Case Report	Case of Benign Phyllodes Tumour Associated with Hypoglycemia
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Abstract -

Benign phyllodes tumour is a rare non-cancerous growth of the breast characterized by a solitary unilateral tumour in one breast, or rarely multifocal in one or both breasts. It is diagnosed on the basis of physical examination, imaging studies, and fine-needle aspiration cytology. In this report, we present a case of benign phyllodes tumour with an unusual presentation of recurrent attacks of hypoglycemia, which can be attributed to secretion of insulin-like growth factor II from the tumour.

Keywords: breast neoplasms, cystosarcoma phyllodes, hypoglycemia, insulin-like growth factor

Introduction

Phyllodes tumours of the breast are rare and account for < 1% of breast tumours, over half of which are benign. They usually present as breast lumps detected during routine examination, but are rarely detected with imaging modalities such as mammography. Phyllodes tumours may resemble fibroadenomas and the two are often confused during diagnosis. Most women who are diagnosed with phyllodes tumour are premenopausal. In very rare cases, adolescent girls may be diagnosed with this type of breast tumour. These tumours can be differentiated from malignant breast tumours on the basis of the mitotic index of the specimen obtained by a core needle biopsy (1-4). On mammography, these tumours appear to have a well-defined edge, and are not usually found near microcalcifications. In many cases, complete surgical removal is advocated because of their high tendency of recurrence (5,6).

Women who undergo surgery for phyllodes tumour removal require close surveillance with follow-up mammograms, and physical examination at regular intervals. Malignant phyllodes tumours are best managed with wide excision of normal breast tissue around the tumour to obtain a 1 cm margin of normal appearing breast tissue. In most cases, radiation therapy is not required. Very large malignant phyllodes tumours may require mastectomy. Axillary dissection is not routinely recommended as metastases rarely occur. There is a low chance of recurrence for a benign phyllodes tumour in patients aged 45 years or older.

Case Report

We report the case of a 20-year-old woman who presented with a large lump in the left breast since three months, and generalized weakness and fatigue since one month. The tumour was extremely large (approximately 34 cm in diameter) and had a distinct boundary. It was firm in consistency with structural distortion of the left breast without any ulceration or involvement of the nipple-areolar complex. Engorged veins were present over the entire left breast, whereas the right breast and axilla appeared normal (Figure 1).

The patient was diagnosed with phyllodes on the basis of clinical examination, mammogram, and fine needle aspiration cytology of the tumour. The patient developed symptoms of sweating, delirium and loss of consciousness, and was found to have significant hypoglycemia (blood glucose: 20 mg/dL). Fasting insulin (12 uIU/ mL) and C-peptide (1.2 ng/mL) were within the normal limits (< 17 uIU/mL and 0.8–3.1 ng/mL, respectively). Ultrasonography of the abdomen ruled out the presence of insulinomas. Despite corrective measures with intravenous dextrose infusions, and oral glucose supplementation, the



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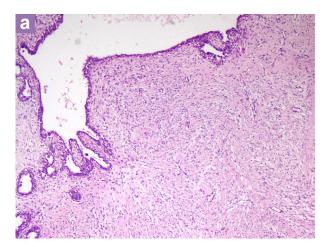
patient persisted to have recurrent episodes of hypoglycemia. A mastectomy of the left breast was performed in view of the bulk of the tumour, and intraoperatively it showed no violation of the deeper planes or invasion into the chest wall. The patient recovered with complete resolution of the hypoglycaemic episodes. Final histopathological analysis confirmed the diagnosis (Figure 2) and complete excision of the tumour.

Discussion

Fasting hypoglycaemia generally occurs in certain types of malignant tumours, as a part of paraneoplastic syndrome or usually in the extra-pancreatic tumours except for insulinomas (7-12). Previous studies by Hino et al. (13,14)



Figure 1: Pre-operative image showing the tumour in the left breast.



have indicated that the insulin-like growth factors (IGFs) are overexpressed in phyllodes tumours of the breast, which has been attributed to the presence of subclinical hypoglycemia in these patients. The clinical hypoglycemia seen in our patient may be attributed to the large tumour size, as tumour size has been correlated to the amount of IGF secreted (15). This report illustrates the rare incident of benign phyllodes tumour-associated hypoglycaemia, which was reconciled following the removal of the tumour. It is likely that elevated tumour IGF levels may have contributed to the hypoglycaemic episodes in this patient.

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Conflict of interest

Nil.

Fund

Nil.

Authors Contributions

Conception and design, analysis and interpretation of the data, drafting of the article, critical revision of the article for the important intellectual content: RPN

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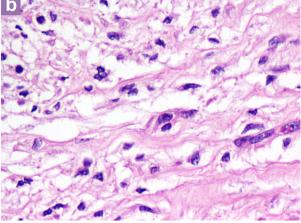


Figure 2: Histopathology slides; (a) Photomicrograph shows a biphasic tumour with glandular epithelium surrounded by cellular stroma (haematoxylin & eosin staining, 20× magnification).
(b) Photomicrograph shows spindle cells with plump nuclei and inconspicuous mitosis (haematoxylin & eosin staining, 40× magnification).

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References

- Zahner J, Bässler R. The rate of mitosis in cystosarcoma phyllodes (phyllodes tumor, WHO) of the breast. An analysis of 47 cases. Arch Gynecol Obstet. 1989;246(3):153–157.
- 2. R Awwal, Shashi SA, Khondokar MS, Khundkar SH. Management of recurrent phyllodes with full thickness chest wall resection. *Bangladesh J Plast Surg.* 2010;**1(1)**:9-13.
- Azzopardi JG, Ahmed A, Millis RR. Problems in breast pathology. *Major Probl Pathol.* 1979; 11(i-xvi):346–364.
- Jacklin RK, Ridgway PF, Ziprin P, Healy V, Hadjiminas D, Darzi A. Optimising preoperative diagnosis in phyllodes tumour of the breast. *J Clin Pathol.* 2006; 59(5):454–459.
- Richard M, Ward MD, Harry L, Evans MD. Cystosaecoma Phyllodes – A Clinicopathologic Study of 26 cases. *Cancer*. 1986;**58(10)**:2282–2289.
- 6. Mangi AA, Smith BL, Gadd MA, Tanabe KK, Ott MJ, Souba WW. Surgical management of phyllodes tumours. *Arch Surg.* 1999;**134(5)**:487–492.
- Bleau B, Vlahos W, Guyga H. High molecular weight insulin-like growth factor-II (HMW IGF-II) associated with fasting hypoglycemia and a phyllodes tumor of the breast. *Clin Invest Med.* 1991;14 Suppl:36(abstr).

- Aguiar Bujanda D, Rivero Vera JC, Cabrera Suárez MA, Aguiar Morales J, Christol R, Bohn Sarmiento U, et al. Hypoglycemic Coma Secondary to Big Insulinlike Growth Factor II Secretion by a Giant Phyllodes Tumor of the Breast. *The Breast J.* 2007;13(2): 189–191.
- 9. Kataoka T, Haruta R, Goto T, Sugino K, Asahara T, Dohi K, et al. Malignant Phyllodes Tumor of the Breast with Hypoglycemia: Report of a Case. *Jpn J Clin Oncol.* 1998;**28(4)**:276–280.
- Ishido H, Kitaoka M, Uchigata M, Kaji M, Maeda M, Nagayama T. A case of phyllodes tumor of the breast producing IGF-II. *Nippon Naibunpitsu Gakkai Zashi*. 1992;68:825.
- Shapiro ET, Bell GI, Polonsky KS, Rubenstein AH, Kew MC, Tager HS. Tumor hypoglycemia: Relationship to high molecular weight insulinlike growth factor-H. *J Clin Invest.* 1990;85(5): 1672–1679.
- 12. Petrek JA. *Phyllodes tumor*. In: Harris JR, Lippman ME, editors. Disease of the Breast. Philadelphia (PA): Lippincott-Raven. 2000. p. 863–869.
- 13. Hino N, Nakagawa Y, Ikushima Y, Yoshida M, Tsuyuguchi M. A case of a giant phyllodes tumor of the breast with hypoglycemia caused by highmolecular-weight insulin-like growth factor II. *Breast Cancer*. 2010;**17(2)**:142–145.
- 14. Reinfuss M, Mitus J, Duda K, Stelmach A, Rys J, Smolak K. The treatment and prognosis of patients with phyllodes tumor of the breast: an analysis of 170 cases. *Cancer*. 1996;77(5):910–916.
- 15. Li TCM, Reed CE, Stubenbord WT Jr, Ettinghausen S, Peterson CM, Jovanovic L. Surgical cure of hypoglycemia associated with cystosarcoma phyllodes and elevated non-suppressible insulin-like protein. *Am J Med.* 1983;**74(6)**:1080–1084.