Primary breast mantle cell lymphoma with atypical relapse patterns

Dear Editor,

The term "primary breast lymphoma" (PBL) is used to define a malignant lymphoma primarily occurring in the breast in the absence of other previously detected lymphoma localizations. PBLs have a reported incidence ranging from 0.04 to 0.5% of all breast malignancies. PBLs account for less than 1% of all patients with non-Hodgkin's lymphomas (NHLs) and approximately 1.7% of all extranodal NHLs [1]. The most frequent subtype of lymphoma diagnosed in the breast is diffuse large B cell NHL. Because only 2 previous case reports of primary breast mantle cell lymphoma (PBMCL) have been reported, we present herein a case of PBMCL with an atypical clinical course in terms of the pattern of relapses.

A 67-year-old Caucasian female presented with a 4-month history of a progressively enlarging painless mass of the left breast in November 2002. The patient had a lumpectomy and a left axillary lymph node dissection performed and was then referred to our clinic. Histopathological examination of the lumpectomy and lymph node dissection material revealed small to medium-sized lymphoid cells, which were CD3+, CD5+, CD 19+, CD 20+, Cyclin D1+, and negative for CD2, CD10, and CD23. Ki67 showed a proliferation index of 45%. The results of fluorescein *in situ* hybridisation (FISH) showed IGH/CCND1 fusion [t(11;14)]. The case characterized as PBMCL, stage II AE, and the patient was treated with 4 cycles of CHOP (cyclophosphamide, adriamycin, vincristine and prednisolone) every 3 weeks and involved-field radiotherapy. After a 15-month follow up period with no evidence of disease the patient presented with a mass in the right palpebral conjunctiva and a mass in the buccal mucosa on the left. The relapse of PBMCL was confirmed by complete excision of both masses. Restaging showed no evidence of postoperative disease and the patient was given 6 cycles of R-ICE (rituximab, ifosfamide, carboplatin and etoposide) chemotherapy and followed up until her second relapse, which was manifested as multiple cutaneous abdominal nodules, also confirmed after excisional biopsy, 24 months after the last R-ICE administration. She was then given rituximab+fludarabine after the excision of some of her cutaneous nodules but without response. The patient died 73 months after the initial PBMCL diagnosis.

Although it is not possible to comment on survival based on one patient, our case survived longer than expected. In the literature, only 4 cases of primary or secondary breast MCL have been reported (Table 1). Due to systemic involvement, 2 of the 4 cases reported were accepted as secondary breast mantle cell lymphoma (cases 4 and 5). Interestingly, in 3 of the 4 patients, both breasts were involved with lymphoma. All but one, the reported PBMCL cases were older than 65 years. In our case, besides conventional chemotherapy, surgery was carried out at the time of diagnosis for the primary treatment of the malignant lesion of the breast and the axillary lymph nodes, and for relapses with palliative intent. Also, radiation therapy was given to the chest wall and left axillary region as adjuvant therapy. After extensive literature search, we could not find any other report of PBMCL relapsed in the palpebral conjunctiva, buccal mucosa and progressed as cutaneous nodules without systemic involvement. A possible explanation for the long survival of our patient may be the atypical relapse pattern without systemic involvement or the fact that the patient underwent local treatments in addition to chemotherapy during the treatment of both primary breast mass and cutaneous nodules. In conclusion, because the diagnosis of BCMCL has prognostic and therapeu-

Table 1. Clinical features in patients with MCL involving the breast

First author [Ref. no.]	*N*	*Age (years)*	*Presenting features*	*PBL or SBL*	*Location*	*Stage*	*Primary treatment*
Windrum et al. [2]	1	53	Breast mass	PBL	Right	1E	HDCT+ASCT
Fadare et al. [3]	2	77	Breast mass	PBL	Bilateral	–	–
Present case	3	66	Breast mass	PBL	Left	2E	Surgery+CT+RT
Boullanger et al. [4]	4	71	Lymphadenopathy	SBL	Bilateral	2E	CT
Hill et al. [5]	5	90	Breast mass	SBL	Bilateral	4E	–

PBL: primary breast lymphoma, SBL: secondary breast lymphoma, CT: chemotherapy, RT: radiotherapy, HDCT: high dose chemotherapy, ASCT: autologous stem cell transplantation

tic implications, its accurate identification using a test repertoire including cyclin D1 and t(11;14) is important.

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Resection of giant follicular thyroid carcinoma infiltrating the trachea

Dear Editor,

A 66-year-old female, coming from an iodine deficiency region, presented to the Department for Endocrine Surgery, Clinical Centre of Montenegro with a neck tumor. She was known to have a nodule in the left thyroid lobe for 14 years, which grew over the last 6 months. Inspection of the neck showed neck deformity on the left side, and a big fixed painless tumor of the left thyroid lobe. FNA of the thyroid showed follicular cells arranged in longitudinal, parallel arrays, with nuclear enlargement and colloid located centrally.

Ultrasonography of the neck showed a hypoechoic heterogeneous 14 cm large and irregular-contour nodule, with calcification in the central part. There was no enlargement of the cervical lymph nodes. Chest X-ray was normal. Thyroid hormones and calcitonin levels were normal but serum thyroglobulin was high (1000 ug/ml). Thyroid scintigraphy revealed a cold nodule in the left lobe. CT scan showed tracheal infiltration without obstruction. Tracheoscopy visualized a 1 cm wide space in the front part of the tracheal wall with malignant infiltration.

Neck exploration under general anaesthesia revealed a tumor infiltrating muscles on the front left neck side. The tumor also infiltrated the left jugular vein and the front part of the trachea. An extended total thyroidectomy with circular tracheal resection and termino-terminal anastomosis were carried out. During surgery we placed another sterile endotracheal tube into the trachea to obtain open airway during the tracheal resection and anastomosis. Frozen sections showed invasive thyroid cancer. We identified and preserved two parathyroid glands on the right side and both recurrent laryngeal nerves. Both sides of the neck were drained, and the neck was fixed in flexion position. There was no evidence of postoperative hypoparathyroidism, recurrent nerve palsy or respiratory insufficiency. In the first 3 postoperative days the patient was admitted at the Intensive Care Unit, and the following 8 days at the Department for Endocrine Surgery. The following day she was discharged in good condition.

The definite pathological diagnosis was invasive follicular carcinoma infiltrating the trachea. Tumor cells invaded the capsule in a mushroom-shaped growth, and also the muscles and the tracheal wall. Vascular invasion was detected in thyroid tissue.

Two months after the operation the patient received 3.7 GBq I-131 therapy. One year later, whole-body iodine scintigraphy was normal with low thyroglobulin level. Two years after surgery the patient received 5.55 GBq I -131 therapy because of appearance of metastasis in the first thoracic vertebra. A complete response was attained. The patient is on regular follow up for 4 years without evidence for local recurrence or distant metastasis.

Comprehensive use of diagnostic methods, especially CT scan, will give detailed information for operation [1]. Incomplete tumor resection impacts negatively disease prognosis [2]. The purpose of extended surgical treatment in differentiated advanced thyroid carcinomas is to guarantee sufficient respiratory and alimentary functions and obtain local disease control. Adjuvant radioiodine therapy follows [3]. In the Gasiert et al. study of 82 patients, the mean survival was 9.4 years and 10-year survival was 40% after resection and reconstruction [4].