Nigerian Journal of Paediatrics 1985; **12 (3): 101–104**

Intra-jejunal Stone Formation associated with Congenital Jejunal Stenosis

SE EFEM† AND AA ASINDI*

Summary

Efem SE and Asindi AA. Intra-jejunal Stone Formation associated with Congenital Jejunal Stenosis. *Nigerian Journal of Paediatrics* **1985; 12:101.** A case of extrabiliary stone formation in a $2\frac{1}{2}$-year old child with congenital jejunal stenosis, is reported. Our patient had no gall-bladder disease or any haemolytic disorder. We believe that the stones resulted from stasis of bile in a loop of jejunum proximal to the site of partial intestinal obstruction. A review of literature revealed that this is probably the first case of extrabiliary stone formation in a Nigerian child and possibly, the first to be reported anywhere in the world.

Introduction

GALLSTONE formation usually takes place within the gall-bladder; it commonly follows an inflammation of the latter. Such stones may migrate into the common bile duct.[1] Their appearance in the small intestine is usually by way of a cholecystoduodenal fistula with subsequent impaction in the terminal ileum to produce a "gallstone ileus".[1] The presence of gallstone within the lumen of the small intestine in the absence of any fistula or gall-bladder disease calls for explanation. The present communication concerns a child with a partial gastro-intestinal obstruction and numerous stones in the upper intestine, without evidence of gall-bladder disease or any fistulous connection with the intestine.

College of Medical Sciences, University of Calabar, Calabar

Department of Surgery

† Lecturer

Department of Paediatrics

* Senior Lecturer

Correspondence: Dr SE Efem

Case Report

A $2\frac{1}{2}$ - year old Nigerian boy was seen at the University of Calabar Teaching Hospital (UCTH), Calabar, with a history of recurrent abdominal pains and progressive abdominal distension of $2\frac{1}{2}$ years duration. The child was born full-term after an uneventful pregnancy. At birth, the abdomen was noticed to be mildly distended and this had progressively increased since. He passed normal meconium at birth. There was no history of vomiting or constipation. No visible peristalsis had been observed by the parents and the patient received no treatment of any sort before presentation at the hospital. None of the parents or the other siblings had similar illness. The family history was negative for gall-stones. Examination revealed a well-developed

and well-looking child, 90cm tall and weighing 12kg. No abnormality was detected either in the respiratory or cardiovascular system. The abdomen was grossly distended with a gap between the two recti abdominis.

There were neither visible peristalsis nor distended superficial veins. The abdomen was not tender on palpation. The liver and spleen were palpable. The epigastrium was tympanitic to percussion, while the rest of the abdomen showed a positive shifting dullness. The bowel sounds were slightly hyperactive. The clinical diagnosis at this stage, was partial intestinal obstruction.

Investigations

The haemoglobin was 9.5g/100ml (9.5g/dl). A plain abdominal radiograph showed evidence of high small intestinal obstruction and calcifications which were thought to be radio-opaque stones in the distended loops of small bowel (Fig.1). Other investigations including serum electrolytes and urea, liver function tests and serum proteins, were normal. The haemoglobin electrophoresis revealed a genotype of AA.

Fig. 1 A plain radiograph of the abdomen showing stones in the distended loops of bowel.

Laparotomy

At laparotomy, the following was found: the duodenum and jejunum were grossly distended; there was approximately 500ml of free straw-coloured fluid in the peritoneal cavity; there were two fibromuscular bands plastering the terminal jejunum to the posterior abdominal wall causing obstruction and massive hypertrophy and dilation of the duodenum and jejunum (Fig. 2). Seventy-eight discrete stones and bile were found in the distended loops of the duodenum and jejunum (Fig. 3). The gall bladder and biliary tree were normal. There was no evidence of fistulous communication between the gall bladder or biliary tree with the intestines. There was a very narrow communication between the jejunum and the proximal ileum. The stenosed segment of jejunum was excised and an end-to-end ileo-jejunal anastomosis carried out, after evacuating the bile and milking out all the stones.

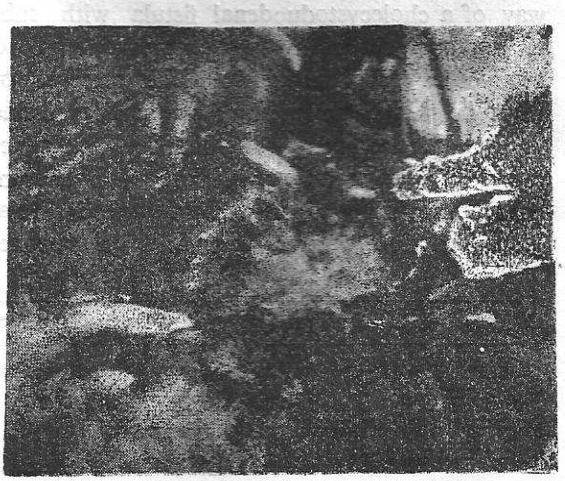

Fig. 2 Grossly dilated and hypertrophied duodenum and jejunum proximal to the obstruction at the jejunal end.

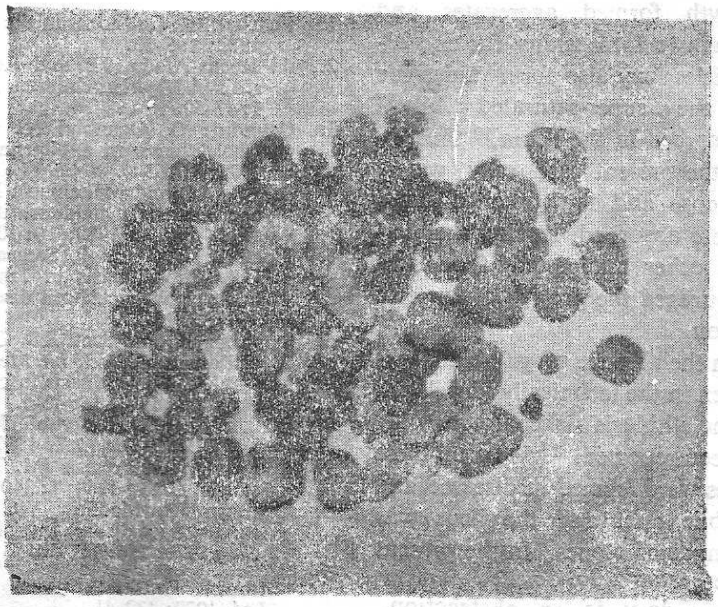

Fig. 3 Seventy-eight stones obtained from dilated jejunum and duodenum.

The patient made a remarkable post-operative recovery and was discharged on the 12th post-operative day. He has since been seen three times in the 'follow-up' clinic without any complaint. A chemical analysis of the stones showed them to be mixed stones consisting of calcium bilirubinate, calcium carbonate and cholesterol.

Discussion

Cholelithiasis is extremely rare among Nigerian children. In 1971, da Rocha-Afodu and Adesola[2] reviewed cases of cholelithiasis in Nigeria and found none among children, but a single symptomatic case has been reported in a child by Ogundipe, Ogunbiyi and Ojemuyiwa.[3] Using plain abdominal radiograph, oral cholecystography and real time ultrasonography, Adekile and Makanjuola[4] reported only 2 children (4.4%) with gallstones among 45 asymptomatic children who had sickle-cell anaemia. Thus, the prediction by some workers[1 5] that 25% of patients with homozygous sickle-cell disease in black tropical countries will develop pigment stones does not appear to hold in Nigeria. The high incidence of gallstones reported in American patients with homozygous sickle-cell disease is multifactorial and not simply dependent on red cell sequestration.[6–11]

Our patient is a unique case for several reasons. His haemoglobin genotype was AA. The gallbladder was completely normal; the stones were formed *in situ* in the jejunum in relation to a congenital stenosis of this part of the gut. This is therefore, a classical case of stasis predisposing to stone formation. To our knowledge, this is the first report of extrabiliary stone formation in a Nigerian child.

The pathogenesis of the stones in the present case appears to be as follows: the congenital band stretching across the distal jejunum gave rise to a partial obstruction that caused the hypertrophy and dilatation of the jejunum proxima

to the stenosis with accumulation of bile therein. Bacterial overgrowth formed aggregates and particles large enough to form nidus for crystallisation. Food particles may also form nidus for crystallisation of the super-saturated bile.[12] Bile is normally absorbed in the ileum,[12] thus, in the duodenum of the present case, only water, calcium, iron, glucose and few other digested food substances were absorbed, leaving a super-saturated bile and other undigested food substances which encouraged the growth of bacteria and stone formation.

Gallstone ileus in the absence of gall-bladder is extremely rare. Even in the technically developed countries where cholelithiasis is common, only four cases have been reported. [13–16] In all these cases, the gallstones were formed in diseased and dilated common bile duct and the ileus was secondary to the impacted stones. In the present cases however, the stones were formed *in-situ* in the jejunum secondary to an obstruction. This is probably the first case in the medical literature of bilirubin containing stones formed outside the biliary system (Hobsley 1983, personal communication).

Acknowledgements

We wish to thank Professors TF Solanke and Michael Hobsley for their useful criticism of this paper and the Department of Chemical Pathology, University of Ibadan, for analysing the stones.

References

1. Fletcher PR. Gallstones in the Tropics. *Med Digest* 1982; **8:** 5–13.
2. da Rocha-Afodu JT and Adesola AO. Cholecystitis in Nigerian children. *J Nig Med Ass* 1971; **1:** 47–50.
3. Ogundipe O, Ogunbiyi AJ and Ojemuyiwa M. Cholelithiasis in a Nigerian child. *Nig Med J* 1978; **5:** 163–4.
4. Adekile AD and Makanjuola D. The prevalence of cholelithiasis in children with sickle-cell anaemia. *Nig J Paediat* 1983; **10:** 109–17.
5. MacCall IW, Desai P, Sergeant BE and Sergeant GR. Cholelithiasis in Jamaican patients with homozygous sickle-cell disease. *Am J Haematol* 1977; **3:** 15–21.
6. Stephens OG and Scott RB. Cholelithiasis in sickle cell anaemia. *Arch Int Med* 1980; **140:** 648–51.
7. Mossa AR. Cholelithiasis in childhood. *J Roy Coll Surg Edin* 1973; **18:** 42–6.
8. Walker CM. Aetiology of cholelithiasis in childhood. *Arch Dis Child* 1957; **32:** 293–7.
9. Bouder IA and Preston JM. The aetiology of gallstones. *Lancet* 1968; **1:** 340–4.
10. Cameron JC, Maddrey WC and Zuidema GD. Biliary tract disease in sickle cell anaemia. *Ann Surg* 1971; **174:** 702–10.
11. Edington GM and Gilles HM. Pathology in the tropics. London: Edward Arnold (Publishers) Ltd, 1979: 432–41.
12. Rains AJH and Ritchie HD. Gallstones. In: Baily and Love's Short Practice of Surgery. Rains AJH and Ritchie HD, eds. London: HK Lewis & Co Ltd, 1977: 886–91.
13. Dulin JM and Peterson RF. Intestinal obstruction due to gallstones. *Arch Surg* 1939; **38:** 351–7.
14. Warner G and Swan H. Gallstone obstruction of the small bowel in the absence of gall bladder. *Arch Surg* 1975; **30:** 865–8.
15. Lundsay TM, Katz-Unger A and Strickland WM. Gallstone ileus in the absence of gall-bladder. *Arch Surg* 1975; **110:** 448.
16. Panapizir K and Sykes PA. Gallstone ileus in the absence of gall-bladder. *J Roy Coll Surg Edin* 1982; **27:** 182–3.

Accepted 26 April 1985