

CASE REPORT

Basal cell adenocarcinoma of the parotid gland detected in a patient with breast cancer

Alpaslan Ozgun,¹ Tolga Tuncel,¹ Levent Emirzeoglu,¹ Abdullah Haholu²

¹Department of Medical Oncology, Gulhane Military Medical Academy Haydarpasa Training Hospital, Istanbul, Turkey

²Department of Pathology, Gulhane Military Medical Academy Haydarpasa Training Hospital, Istanbul, Turkey

Correspondence to
Dr Alpaslan Ozgun,
alpozgun@yahoo.com

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SUMMARY

Basal cell adenocarcinoma (BCAC) is a rare tumour of the salivary glands and often associated with a good prognosis. The present case had BCAC of the parotid gland as the second primary tumour in addition to breast cancer. The patient was a 66-year-old woman who underwent mastectomy due to breast cancer. She then underwent adjuvant chemotherapy and adjuvant hormone therapy. After 4 years of disease-free follow-up, the patient presented with a swelling on the left cheek. The examination of the biopsy specimen revealed BCAC of the parotid gland. The patient then underwent left parathyroidectomy plus left neck dissection. Adjuvant radiotherapy was performed. Despite the therapy, the patient developed four local recurrences within 1 year, and then developed metastasis to the pleura. A swelling in the parotid gland in a patient with breast cancer should be carefully screened for the presence of a second primary tumour.

BACKGROUND

Breast cancer is the most common cancer in women. Patients with breast cancer may develop a second primary tumour.¹ Basal cell adenocarcinoma (BCAC) is a rare low-grade tumour of the salivary glands, and it is associated with a good prognosis.² The present case had BCAC of the parotid gland in addition to breast cancer, and the tumour showed an aggressive course despite the administered therapy.

CASE PRESENTATION

This case is of a 66-year-old female patient who presented with a mass in the left breast. The Tru-Cut biopsy obtained from the tumour mass

revealed invasive carcinoma of the breast. The patient therefore underwent a left-sided modified radical mastectomy. The pathological examination revealed invasive ductal carcinoma. The patient underwent adjuvant chemotherapy consisting of four cycles of epirubicin and cyclophosphamid. The tumour was oestrogen receptor positive and adjuvant hormone therapy (anastrozol 1 mg tablet) was initiated. After 4 years of disease-free follow-up, the patient presented with a swelling on the left cheek. A Tru-Cut biopsy was obtained from the swelling on the left cheek and the examination of the biopsy specimen revealed BCAC of the parotid gland ([figure 1](#)). Diagnosis was confirmed by the immunohistochemical analysis. Immunohistochemical staining showed that carcinoembryonic antigen (CEA) positive, S-100 positive ([figure 2](#)). Distant organ metastasis was not detected. The patient then underwent left parathyroidectomy plus left neck dissection. Adjuvant external radiotherapy (6600 cGy/33 fractions) was performed on the left parotid region. Tumour recurrence occurred in the parathyroidectomy operation site after 3 months. A large resection was performed. The pathological examination revealed BCAC. Surgical margins were tumour positive. The patient underwent another course of radiotherapy and she was placed on follow-up. She then developed three more local recurrences within 1 year, and underwent three operations. In the last operation, the patient underwent a resection of the left half of the lip and left eyelid involving a large area and flap reconstruction was performed after the resection. She developed recurrence despite the given treatment. The tumour mass was deemed to be unresectable. The patient

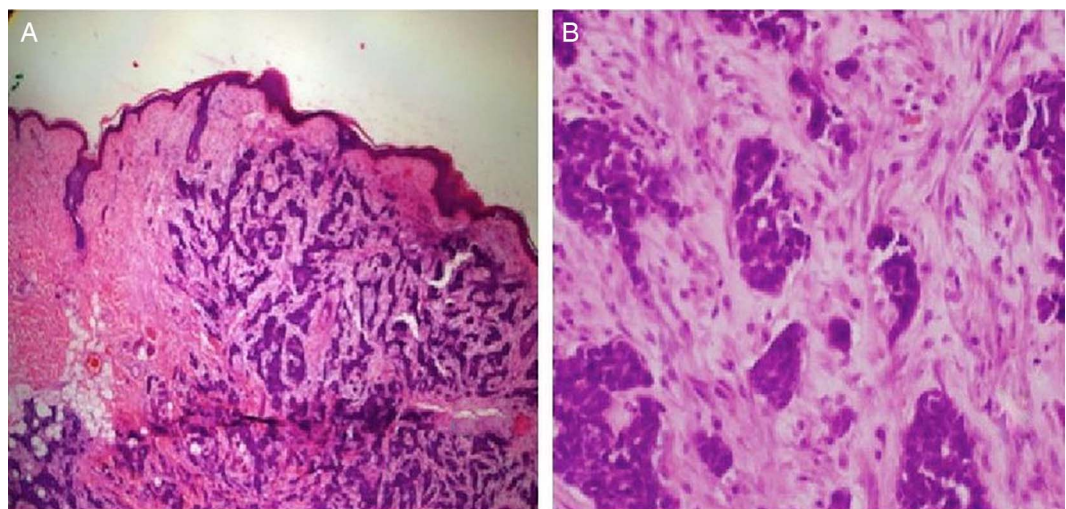


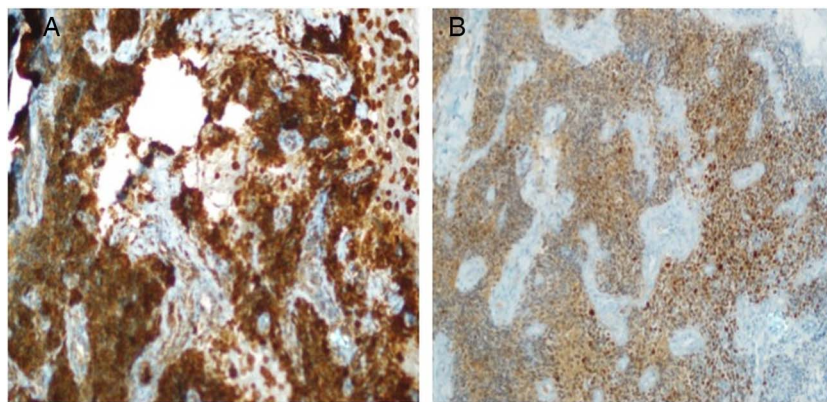
Figure 1 Histological appearance of basal cell adenocarcinoma (A) H&E, $\times 40$ and (B) H&E, $\times 400$.



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Figure 2 Immunohistochemical staining in basal cell adenocarcinoma. (A) Carcinoembryonic antigen (CEA) positive expression and (B) S-100 positive expression (H&E, $\times 200$).



therefore underwent chemotherapy with cisplatin and 5 FU, and minimal response was achieved after three cycles of chemotherapy. Chemotherapy was continued. She developed shortness of breath during the treatment. CT showed pleural thickness and pleural effusion on the right side. The patient underwent thoracentesis. A Tru-Cut biopsy was obtained from the pleural tissue. The examination of the pleural biopsy revealed findings consistent with metastasis of BCAC. Weekly paclitaxel therapy was initiated. She is still receiving paclitaxel therapy, and is followed-up by our clinic.

INVESTIGATIONS

A Tru-Cut biopsy was obtained from the swelling on the left cheek and the analysis of the biopsy specimen revealed BCAC of the parotid gland. CT showed pleural thickness and pleural effusion on the right side. A Tru-Cut biopsy was obtained from the pleural tissue and the analysis of the pleural biopsy revealed metastasis of BCAC. The diagnosis of BCAC confirmed by the immunohistochemical analysis.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis includes basal cell adenoma, adenoid cystic carcinoma and metastatic breast carcinoma. BCAC can be differentiated from these tumours by use of morphological and immunohistochemical analysis. Immunohistochemical staining shows CEA positive, S-100 positive.

TREATMENT

The patient underwent left parathyroidectomy plus left neck dissection due to BCAC. Adjuvant radiotherapy was performed on the left parotid gland area. The patient underwent four resection operations due to local recurrences. The patient received adjuvant chemotherapy with cisplatin and 5 FU. After progression, the patient received paclitaxel as salvage chemotherapy.

OUTCOME AND FOLLOW-UP

Despite the therapy, BCAC developed four local recurrences within 1 year, and then developed metastasis to the pleura. The patient is still receiving paclitaxel therapy, and she is followed-up by our clinic.

DISCUSSION

Breast cancer is the most common cancer type in women, and the incidence of developing a second primary tumour is higher in women with breast cancer. In the study by Molina-Montes *et al*, approximately 6000 patients with breast cancer were followed for the duration of 22 years, and they reported a 39%

higher risk of developing a second primary tumour in patients with breast cancer. The same study also indicated that the most common second primary tumour in patients with breast cancer was endometrium cancer followed by ovarian cancer in the third place.¹ The current patient developed BCAC of the parotid gland as the second primary tumour in addition to breast cancer.

BCAC is a rare tumour of the salivary glands. This tumour was described as a different entity for the first time by Klima in 1978.² Previous names used for BCAC include malignant basalioid tumour, malignant basal cell tumour, hybrid basal cell adenoma, adenoid cystic carcinoma, basalioid carcinoma of the salivary gland and atypical monomorphic adenoma. Ellis and Wiscovitch³ described the histological and clinicopathological characteristics of BCAC in a study involving 29 cases in 1990. In 1991, this tumour was classified by the WHO under malignant tumours of the salivary glands. In 2005, the tumour was renamed 'basal cell adenocarcinoma' and classified under basalioid tumours of the salivary glands by the WHO.⁴

BCAC is a rare low-grade tumour of the salivary glands, and it is associated with good prognosis. This tumour accounts for 1.6% of all tumours of the salivary glands and 2.9% of all malignant tumours of the salivary glands.⁵ This tumour is a malignant counterpart of the basal cell adenoma,⁶ and 90% of cases occur in the parotid gland. The tumour may rarely occur in the submandibular gland and minor salivary glands.⁷ Mean age at disease onset is 60 years. Local recurrence occurs in 35% of the cases. Metastasis occurs in less than 10% of the cases.⁸ The most common symptoms associated with BCAC include enlargement and tenderness in the salivary gland. The current case also presented to the doctor with these symptoms.

BCAC has four histological subtypes: solid, membranous, trabecular and tubular. One-third of the BCAC are of the solid type. The second most common type is membranous. Trabecular and tubular subtypes rarely occur.⁵ The present case had solid type BCAC. BCAC may sometimes occur in immunocompromised individuals. Markkanen-Leppänen *et al*⁹ reported a renal transplant recipient receiving immunosuppressive therapy that developed BCAC in the bilateral parotid glands.

Basal cell adenoma ranks first in the differential diagnosis of BCAC. BCAC and basal cell adenoma share common morphological and immunohistochemical features. However, BCAC shows nuclear pleomorphism, increased mitotic activity, necrosis, vascular invasion and lymphatic invasion, the features distinguishing it from basal cell adenoma.¹⁰ Adenocystic carcinoma is another tumour type that needs to be considered in the differential diagnosis. Adenocystic carcinoma is a high-grade tumour of the salivary gland and exhibits an aggressive course. This feature

distinguishes it from low-grade BCAC that is associated with a good prognosis.¹⁰ The present case exhibited an aggressive course despite the anticipation for classical BCAC. The patient developed four local recurrences in 1 year despite the given therapy, and then she developed pleural metastasis.

The curative treatment modality is surgery in the management of BCAC. Chemotherapy and radiotherapy possesses limited efficiency. Furthermore, carbon ion radiotherapy was introduced into practice in recent years.¹¹ Surgery, radiotherapy and chemotherapy were employed in the treatment of the present case.

To the best of our knowledge, this is the first case of BCAC of the parotid gland occurring in a patient with breast cancer. BCAC is generally associated with a good prognosis. However, the present case developed local recurrences despite the administered therapy, and the patient finally developed distant metastasis. A recent swelling in the parotid gland in a patient with breast cancer should be screened for the possible occurrence of BCAC, and it should be remembered that some cases with BCAC can show aggressive disease course.

Learning points

- Swelling in the parotid gland in a patient with breast cancer should be screened for the possible occurrence of basal cell adenocarcinoma (BCAC).
- It should be remembered that some cases with BCAC can show an aggressive disease course.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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