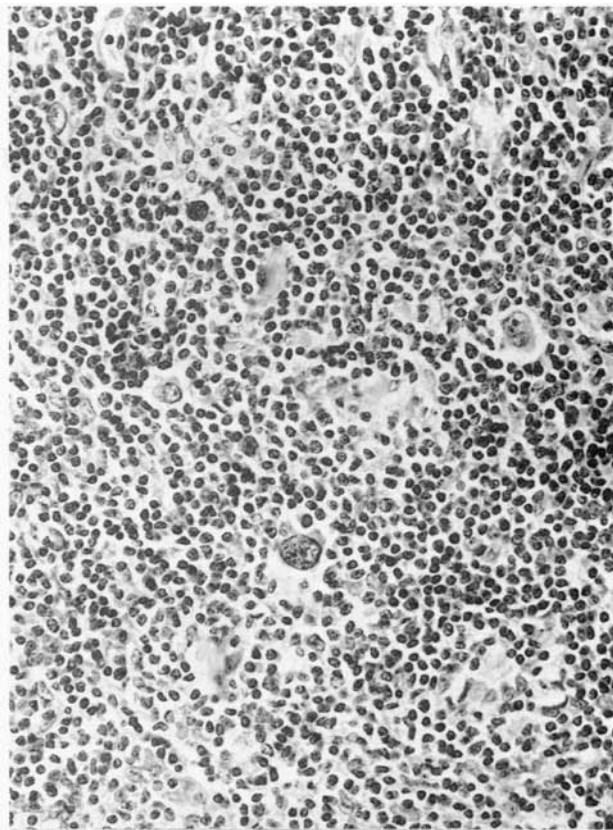


FIG. 4

Lymphocyte-predominant Hodgkin's disease. A monomorphic growth pattern is present with some Reed-Sternberg cells. H&E, ($\times 240$).

females (Schindel and Castoriano, 1972), that the incidence of these tumours increases with age (Spratt and Hoag, 1966), and that their number can vary, as cases have been reported with more than one neoplastic localization (Dellon *et al.*, 1975; Megighian and Zorat, 1985; Friedmann and Ferlito, 1988). As mentioned above, some tumours are of different histological type from the primary tumour. Since this is a fairly uncommon event, we thought it interesting to report a case that we have observed.

Case report

A 42-year-old male was admitted to the Otolaryngology Clinic, in January 1977, for a tumour of the left parotid gland and subsequently submitted to total parotidectomy with preservation of the facial nerve (Fig. 1). Histology showed an adenoid cystic carcinoma of the parotid gland.

Eight years later, during a follow-up examination, a small mobile, well-circumscribed mass, measuring 2–3 cm in diameter was found. It was located on the lateral surface of the sternocleidomastoid muscle, facing the parotid region on the operated side, which appeared to be normal.

The tumour was excised and a histological diagnosis of a recurrence of the adenoid cystic carcinoma was made presenting morphological features similar to those observed previously (Fig. 2). Microscopy shows nests, cords and solid sheets of epithelial cells, often interspersed with glandular and cyst-like spaces containing PAS-positive material. The fibrous stroma shows zones of hyalinization (Fig. 3). A lymph node present in the biopsy was normal.

About two years later, in February 1987, the patient was readmitted to the clinic for the presence of enlarged right lateral cervical and supraclavicular lymph nodes. Microscopic examination of an excised lymph node revealed lymphocyte-

predominant Hodgkin's disease. The underlying structure of the node was obliterated by the presence of a diffuse, occasionally nodular infiltration of lymphocytes. Scattered throughout were a few eosinophils and plasma cells and some large cells with a prominent nucleolus resembling a mono- or bi-nucleated Sternberg cells (Fig. 4). Some thin strands of fibrous connective tissue were present.

The patient underwent radiotherapy with a total dosage of 400 cGy (Mantlefield) for HD PL in Stage Ia. Chest X-rays, abdominal lymphography, macro- and microscopic laparoscopy, abdominal CT scans, bone biopsy, needle biopsy of bone marrow and immunoelectrophoresis were all negative. At the follow-up in September 1989, the patient's condition appeared to be satisfactory. At the time of proof correction, the patient's condition at the last follow-up (January 1991) showed that he was still satisfactory.

Discussion

This case is rather unusual due to the long period of time which elapsed between the appearance of the parotid gland tumour and the development of the metachronous lymph node neoplasia, the different histological type and the difficulty in evaluating possible aetiopathogenetic factors associated with the subsequent primary neoplasia. In fact, Hodgkin's disease appeared ten years after the total parotidectomy for adenoid cystic carcinoma.

Furthermore, Hodgkin's disease is rarely observed, not only as secondary neoplasia (Bandini *et al.*, 1981), but particularly as metachronous neoplasia to malignant salivary gland tumours (Prior and Waterhouse, 1977; Biggar *et al.*, 1983).

The present case has offered us clues regarding the aetiological problem of metachronous malignant tumours. The patient had no past history of radiotherapy or chemotherapy, he did not smoke or drink and there was no evidence of a cancer diathesis. There remains only one important element to consider:

the appearance of Hodgkin's disease about a year after the recurrence of adenoid cystic carcinoma. This may be a fortuitous coincidence.

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