

Bilateral basal-cell adenomas in the parotid glands

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Abstract

A rare case of bilateral basal-cell adenomas in the parotid glands of a 65-year-old woman is reported. There have been only four previous reported cases of bilateral parotid basal-cell adenomas, all of which occurred synchronously with dermal cylindromas. The present lesion is the first case reported in the English literature of bilateral parotid basal-cell adenoma without coexisting dermal cylindroma. Histological examination revealed that the left tumour had adenoid cystic change, which closely resembled adenoid cystic carcinoma. This case is of interest not only because of the rarity of bilateral parotid basal-cell adenomas, but also because of the necessity for differentiation from adenoid cystic carcinoma in order to plan appropriate treatment.

Key words: Parotid neoplasms; Adenoma; Carcinoma, adenoid cystic

Introduction

The majority of bilateral parotid tumours are Warthin's tumours.^{1,2} The occurrence of other benign tumours in the bilateral parotid glands is extremely rare. Only four cases of bilateral basal-cell adenoma of the parotid glands have been reported,^{3–6} all of which occurred synchronously with dermal cylindromas. The present lesion is the first case reported in the English literature of bilateral parotid basal-cell adenoma without coexisting dermal cylindroma.

Basal cell adenoma occasionally shows adenoid cystic change.⁷ Because basal cell adenoma has various histological variants the histopathological diagnosis should be made very carefully. Adenoid cystic changes in particular need to be distinguished from adenoid cystic carcinoma.

Case report

A 65-year-old woman was referred for evaluation of bilateral parotid masses, which had been pointed out during a health screening a month previously. The masses were able to be mobilized, were elastically hard and poorly defined. Ultrasonography revealed that the left mass was echographically homogeneous, measuring 27 × 24 mm in diameter, whereas the right lesion showed cystic and solid patterns, measuring 28 × 26 mm in diameter. Magnetic resonance imaging (MRI) revealed bilateral parotid masses in the deep lobes (Figure 1). Because of the pre-operative cytologic diagnosis of benign tumours, as well as the location of the lesions in the deep lobes, we planned a staged operation on each side.

The left tumour was located in a deep lobe of the parotid gland and total parotidectomy with facial nerve preservation was performed. Histopathological examination revealed that monomorphic adenoma was complicated by adenoid cystic carcinoma. There was a question of whether to perform surgery on the right parotid gland or postoperative irradiation on the left one. As the left lesion

was completely removed and no extracapsular invasion was found histologically, it was decided to operate on the right lesion prior to irradiation on the left side.

Like the left lesion, the right tumour originated from a deep lobe and was excised together with that lobe. Histological examination revealed basal-cell adenoma with tubular variants (Figure 2). As the histopathological diagnoses of the two excised tumours were different, a detailed histological re-examination of the left lesion was then carried out, revealing the following three findings: i) the tumour nests that had been diagnosed as adenoid cystic carcinoma showed histological transition into adenoma cells, ii) no capsular invasion was found, and iii) there were few atypical changes in the tumour cells. Based on these findings, the final histopathological diagnosis of the left tumour was confirmed as basal-cell adenoma with an adenoid cystic change (Figure 3). Post-operative therapies



FIG. 1

Magnetic resonance (MR) imaging revealed bilateral parotid masses in the deep lobes.

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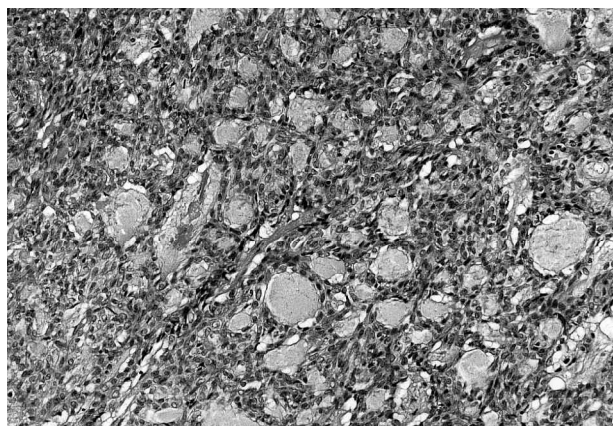


FIG. 2

Basal-cell adenoma, tubular type, of the right parotid gland: characteristic small lumens lined by ductal cells are observed (H&E; $\times 50$).

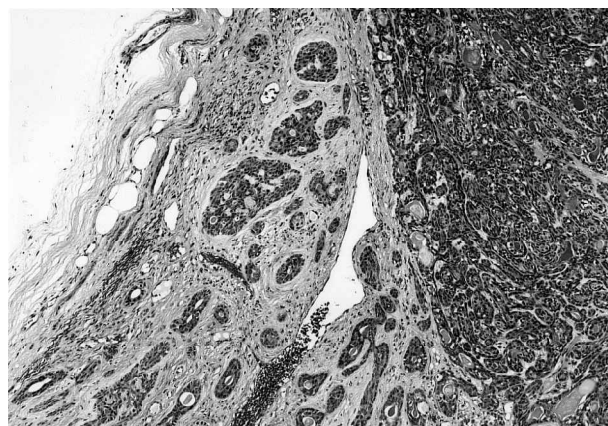


FIG. 3

Basal-cell adenoma with an adenoid cystic pattern of the left parotid gland. This histologic pattern may be confused with adenoid cystic carcinoma but capsular infiltration is absent (H&E; $\times 25$).

TABLE I
REPORTED CASES OF BILATERAL PAROTID BASAL CELL ADENOMAS IN THE ENGLISH LITERATURE

Report	Patient	Occurrence of parotid lesions	Coexisting dermal cylindromas	Family history of dermal tumours
Reingold <i>et al.</i> ³	43 / M	metachronous	Yes	Yes
Herbst and Utz ⁴	54 / F	metachronous	Yes	Yes
Zarbo <i>et al.</i> ⁵	58 / M	synchronous	Yes	Yes
Schmidt <i>et al.</i> ⁶	72 / F	metachronous	Yes	No
Present case (1999)	65 / F	synchronous	No	No

such as irradiation were therefore not performed. Post-operatively there were no complications and no sign of recurrence after three years' follow-up.

Discussion

Turnbull *et al.*¹ reported the incidence of bilateral parotid tumours to be 0.8 per cent (24 of 1805 cases) of all parotid tumours, and Catanina *et al.*⁸ as 0.5 per cent (three of 544 cases). The most common tumour to occur bilaterally is Warthin's tumour, reported to represent 5-10 per cent of all Warthin's tumours.^{2,9} Bilateral involvement of other benign tumours is extremely rare. Other than Warthin's tumour, there were 35 reported cases of pleomorphic adenoma, 12 cases of oncocytoma and six cases of basal-cell adenoma in a review by Seifert² of bilateral benign salivary tumours, including the submandibular lesions. A review of the English literature revealed only four previously recorded cases of bilateral basal-cell adenoma of the parotid glands³⁻⁶ (Table I). Histologically, parotid basal-cell adenomas resemble dermal cylindromas. Many authors have also reported the synchronous occurrence of both types of lesion.^{10,11} All four reported cases of bilateral parotid basal-cell adenoma occurred synchronously with dermal cylindroma. To the best of our knowledge the present lesion is the first reported in the English literature to occur without coexisting dermal cylindroma.

Histologically basal-cell adenoma has various variants, including solid, trabecular, tubular and membranous variants.¹² As in the left lesion of the present case, basal-cell adenoma occasionally shows adenoid cystic changes, which are found in 10 per cent of patients.⁷ There is a notable histologic similarity between the adenoid cystic change in basal-cell adenoma and adenoid cystic carcinoma. The change can be differentiated from adenoid cystic carcinoma because it is based upon a gradual structural alteration of a benign tumour. In addition,

neither invasive growth nor mitotic figures should be present in the area.⁷ Adenoid cystic carcinomas invade the stroma and exhibit perineural invasion. Basal-cell adenoma with the adenoid cystic change must be distinguished from adenoid cystic carcinoma in order to plan appropriate treatment, as the oncologic prognoses of the two are quite different.

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