A rare presentation of Y-duplication of the pancreatic tail with enteric duplication cyst as intussusception in a child: A case report

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ARTICLE INFO

Article history:
Received 4 November 2020
Received in revised form
24 November 2020
Accepted 25 November 2020
Available online 5 December 2020

Keywords:
– Pain
– Vomiting
– Obstruction
– Duplication

ABSTRACT

We report a case of a 2-year-old girl who presented to us with complaints of pain abdomen and non-bilious vomiting. USG was suggestive of intussusception with necrotic bowel and free fluid in the left lumbar region. Operative findings were of enteric duplication cyst of the pancreas which was later confirmed on histopathology. Enteric duplication cysts can occur anywhere in the gastrointestinal tract but duplication cyst within the pancreas is rare. The most common presentation in other reported cases is pain abdomen due to pancreatitis but in our case, we had findings of intussusception. Children usually present with a severe complication like perforation of duplication cyst as in our case. Excision of the cyst and the duplicated pancreatic tail is the mainstay of treatment.

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1. Introduction

Enteric duplication associated with pancreas is rare [1]. Bifid pancreas is a branching anomaly of pancreatic duct which present as Y-duplication of pancreatic tail. It is thought that bifid pancreas develops from bifid dorsal bud [2].

Most common presentations are abdominal pain, vomiting, pancreatitis, or incidental mass abdomen and rarely they present with peritonitis. Enteric duplication cyst within pancreas presenting as intussusception has never been reported in literature. Diagnosis of these cysts is difficult as they may be confused with pseudocyst of pancreas. Surgical excision is the treatment of choice. Here in line with the SCARE criteria [3,4]. We report a rare presentation of Y-duplication of pancreatic tail with enteric duplication cyst masquerading as intussusception.

2. Case report

A two-year-old girl presented with complaints of acute abdominal pain, non-bilious vomiting and decreased urine output for last 3 days. She was admitted in some local facility and was diagnosed with intussusception and then managed there conservatively and then referred to us. She had intermittent pain abdomen for last 6 months which were managed conservatively. There is no history of bilious vomiting, abdominal distension, constipation, blood in stool, fever or UTI. There was no significant past, family, psychological or pharmacological history. On examination, she was dehydrated and irritable. Abdomen was distended and tender. Lump was palpable in left lumbar region. It was soft, tender, ill-defined margins and immobile. Kidneys and bladder were not palpable. Fluid resuscitation was done and broad-spectrum antibiotics ceftriaxone, metronidazole and amikacin was started. USG abdomen revealed intussusception with necrotic bowel and fluid collection in the left lumbar region (Fig. 1). With a differential diagnosis of mesenteric cyst, pseudocyst of pancreas and duplication cyst CECT was done which revealed possibility of duplication cyst with perforation and phlegmonous collection within the mesentery (Fig. 1b). She underwent exploratory laparotomy by senior consultant and chairman of the department with surgical experience of 30 years and intra-operative findings revealed duplication cyst adjacent to splenic flexure with necrotic omentum on it. Tongue of pancreatic tissue going into the cyst. Cyst received its blood supply from left gastric artery and drained into splenic vein (Fig. 2a). Cyst was dissected from pancreas and pancreatic tissue going into the cyst was ligated without injuring the pancreas (Fig. 2b,c). Post-operative recovery was smooth. Histopathology revealed presence of pancreatic tissue and cyst wall had enteric mucosal lining and outer smooth muscle layer. USG abdomen after 1 week revealed no fluid in the bed. On follow after 6 months, she is doing well.

3. Discussion

Enteric duplication cyst can occur anywhere along the intestinal tract, of which 44% are found in small intestine [4]. Two widely accepted theory of enteric duplication is the errors of recanalization

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https://doi.org/10.1016/j.ijscs.2020.11.131
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by Brener and neurenteric band theory postulated by McLetcheis. In our case, the cyst derived its arterial supply from left gastric artery and drained into splenic vein. Review article reported by Deng et al. states about enteric duplication cyst associated with an accessory pancreatic lobe [5,6]. To date, 23 such cases have been reported. In our case it appears to be a bifid pancreatic tail rather than an accessory lobe and cyst was seen arising from one of the pancreatic tail. It seems there were repeated episodes of infections leading to perforation and further formation of phlegmon.

Most common presentation of duplication cyst is acute pain abdomen which is due to pancreatitis which is seen in 72% of cases. Our case had normal level of amylase and lipase and presented as intussusception which has not been reported in literature. Ultrason sound is the diagnostic modality where double wall or muscular rim sign is seen in cases of enteric duplication cyst [7]. But in our case due to thick cyst wall and fluid collection, it was diagnosed as intussusception with necrotic bowel. CECT abdomen can show the location, extent, complications and anatomic relation with the surrounding structures. Surgical excision is the treatment of choice and children usually don’t need any other management. As in our case, child is doing well on follow-up and had no similar complaints.
4. Conclusion

Here the purpose of reporting the case is a) Bifid pancreas presenting as Y-duplication of tail is seen in 0.9% cases of pancreatic anomaly and is generally an incidental finding, b) Enteric duplication cyst of Y-duplication is another rare entity other cases and it’s not the same as accessory lobe, c) Most common presentation of these cases is pancreatitis due to blockage of pancreatic duct which was not seen in our case, even though there is evidence of infections, d) Diagnostic dilemma on ultrasound reporting it as intussusception. The most important thing here is the basic knowledge which we should never forget. The clinical scenario didn’t match the radiological investigations and that lead to getting a CECT abdomen done and that clichéd the diagnosis.

Declaration of Competing Interest

No disclosures or conflicts of interest

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

The study is exempt from ethical approval in my institution.

Consent

Written informed consent was obtained from the parents of the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr Satish Kumar Aggarwal: writing the manuscript, study design, final review.

Dr Rupa Banerjee and Dr Gaurav Singh: data collector, writing the manuscript.

Registration of research studies

Not applicable.

Guarantor

Dr Satish Kumar Aggarwal is the guarantor and accepts full responsibility.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

[1] S. Andronikou, C. Sinclair-Smith, A.J. Millar, An enteric duplication cyst of the pancreas causing abdominal pain and pancreatitis in a child, Pediatr. Surg. Int. 18 (2002) 190–192.
[2] D. Dinter, J.M. Lohr, K.W. Neff, Bifid tail of the pancreas: benign bifurcation anomaly, AJR Am. J. Roentgenol. 189 (2007) 251–253.
[3] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus surgical CASE REPORT (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136.
[4] T.R.S. Prasad, C.E. Tan, Duodenal duplication cyst communicating with an aberrant pancreatic duct, Pediatr. Surg. Int. 21 (2005) 320–322.
[5] K.K. Christians, S. Pappas, P. Pilgrim, S. Tsai, E. Quebbeman, Duplicate pancreas meets a gastric duplication cyst: a tale of two anomalies, Int. J. Surg. Case Rep. 4 (2013) 735–738.
[6] Y. Deng, Z. Hu, W. Hao, G. Hu, Isolated retroperitoneal enteric duplication cyst associated with an accessory pancreatic lobe, Int. J. Clin. Exp. Pathol. 12 (2019) 3089–3095.
[7] S.A. Shabtai, J.C. Infante, G. Dantan, H.L. Neville, E.A. Perez, J.E. Sola, A.R. Hogan, Accessory pancreatic lobe in association with a gastric duplication cyst, J. Pediatr. Surg. 96 (2017) 7–10.