Gall bladder agenesis in Prader Willi syndrome

Sir,

We earlier described a 21-year-old morbidly obese man with Prader Willi syndrome (PWS)[1] who presented with poor glycemic control (HbA1c 14%) on large doses of multiple subcutaneous doses of insulin. He had unsatiable hunger. For benefit of hypothalamic action, subcutaneous liraglutide 0.6 mg once daily was added to his insulin and metformin regimen.

Two weeks after starting liraglutide, he developed severe abdominal pain and vomiting. Pain was severe, epigastric and radiating to the back. His family took him to a local physician where he was admitted. His serum amylase levels were found to be elevated more than 3 times the normal range of 360 SU (28–100 SU). There was no history of recent alcohol intake. He was initially managed locally and then referred to us. His liraglutide was stopped and he was managed conservatively for pancreatitis with bowel rest, intravenous fluids and analgesics. Patient had an uneventful recovery. He was started back on multiple subcutaneous insulin regimen and metformin. Two weeks later, his serum amylase was 99 SU (28–100 SU) and abdominal pain disappeared.

Ultrasound abdomen was unable to visualize the gallbladder. Head of pancreas was seen normally, body and tail were obscured by bowel gas.

Computed tomography scan abdomen showed features of acute pancreatitis involving body and tail region with extensive peripancreatic inflammation and left minimal pleural effusion [Figure 1]. Gall bladder fossa was empty.

Tc-99m Mebrofenin Hepatobiliary scan revealed patent biliary system with good hepatocyte radiotracer extraction with prompt excretion of tracer in the bowel loops. The gall bladder was not visualized [Figure 2]. Patient did not have any history of previous abdominal surgery either open or laparoscopic.

Gall bladder agenesis is rare[2,3] with only over 400 cases reported. Pancreatitis can occur in biliary stasis and gall bladder abnormalities.[4] PWS is associated with many other congenital anomalies. To the best of our knowledge, this is the first reported case of agenesis of gall bladder in PWS. Though a recent study showed no additional risk of pancreatitis with the use of Liraglutide, we did not restart liraglutide in this patient as pancreatitis following use of liraglutide has earlier been reported.[5]

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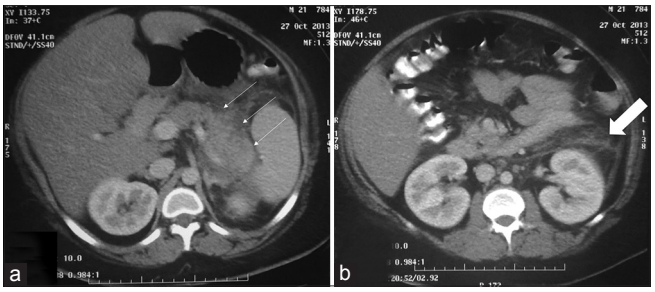

Figure 1: (a, b) Contrast enhanced CT scan images in axial plane (a, b) show bulky appearance of body and tail of pancreas (arrows) with extensive peripancreatic fat stranding and minimal fluid collection and inflammatory thickening of left side Gerota's fascia. There is no necrosis or calcification in the pancreas. The above findings are suggestive of acute pancreatitis

Figure 2: Tc-99m Mebrofenin Hepatobiliary study, anterior serial static images of the abdomen showing good hepatocyte radiotracer extraction with prompt excretion of tracer in the bowel loops visualized at 10 minutes, the gall bladder is not visualized during the study duration

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