Internal Biliary Fistula Secondary to Cushing’s Ulcer in a Child

by Allison Ta, Suchitra Hourigan, Otto Louis-Jacques

INTRODUCTION

Internal biliary fistulas are abnormal communications between the extrahepatic biliary tree and another organ. These fistulas are rare disorders, typically affecting elderly patients as complications of biliary disease or, less commonly, peptic ulcer disease or cancer requiring surgical intervention. We present the case of a cholecystoduodenal fistula (CDF) secondary to a perforated duodenal ulcer in a pediatric patient with posterior fossa tumor. We discuss the success of medically treating this patient with proton pump inhibitors and subsequent resolution of the fistula.

Case Report

A 10 year-old female presented to the hospital with complaints of headache, vomiting, abnormal gait and upper extremity weakness for approximately eight months. She utilized ibuprofen or acetaminophen several times a week for her symptoms. Her past medical history was significant only for hypothyroidism.

A magnetic resonance imaging (MRI) of her brain showed a large posterior fossa tumor (measuring 8x4x3 cm). The patient was admitted to the pediatric intensive care unit (PICU) for stabilization and pre-operative planning. Dexamethasone was started on hospital day (HD) 1 and weaned post-operatively. On HD 4, she underwent surgical debulking of the tumor. On HD 8, she was started on nasoduodenal feeds and famotidine. The patient complained intermittently of epigastric pain, but otherwise had an uncomplicated initial postoperative course.

On HD 11, she became acutely hypotensive and her hemoglobin dropped from 10 to 5.1g/dL. Nasogastric (NG) tube lavage revealed brown colored fluid. She was resuscitated with packed red blood cells and fluids. Urgent esophagogastroduodenoscopy (EGD) discovered a large, non-bleeding duodenal ulcer. An orifice was seen medially within the ulcer bed (Figure 1). Biopsies taken at the time of EGD confirmed ulceration and ruled out Helicobacter infection. An abdominal computed tomography (CT) scan revealed air and contrast in a normal-appearing gallbladder, with a small communication between gallbladder and duodenum, consistent with a cholecystoduodenal fistula (Figure 2). Pediatric surgery was consulted and recommended medical management.

The patient was started on intravenous pantoprazole at 120 mg daily (roughly 1.6mg/kg/day), total parenteral nutrition and a NG tube was placed for gastric decompression. An EGD was repeated on hospital day 20 due to persistent melena. It showed the fistula surrounded by ulcerated tissue with a small amount of blood and bile flowing out of fistula. Attempts to place a clip on the edges of the ulcer bed were unsuccessful. Her proton pump inhibitor (PPI) was switched to continuous drip after which the bleeding resolved. Repeat EGD and abdominal CT on HD 36 showed a healed ulcer and closure of the fistula. The patient was transitioned to oral PPI and NG feeds were resumed. She had no further complications before transfer to a rehabilitation facility. At clinic follow-up more than three months later, there was no recurrence of bleeding.
Patients with smaller choledochoduodenal fistulas (orifice <0.5 cm) can be successfully treated medically, however surgical intervention may be necessary in case of complications from the ulcer, including uncontrolled bleeding or perforation.

Given the lower incidence of gallstones and the rarity of peptic ulcer disease in children, it is not surprising that there are very few reports of IBF in children. In one case, a 6 year-old child presented with recurrent cholangitis. MRCP revealed a choledochoduodenal fistula for which she underwent cholecystectomy, common bile duct excision and Roux-en-Y hepaticojejunostomy. The only previous report of a child with a choledochoduodenal fistula secondary to peptic ulcer disease is a 10 year-old male with a three month history of abdominal pain and non-bilious vomiting. An EGD revealed a scarred ulcer in first part of duodenum causing obstruction which was treated surgically.

This is, to our knowledge, the first report of a pediatric patient with a CDF caused by a penetrating duodenal ulcer who was successfully treated medically. Our patient had multiple risk factors that predisposed her to developing an ulcer and subsequent bleeding including prolonged use of non-steroidal anti-inflammatory drugs (NSAIDs), the peri-operative use of systemic steroids and the stress of PICU hospitalization all in the setting of a posterior fossa tumor. An important aspect of our patient’s case is the development of a
Internal Biliary Fistula Secondary to Cushing’s Ulcer in a Child

A CASE REPORT

peptic ulcer in the setting of a posterior fossa tumor which has been described as a Cushing’s ulcer. Harvey Cushing’s initial work in 1932 described an association between intracranial tumors or injury and the development of ulceration in the upper gastrointestinal tract. He theorized that these intracranial masses caused stimulation of the vagal nerve leading to increased gastrin predisposing patients to develop an ulcer in the esophagus, stomach or duodenum. This suggests prophylactic acid suppressant medication may be necessary.7,8

CONCLUSION

This case highlights the successful use of acid suppressants to heal a CDF caused by a perforated duodenal Cushing’s ulcer, a complication rarely seen in children. In pediatrics, some patients are predisposed to develop peptic ulcer disease (NSAID use, steroids, posterior fossa tumors, to name a few) and should have prophylactic acid suppressant started. Hemorrhage from an ulcer can have high morbidity and aggressive treatment, including the use of continuous PPI infusion, should be initiated until bleeding is controlled.

References


Answers to this month’s crossword puzzle:

1. Tubular
2. Iliac
3. Adenoma
4. Derived
5. Kidneys
6. Eilean
7. Sacred
8. Pair
9. Ill
10. Spike
11. Tuba
12. Cactus
13. Burning
14. Poylops
15. Copper
16. Biuret
17. Side
18. Cie
19. Itch
20. Ions
21. Used
22. Nor
23. Eau
24. Aicy
25. Iron
26. Inflamed
27. Hatly
28. Up
29. Genesis
30. Salves
31. Tuba
32. Iliac
33. Sacred
34. Pair
35. Ill
36. Spike
37. Tuba
38. Cactus
39. Burning
40. Poylops
41. Copper
42. Biuret
43. Side
44. Cie
45. Itch
46. Ions
47. Used
48. Nor
49. Eau
50. Aicy
51. Iron
52. Inflamed
53. Hatly
54. Up
55. Genesis
56. Salves
57. Tuba
58. Iliac
59. Sacred
60. Pair
61. Ill
62. Spike
63. Tuba
64. Cactus
65. Burning
66. Poylops
67. Copper
68. Biuret
69. Side
70. Cie
71. Itch
72. Ions
73. Used
74. Nor
75. Eau
76. Aicy
77. Iron
78. Inflamed
79. Hatly
80. Up
81. Genesis
82. Salves