Phyllodes tumour in pregnancy: a case report

Jeffrey C. Way, MD; Beverley A. Culham, BSc, MHSA

Phyllodes tumour (cystosarcoma phyllodes) is a rare breast tumour that grows rapidly and to a relatively large size, especially during pregnancy. These tumours may be classified as benign, borderline or malignant. They have a high incidence of local recurrence but little tendency to metastasize to distant organs. The question of whether the tumour is hormone dependent remains unresolved. This report describes the case of a patient who had a phyllodes tumour that first became apparent in her 31st week of pregnancy. After enucleation and subsequent wide excision she remained tumour free through a second pregnancy. Although the follow-up period is short, it appears that subsequent pregnancy is not necessarily associated with recurrent or new disease for patients who have had their initial tumour completely excised. The goal for the management of these tumours is complete surgical excision.

La tumeur phyllode (cystosarcoma phyllodes) est une tumeur rare du sein, qui grossit rapidement et atteint une taille relativement importante, surtout au cours de la grossesse. Ces tumeurs peuvent être bénignes, à la limite de la malignité, ou malignes. Elles ont une forte incidence de récidive locale, mais elles ont peu tendance à produire des métastases dans des organes distants. On n'a pas encore déterminé si la tumeur est hormonodépendante. Ce rapport décrit le cas d'une patiente dont la tumeur phyllode est devenue apparente pour la première fois au cours de la 31e semaine de grossesse. Après une énucléation et une excision large subséquente, elle n'a pas eu de tumeur pendant une deuxième grossesse. Même si la période de suivi a été brève, il semble qu'il n'y ait pas nécessairement de lien entre une grossesse subséquente et la récidive ou l'apparition d'une tumeur chez les patientes qui ont subi une excision complète de la tumeur initiale. L'excision chirurgicale complète constitue le but du traitement de ces tumeurs.

Phyllodes tumour (cystosarcoma phyllodes) is a rare tumour of the breast representing less than 1% of all breast tumours[1] and approximately 2.5 % of all fibroepithelial breast lesions.[2] These tumours are classified as benign, borderline or malignant based on their histologic appearance.[2,3] In various studies the incidence of benign tumours has been reported to be between 35% and 91%.[2,4–6] The incidence of the malignant form is estimated to be 6.2% based on data from a general surgical facility.[7]

Phyllodes tumour grows rapidly especially during pregnancy.[8,9] The presence of steroid receptors was noted by Rao, Meyer and Fry[10] and Palshof and colleagues;[11] however, the hormone dependency of this disease has remained controversial.

Case report

A 35-year-old woman at approximately 34 weeks' gestation was referred with a rapidly enlarging tumour, 5 cm in dimension, in the upper inner quadrant of the right breast. This tumour had first become apparent to the patient 3 weeks earlier. The mass was firm, mobile and smooth and suggestive of a fibroadenoma. Needle aspiration showed overall features consistent with a fibroadenoma; however, it was suggested this initial diagnosis be confirmed by excision biopsy.

The tan–pink tumour measured about 5 cm in its greatest dimension and was fairly well circumscribed but not encapsulated. Cut sections showed numerous large fronds of pink tissue within the lesion with no evidence of hemorrhage or necrosis. Microscopic examination revealed that the fronds consisted of stroma with an

From the Department of Surgery, Division of General Surgery, Calgary District Hospital Group, Calgary, Alta.

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Correspondence to: *Dr. Jeffrey C. Way, A304 1600–90th Ave. SW, Calgary AB T2V 5A8*

increase in cellularity. Mitotic figures were noted but these were not numerous. Although the stromal cells contained slightly enlarged nuclei, pleomorphism was not striking and the borders of the tumour appeared to be noninfiltrating. For these reasons we thought the tumour was benign. The lesion was reported to be estrogen receptor- and progesterone-receptor negative.

Because the tumour extended to the margin of the biopsy there was a possibility that it had not been completely excised, so a subsequent wide excision with lumpectomy was carried out to ensure tumour-free margins. The pathology report showed no residual phyllodes tumour. A 6-month follow-up mammogram gave negative results. One year after tumour excision the patient became pregnant a second time and delivered a healthy infant. Mammograms obtained 6 months and 18 months after the delivery showed no abnormalities.

Discussion

Even though the study by Norris and Taylor[3] more clearly distinguished the histologic variation of phyllodes tumour, little is known about the correlation between the histotypes and the recurrence of the tumour or metastatic spread. Because of continued uncertainties about the natural history and prognostic variations of this disease and its relative rarity, there is still some confusion about how to appropriately manage such tumours.

There is no literature that documents how this tumour is affected by pregnancy apart from the fact that phyllodes tumour appears to grow more rapidly during this state.[8] In general, phyllodes tumours have a low risk of metastasis[2,7] but a very high recurrence rate.[2,4,12–15]

Local recurrence of phyllodes tumour ranges from 14% to 29%[2,4,12–15] and generally occurs within 2 years of primary treatment.[2,6,13] Local recurrence can not be predicted by the size of the tumour or its histotype, and recurrence is generally believed to be a result of inadequate excision.[2,4] Hajdu, Espinosa and Robbins[13] speculated that because of the multinodular centrifugal growth pattern of these tumours, peripheral stromal nodules or microscopic extensions may be inadvertently amputated during excision.

Even though local recurrence rates appear to be higher for more conservative breast surgery the long-term survival rate does not appear to be affected.[4,15] In general, it is accepted that treatment options should be individualized with the primary goal of complete surgical removal with a free margin of 2 or 3 cm.[16] Tumour management should take into consideration tumour and breast sizes, extent of disease, tumour aggressiveness and the patient's decision.[6]

Several biochemical studies have investigated the existence of estrogen and progesterone receptors in phyllodes tumours, but the results have been variable, with some tumours exhibiting no receptors and others exhibiting low to relatively high levels.[9–11,17,18] Friedl and associates[19] found that estrogen-receptor negativity does not necessarily indicate that the tumour is unresponsive to estrogen. As a result, the hormone dependency or responsiveness of these tumours continues to be controversial.

Summary and conclusions

Phyllodes tumour is a rare, fibroepithelial tumour of the breast that may be considered benign, borderline or malignant depending on its histologic make-up. The tumour grows rapidly, especially during pregnancy, but its hormone dependency remains controversial. These tumours have a high recurrence rate possibly due to inadequate excision. Therefore, wide resection must be achieved if at all possible. It appears from this case report that pregnancy is not necessarily associated with recurrent or new disease. Even though the follow-up was short, the patient in this case report went through a second pregnancy with no indication of either a recurrence or development of another tumour at another site.

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Notices
Avis

Quebec Association of Urologists

The 23rd annual meeting of the Quebec Association of Urologists will be held from Nov. 13 to 15, 1998, at the Westin Mont Royal Hotel, Montreal. For further information please contact: Ms. Jacqueline Deschênes, Quebec Association of Urologists, 2 Complexe Desjardins, East Tower, Door 3000, Montreal QC H5B 1G8; tel 514 350-5131, fax 514 350-5181

Gastrointestinal malignant disease

Sponsored by Continuing Education, Faculty of Medicine, University of Toronto, the 1998 update on digestive diseases: gastrointestinal malignancies will be held on Nov. 13, 1998, in the 18th Floor Auditorium at Mount Sinai Hospital, Toronto. Course credits: MOCOMP and AMA Category I. For further information contact: Continuing Education, Faculty of Medicine, University of Toronto, 150 College St., Toronto ON M5S 3E2; tel. 416 978-2718, fax 416 971-2200, jan.spencer@utoronto.ca

Colorectal disease in 1999

The Cleveland Clinic Florida will sponsor the 10th annual international colorectal disease symposium under the title "Colorectal Disease in 1999: an International Exchange of Medical and Surgical Concepts." The course will be held from Feb. 11 to 13, 1999, at the Marriott's Harbor Beach Resort in Fort Lauderdale, Fla. The symposium director is Steven D. Wexner. Topics of guest lectures and panels include controversies in ulcerative colitis surgery, frustrating fistulas, treating complications, and reoperative surgery. Four areas will be analysed in detail: colorectal neoplasia, inflammatory bowel disease, colorectal physiology and technical aspects of surgery. For further information contact: Cleveland Clinic Florida, Department of Continuing Education, 2950 West Cypress Creek Rd., Fort Lauderdale FL 33309; tel 800 359-5101 x5056, fax 954 978-5539, jagelms@cesmtp.ccf.org

Mayo interactive surgical symposium

The Mayo interactive surgical symposium will be held from Feb. 18 to 20, 1999, at Marriott's Camelback Inn Resort, Scottsdale, Ariz. Sponsored by the Mayo Clinic Scottsdale, it will present interactive sessions designed to update general surgeons on state-of-the-art current issues in breast surgery, trauma and critical care, endocrine, gastrointestinal/hepatobiliary and vascular and thoracic surgery. Course directors are John H. Donohue and William M. Stone. Course credits: AMA Category 1. For further information contact: Kristin Eberhard, Mayo Clinic Scottsdale, 13400 East Shea Blvd., Scottsdale AZ 85259; tel 602 301-7552, fax 602 301-8323

Urogynecology and disorders of the female pelvic floor

The Mayo Clinic Scottsdale will sponsor the 8th annual course on urogynecology and disorders of the female pelvic floor, to be held from Mar. 25 to 27, 1999, at Marriott's Camelback Inn Resort, Scottsdale, Ariz. The course director is Jeffrey L. Cornella. Course credits: AMA Category I and ACOG. For further information contact: Kristin Eberhard, Mayo Clinic Scottsdale, 13400 East Shea Blvd., Scottsdale AZ 85259; tel 602 301-7552, fax 602 301-8323

Interventional ultrasonography

The 8th International Congress on Interventional Ultrasound will be held from Aug. 31 to Sept. 3, 1999, at Herlev Hospital, University of Copenhagen, Copenhagen, Denmark. Further information may be obtained by contacting the Department of Ultrasound — Herlev Hospital, University of Copenhagen, DK-2730 Denmark; tel +45 44 88 32 40, fax +45 44 94 80 09, ultrasound@herlevhosp.dk