Letters to Editor

Non-islet cell tumour hypoglycaemia: A case report

Sir,

Hypoglycaemia is usually a manifestation of pancreatic islet cell tumours producing insulin, primary or secondary adrenal insufficiency, advanced liver disease, phaeochromocytoma, insulin growth factor (IGF)-1 secreting tumours, hypothyroidism, substances interfering with insulin, and insulin receptor-mediated metabolism or antibodies interfering with insulin. Non-islet cell tumour hypoglycaemia (NICTH) has been related to the production of IGF-II or its high-molecular-weight precursor (big IGF-II) by tumours, which are mesenchymal malignancies such as fibrous tumours, sarcomas and mesothelioma. It results from inhibited hepatic glycogenolysis, gluconeogenesis and diminished lipolysis, indicating increased insulin-like activity in the body. In addition, there is enhanced glucose utilisation due to the high metabolic needs of a large tumour. Big IGF- II is a form of pro-IGF-II, which is biologically active and has a greater bioavailability.[1]

There have been a very few case reports of fasting hypoglycaemia in patients with phyllodes tumour (PT). PTs are rare fibroepithelial lesions that make up 0.3–0.5% of breast tumours. We describe such a case, its management and a brief review of the literature.

A 37-year unmarried female presented with a large chest wall tumour (25 cm × 20 cm × 20 cm), which was diagnosed to be PT on core biopsy. On the day of surgery, she complained of dizziness and the blood sugar testing showed a value of 16 mg/dL. On eliciting history, she said that she had experienced such episodes previously. She had difficulty in getting up in the morning, sometimes did not recognise relatives, experienced sweating, and palpitations; mostly during the early morning. The supervised overnight fast test was done. The patient went into hypoglycaemia after 6 h, the critical samples for glucose and insulin C-peptide were tested, levels were very low. This ruled out the possibility of an insulinoma. This suggested that hypoglycaemia might be tumour induced. The growth hormone (GH) and IGF-I levels were low. Oral prednisolone was administered at night and morning 4 am and 6 am and glucose levels were done, which were found to be normal. Pre-operative blood sugar values were low (60 mg%) and supplementary dextrose infusion was continued intraoperatively, titrated with repeated blood samples. Mastectomy was performed and breast reconstructed with deep inferior epigastric perforator flap. Intraoperatively, dextrose requirements decreased after excision of the tumour. Postoperatively, blood sugar was measured 2nd hourly for first 6 h and later 4th hourly and the levels remained normal.

The first case of NICTH was reported in 1983 in a 43-year-old woman with a large left breast PT, 28 cm in diameter and weighing 4.2 kg, successfully treated with mastectomy. This patient's hospital course was marked by recurrent profound hypoglycaemia followed by loss of consciousness but recovered without any neurological deficit each time after dextrose administration. The plasma levels of non-suppressible insulin-like protein were increased, whereas levels of insulin were normal.[2]

Another similar case presentation was found in a 66-year-old woman who also recovered after removing a 6 kg weight PT, describing elevated values of big IGF-II in the blood and in the tumour extract.[3] Furthermore, a case of hypoglycaemia has been reported in a 54-year-old woman who was treated with surgery (malignant PT of 9 kg and 35 cm), and post-operative medroxyprogesterone acetate because of lung metastasis. In this case, the plasma level of IGF-II was moderately diminished postoperatively, whereas the tumour extract IGF-II level was high.[4]

Moreover, an unfortunate case of irreversible hypoglycaemic coma during fine-needle aspiration of a PT breast has been described, but the patient subsequently died on the 8th day of admission due to aspiration pneumonia and renal failure. Low levels of Insulin, IGF-I, C-peptide, and GH suggested an insulinomimetic compound to be the cause of hypoglycaemia. Postmortem of the patient revealed hypoglycaemic brain damage with hippocampal necrosis.[5]

To conclude, big sized PTs may manifest with NICTH, and simple management can prevent poor outcomes. The complete resection of the tumour produces a prompt normalisation of the serum glucose level.

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Conflicts of interest

There are no conflicts of interest.

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