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Giant insulinoma in a 15-year-old man: A case report

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ABSTRACT

INTRODUCTION: Giant insulinomas are extremely rare pancreatic neuroendocrine tumor.
PRESENTATION OF CASE: A 15-year-old man presenting with acute onset of lightheadedness was found to have serum glucose of 1.5 mmol/L. The blood collected from the hypoglycemic episode showed an inappropriately high insulin and C-peptide level. Abdominal computerized tomography showed a 12.5 cm well-defined, lobulated hypervascular mass at pancreatic tail, without any evidence of metastasis. En bloc resection with distal pancreatectomy, and splenectomy was successfully performed. The pathological examination confirmed insulinoma, with benign characteristics. Follow-up after the procedure revealed neither hypoglycemic, nor hyperglycemia.
CONCLUSION: We report the youngest case of a giant insulinoma. Despite the size of the tumor, the pathological report confirmed the benign characteristics. However, long-term follow-up is still essential to detect recurrence in the future.

1. Introduction

Insulinomas are rare neoplasms with a reported incidence of 4 cases per million population-year [1]. The majority of patients are above 50 years of age and is slightly more common in women than men [2]. Insulinomas are usually very small, with 80% being less than 2 cm in diameter [3]. Giant insulinomas (>9 cm in diameter) are even extremely rare. Less than 40 cases were reported since 1927 [4–6]. Mean age at the presentation was 54 years, with the youngest reported case at 29 years. Most giant insulinomas showed metastatic features at the time of diagnosis. We report an exceptional case of a 12.5 cm insulinoma in a 15-year-old patient with benign characteristics.

1.1. Presentation of case

A 15-year-old man was seen at emergency department with acute onset of lightheadedness, diaphoresis, and palpitations in the evening after fasting for 7 h. He had experienced repetitive episodes of dizziness for 2 months without any weight change. His serum glucose level was 1.5 mmol/L. His symptoms resolved after dextrose administration. He had no family history of insulinoma or multiple endocrine neoplasia type 1 (MEN-1). Physical exams showed a large palpable mass at left upper quadrant of the abdomen.

The subsequent analysis of the blood collected from the hypoglycemic episode showed an inappropriately high insulin level of 13.34 μU/mL (normal <3 μU/mL), and a high C-peptide level of 2.2 ng/mL (normal < 0.6 ng/mL). Abdominal contrast-enhanced computed tomographic (CT) scan showed a large well-defined, lobulated hypervascular mass with areas of low attenuation and calcifications at pancreatic tail, measuring 12 cm in largest diameter. The mass was adjacent to stomach, spleen, and left kidney. There were no pancreatic duct dilatation, no liver mass and no significant abdominal lymphadenopathy (Fig. 1). Serum intact parathyroid hormone (iPTH), calcium, and prolactin level were all normal.

The patient was admitted for continuous glucose infusion and frequent meals intake. The plasma glucose was maintained between 3.5–5 mmol/L by average glucose infusion at 10 g/hour. Esophagogastroduodenoscopy (EGD) was performed to exclude gastrointestinal tract invasion. Ten days after his admission, the patient underwent surgical exploration. Careful palpation demonstrated splenic attachment but not kidney. Therefore, en bloc resection with distal pancreatectomy, together with splenectomy was performed. The patient did not receive vaccination 2-week prior to surgery due to the urgency of the operation. The gross appearance showed 12.5 × 10.0 × 8.3 cm mass located at the pancreatic tail. Cut sections of the mass showed gray tan surface with

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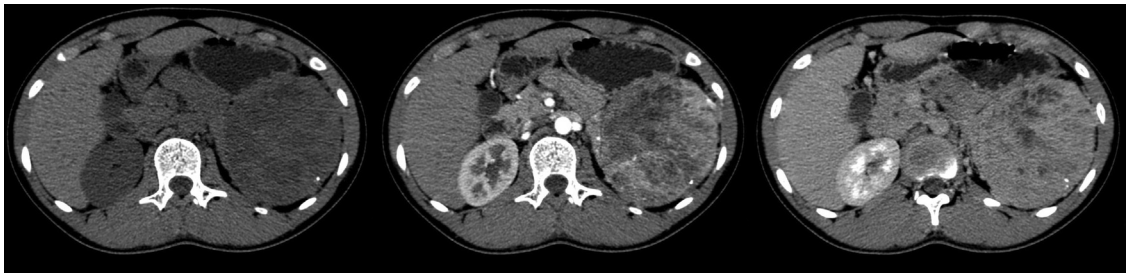

Fig. 1. CT of the abdomen showing the pancreatic mass in (from left to right, respectively) pre-contrast, arterial, and portal phase.

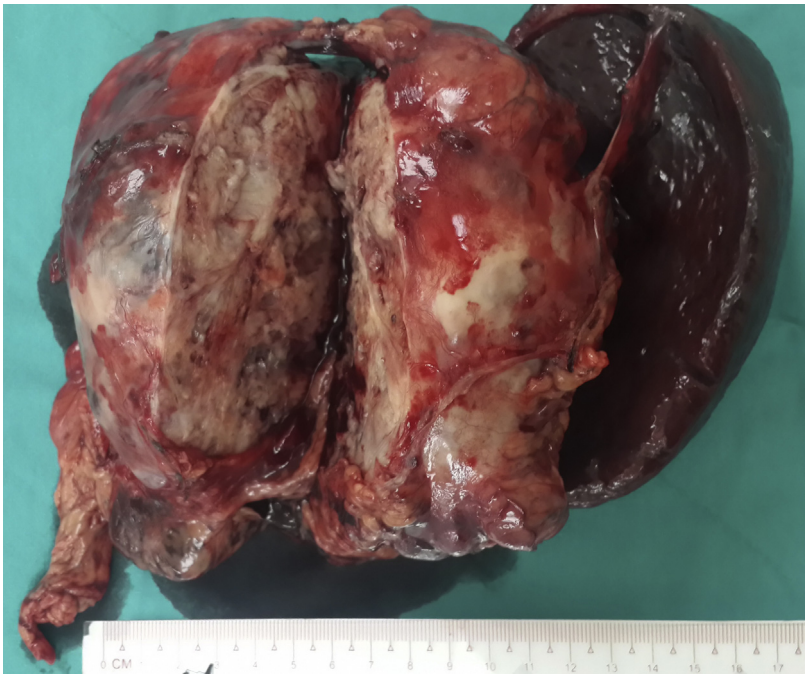

Fig. 2. Gross pathology.

focal hemorrhage. The mass was attached with the splenic capsule without splenic invasion (Fig. 2).

Histopathological examination revealed complete fibrous encapsulated mass composed of neoplastic cells arranging in cord, nest, insular and trabecular pattern (Fig. 3A–C). Immunological staining was positive for chromogranin, synaptophysin, insulin. Ki67 nuclear staining was positive at about 1–2%. Staining for Congo red, glucagon, and somatostatin was negative (Fig. 3D–H). The neoplastic cells had uniformed round nuclei with stippled nuclear chromatin and indistinct nucleoli without angiolymphatic or perineural invasion. Mitotic figures were 3–4/10 high power field (HPF). The tumor was confined in pancreas without splenic capsular invasion. All three resected lymph nodes showed no sign of metastasis.

Postoperative cause was uneventful. The latest follow-up of the patient (6 months after the procedure) revealed neither hypoglycemic, nor hyperglycemia. Post-splenectomy vaccination was performed.

2. Discussion

Insulinomas are rare pancreatic neuroendocrine tumors (PNET), which are mostly small at diagnosis. Giant insulinomas (>9 cm diameter) are even rarer. A giant insulinoma measuring 12.5 cm found in a 15-year-old man is very atypical. Our patient presented with typical Whipple triad. He reported only 2 month duration of symptoms before the diagnosis. High serum C-peptide and insulin level, in contrast with low plasma glucose level, confirmed the endogenous hyperinsulinemia. From these laboratory findings, the differential diagnosis was insulinoma, malignant insulinoma, ectopic insulin secreting tumor, and noninsulinoma pancreatogenous hypoglycemia syndrome (NIPHS). The mass palpable in this patient might represent intraabdominal insulin-secreting tumor such as insulinoma, and malignant insulinoma. In extremely rare cases, some malignancies were also previously reported to secrete insulin, such as gastrointestinal stromal tumor [7]. NIPHS could cause hypoglycemia from endogenous hyperinsulinism, but palpable mass in this patient could not explain this syndrome. Tumor was easily localized at pancreas by abdominal CT due to the large size. The CT result helped rule out NIPHS and made ectopic insulin secreting tumor unlikely. The diagnosis of insulinoma was confirmed by the positive staining for insulin while being negative for other hormones.

Diagnosing an insulinoma in youths as in our patient should raise the concern for genetic diseases. A case of giant insulinoma was previously reported to be associated with MEN-1 [8]. Yet, normal serum intact PTH, calcium and prolactin level helped exclude MEN-1 syndrome in this patient.

Surgical excision is the treatment of choice for insulinomas. In the small ones, simple enucleation or a distal pancreatectomy could be done. More aggressive surgical procedure is needed for larger tumors, requiring Whipple or total pancreatectomy