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A CASE OF BENIGN RECURRENT PHYLLODES TUMOUR WITH HYPOGLYCEMIA

General Surgery

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KEYWORDS

Introduction

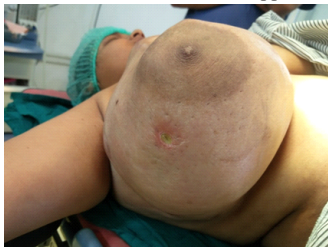
- Phyllodes tumours (PT) of the breast are fibro-epithelial neoplasms that are known to recur locally in up to 19% of patients.
- Phyllodes tumours of the breast are rare and account for < 1% of breast tumours, over half of which are benign. They usually present as breast lumps detected during routine examination, but are rarely detected with imaging modalities such as mammography.
- Most women who are diagnosed with phyllodes tumour are pre-menopausal. 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 micro-calcifications. In many cases, complete surgical removal is advocated because of their high tendency of recurrence (5,6).
- The failure to achieve adequate surgical margins is an important risk factor for local recurrence.
- This, however, is a common problem as PT are clinically similar to the more common fibro-adenoma and are therefore often locally excised without any gross surgical margins. It is still debatable as to whether it is necessary to subject the patient to repeat surgery to obtain pathologically negative margins after a diagnosis of a benign or borderline PT is made.
- Although the majority of recurrences are histologically similar to the initial tumour, a malignant recurrence is possible.
- Malignant tumours can metastasize through the haematogenous route and metastases are associated with a poor prognosis as they are poorly responsive to conventional chemotherapy.

Objectives

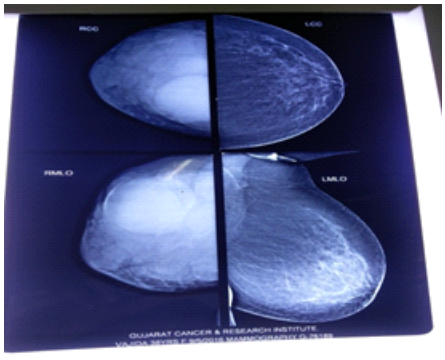
- In this report, we present a case of recurrent benign phyllodes tumour with an unusual presentation of recurrent attacks of hypoglycemia.

Case Report

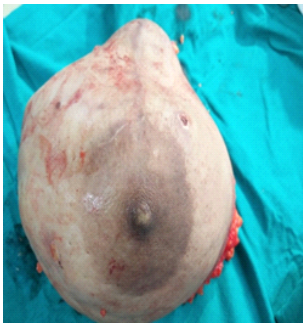
- We report the case of a 38-year-old woman who presented with a large lump in the right breast since three months, and generalized weakness and fatigue since one month. The tumour was extremely large (approximately 34 cm in diameter) and had a distinct boundary. It was firm in consistency with structural distortion of the left breast without any ulceration or involvement of the nipple-areolar complex. Engorged veins were present over the entire right breast, whereas the left breast and axilla appeared normal



- The patient operated twice on same breast two years before for similar complain and was having phyllodes tumour on histopath report.
- 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 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.
- For reconstruction , LD flap was used.
- The patient recovered with complete resolution of the hypoglycaemic episodes. Final histopathological analysis confirmed the diagnosis and complete excision of the tumour



- Final histopath report was suggestive of recurrent phyllodes tumour , high grade malignant involving all the quadrants of breast
- Tumour size 32*20*11cms
- Stromal proliferation seen and stromal pleomorphism seen

- 6-8 mitosis /10hpf seen
- Overlying skin and nipple free of tumour

All 11 Lymphnodes are free of tumour

Discussion

- 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.
- In this report, we have presented 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.
- 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) 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
- Local recurrence can usually be controlled by further wide excision and mastectomy is not invariably required. Mastectomy should, however, be considered for local recurrence after local surgery for borderline or malignant tumors. Kaprisi et al. [10] concluded that tumor size and surgical margins were found to be the principal determinant of local recurrence Occasionally aggressive local recurrence can result in widespread chest wall disease with direct invasion of the underlying lung parenchyma. Isolated reports of good palliation in this situation with radiotherapy have been published.

Conclusion

The clinical finding of a large, firm, non-tender, well-defined, mobile tumour with gradual growth should lead to a suspicion of a phyllodes tumour.

Our results and the review of the other literatures lead to the following therapeutic recommendations:

1. Benign phyllodes tumour warrants wide local excision with a 2 cm tumour-free zone.
2. Borderline malignant phyllodes tumours should be treated via simple mastectomy.
3. Axillary dissection is recommended only if nodes are palpable.
4. Adjuvant radiation is necessary, if wide local spread of the tumour is present and a resection of the lesion with a 2 cm tumour-free zone is not possible. Close follow-up is mandatory.