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Signet Ring Cell Carcinoma of the Breast as a Source of Pelvic Floor Metastatic Mass. A Case Report

T. Colak, T. Akca*, M. Dirlik*, M. Caglikulekci*, E. Seyrek**, L. Cinel***, R. Bozdogan***, S. Aydin**

Departments of Surgery*, Medical Oncology** and Pathology***, Medical Faculty of Mersin University, Mersin, Turkey.

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Abstract. Primary signet ring cell carcinoma of the breast is a very rare tumour. We present a case with pure signet ring cell carcinoma of the breast, which was recognized as metastasis on the pelvic floor, before developing breast symptoms and signs. A 40-year old woman was admitted with abdominal pain. First diagnostic effort revealed a cystic mass on the pelvic floor, compressing the colon and other neighbouring organs. A biopsy of the pelvic mass was performed. The histopathological examination revealed metastatic signet-ring cell carcinoma. At the time of the first operation, the mammary glands were not suspicious. No other sources of primary tumour were evidenced. An inflammatory sign developed in right breast two months after biopsy of the pelvic metastasis. The histopathology of the breast incisional biopsy revealed primary pure signet ring cell carcinoma of the breast. Because the oestrogen and progesterone receptor were negative in the tumoral tissue, the patient underwent chemotherapy followed by modified radical mastectomy, chemotherapy, and palliative resection of the metastatic mass. The patient was followed up for eight months. To our knowledge, in English literature, we believe that this case is the first report of signet ring cell carcinoma of the breast presenting with pelvic floor metastasis without breast sign.

Introduction

Primary signet ring cell carcinoma (SRCC) of the breast is a very rare tumour. It is usually considered as a variant mucinous carcinoma because of their intracellular mucin accumulation (1-2). But it is morphologically and prognostically different from mucinous carcinoma. STAINBRECHER & SILVERBERG recognized signet ring cell carcinoma as a distinct clinicopathological entity characterized by a large number of neoplastic cells (> 20% of cell population) (3). Moreover, the frequency of primary SRCC was difficult to evaluate because World Health Organization classification did not recognize it as a distinct entity (4).

We present a case of SRCC of the breast, which manifested initially by its pelvic floor metastasis.

Case report

A 40 years old woman was referred to our department with pelvic floor mass and right breast inflammation. She was admitted to the city hospital in June 2002 with abdominal pain since one year. Diagnostic tests revealed that the patient had a cystic mass on the pelvic floor compressing the colon externally. The cystic mass was drained transabdominally and a small portion of the cystic wall was also excised for pathological evaluation. The colon, ovary, uterus and other abdominal organs were normal at intraoperative exploration. Histopathological examination was reported as benign. However, the patient was readmitted with recurrence of pelvic mass one month after the operation and drainage with excisional biopsy was performed transvaginally. Histological examination identified a SRCC metastasis of unknown origin.

At the same time of the second operation, inflammatory sign developed in the right breast. The whole mammary gland and the subcutaneous tissue seemed to be substituted by an irregularly structured very firm tumour. A “peau d’ orange” phenomenon was seen with diffuse reddish sign and local pain. Non steroid anti-inflammatory drugs and antibiotics were advised for the breast inflammation and the patient was referred to our department for further investigations.

Our physical examination of the abdomen was normal and rectal digital examination was painful. There was diffuse expansion, erythema, oedema and sensitivity in the right breast. Left breast and bilateral axillary regions were normal. Right leg pain and restricted mobility developed subsequently.

Whole blood count and routine biochemical parameters were in normal limits. Carcino-embryonic antigen

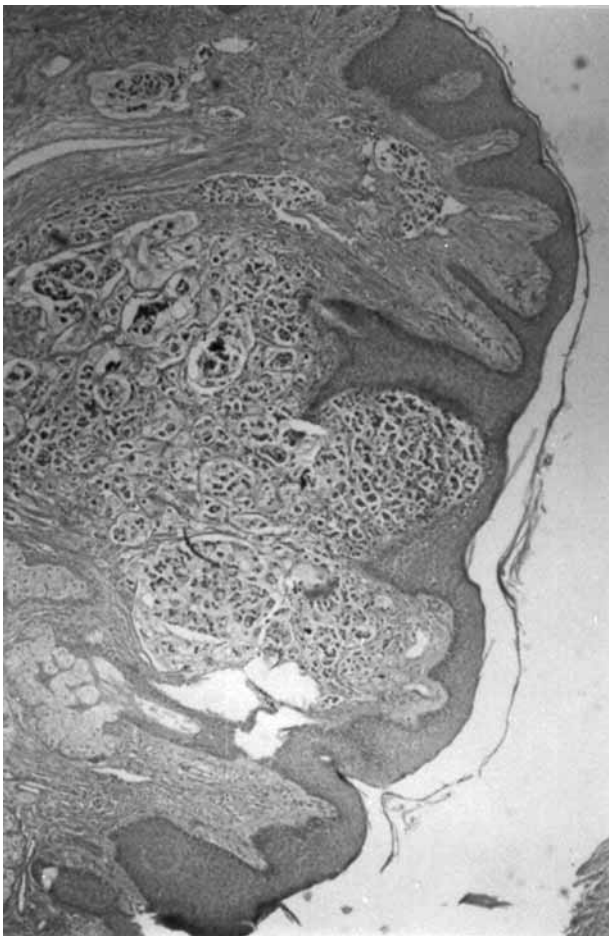

Fig. 1
Signet ring cell carcinoma of the breast characterized by a large mucin vacuole occupying the entire cytoplasm and displacing the nucleus to the periphery. Tumoral infiltration in breast skin, subdermal lymphatics and fascia. Tumoral cell thrombus in the epidermis and subdermal lymphatics is also shown (H&E × 200).

(CEA), carbohydrate antigen (CA) 15-3 and human chorionic gonadotrophin were increased but alpha-fetoprotein, CA 125, CA 19-9 as well as progesterone, estrodiol, prolactin, follicle stimulating hormone and testosterone were in normal ranges.

Plain chest radiography was normal. Computed abdominopelvic tomography showed a 31 × 22 mm diameter hypo dense image at the right ovary and 65 × 57 mm diameter metastatic soft tissue mass on the pelvic floor with invasion of the left gluteus minimus muscle and left side of the rectum. Plain bone radiography showed sub capital fracture of the right femur and whole bone scintigraphy revealed ischaemic necrosis and lytic lesions on the right femur head.

Panendoscopic examination revealed antral gastritis, at the colonoscopy there was no intraluminal pathology but an external mass image on the distal colon and rectum.

Routine gynaecological examination was normal and cervicovaginal smear showed atypical squamous cells.

Mammography and breast ultrasonography revealed a diffuse malign lesion in the right breast. However, left breast and bilaterally axillary regions were normal. Fine needle cytology and incisional biopsy of the lesion was reported as SRCC of the breast. Oestrogen receptors, progesterone receptors and carcinoembryonic antigen were negative in the specimen.

Final diagnosis was primary breast signet cell carcinoma with pelvic metastasis. Preoperative adjuvant chemotherapy was started. After the two cures of cisplatine and taxane protocol the breast mass had partially regressed and modified radical mastectomy, pelvic mass debulging and right oophorectomy were performed. A right partial hip replacement by prosthesis was subsequently done by the orthopaedist.

Histological examination of the surgical material revealed pure signet ring cell type of inflammatory breast carcinoma and metastasis of the SRCC in the pelvic floor and right ovary (Fig. 1).

Postoperative adjuvant chemotherapy was applied in the oncology department with four cures of cisplatine and taxane protocol. The patient is still alive with no further symptoms after eight months follow-up.

Discussion

SRCC of the breast behaves aggressively and is associated with a poor prognosis. Spread of disease past the regional lymph nodes often presents as haematogenous metastasis to the lung, liver and bone. Other common sites of metastasis include the stomach, endometrium and cervix (5-6). The incidence range of SRCC is 0.7-4.5% and 5 years mortality ranges from 45.5 to 60% (7, 8). However, the prognosis of SRCC of the breast is not well known on account of its rarity. It was reported that poor prognosis was seen in 60% of cases and almost all patients died within 7 years (9).

Signet ring cell carcinoma is encountered most commonly in gastric and gastrointestinal tract malignancies. Such cells can also be seen in other cancers, including bladder (10) and breast carcinoma (3). Numerous reports mention that the SRCC occur with distinct metastasis in unusual organs, including stomach (5), endometrium and cervix (6). Furthermore, more than 80 cases of signet ring cell carcinoma of the breast have been reported in literature. However, the present case is different as the painful pelvic mass clinically appeared 18 months before the primary breast tumour signs. This situation is very rare in the presentation of breast carcinoma. This is the first case of signet ring cell carcinoma revealed by pelvic floor metastasis without breast signs.

This situation caused the misdiagnosis, loss of time before appropriate diagnosis was made and therapy was started.

References

1. Izushi K., Imoto S., Hasebe T. Signet ring cell carcinoma associated with invasive ductal carcinoma of the breast : A case report. *Breast Cancer*, 1999, **25** : 223-6.
2. Hull M. T., Seo I. S., Battersby J. S., Csicsko J. F. Signet-ring cell carcinoma of the breast. A clinicopathologic study of 24 cases. *Am J Clin Pathol*, 1980, **73** : 31-5.
3. Steinbrecher J. S., Silverberg S. G. Signet ring cell carcinoma of the breast : The musinous variant of infiltrating lobular carcinoma. *Cancer*, 1976, **37** : 828-40.
4. Liu S. M., Chen D. R. Signet ring cell carcinoma of the breast. *Pathol Int*, 2000, **50** : 67-70.
5. Yim H., Jin Y. M., Shim C., Park H. B. Gastric metastasis of mammary signet ring cell carcinoma – a differential diagnosis with primary gastric signet ring cell carcinoma. *J Korean Med Sci*, 1997, **12** : 256-61.
6. Kennebeck C. H., Alagoz T. Signet ring cell carcinoma metastases limited to the endometrium and cervix. Case report. *Gynecol Oncology*, 1998, **71** : 461-4.
7. Eltorky M., Hall C., Osborne P. T., Zeky F. E. Signet ring cell variant of invasive lobular carcinoma of the breast. A clinicopathologic study of 11 cases. *Arch Pathol Lab Med*, 1994, **118** : 245-8.
8. Hull M. T., Seo S., Battersby J., Csicsko J. Signet ring cell carcinoma of the breast. A clinicopathologic study of 24 cases. *Am J Clin Pathol*, 1980, **73** : 31-5.
9. Merino M. J., Livolsi V. A. Signet ring cell carcinoma of the female breast. A clinicopathologic analysis of 24 cases. *Cancer*, 1981, **48** : 1830-37.
10. Moll C., Landolt U., Pedio G. Signet ring cell differentiation of transitional cell carcinomas of the bladder. *Acta Cytol*, 1996, **40** : 619-21.

T. Akca
Department of General Surgery
Mersin University Medical Faculty
Tip Fakultesi Hastanesi Zeytinlibahce C.
TUR-33079 Mersin, Turkey
Tel. : +90 324 337 43 00
Fax : +90 324 337 43 05
E-mail : takca@mersin.edu.tr