CASE REPORTS

Giant gas-filled cyst of sigmoid colon Report of a case and review of the literature

D. E. SIBSON
M.B., F.R.C.S.

A. J. EDWARDS
M.Sc., M.Chir., F.R.C.S.

Department of Surgery, Hackney Hospital, London, E.9

Summary

The largest recorded gas-filled cyst of the sigmoid colon, in a man of 67, is presented. The literature of the other recorded cases is reviewed. The gas in the cyst was analysed and a suggestion put forward that bacteria play some part in the aetiology of the gas in the cyst.

Introduction

A giant gas-filled cyst of the abdomen is a rare occurrence there being only fifteen cases previously recorded in the literature. Plain X-ray of the abdomen makes the diagnosis and by this method the recorded sizes have varied from 8 to 25 cm. The presence of muscle in the wall of two of the smallest cysts implies that they arise from elements of reduplication of the gut (Hughes & Greene, 1953; Melamed & Pantone, 1960). Of the remainder, nine with histological reports do not show this feature and, arising in conjunction with diverticular disease of the colon, are thought to take origin from a single diverticulum. The origin of the gas in the cysts has been held to be intraluminal colonic gas entrapped by valvular communication (Case records of the Massachusetts General Hospital, Case 43402, 1957) although diffusion with blood gases in the cyst wall may modify the constituents. This case report describes the largest cyst recorded and includes chemical analysis of the entrapped gas. Since no similar data have been found in the literature some evidence is provided as to the origin of the cyst gas.

Case report

A 67-year-old man was admitted as an emergency on 2 October 1970, complaining of abdominal distension and discomfort. He had had recurrent distension of the abdomen for 3 years which had

become constant over the past 6 months, although his abdomen still varied in size. His discomfort was related to the distension, but he had no severe pain. He had a marked tendency to constipation but no rectal bleeding. He had been a chronic bronchitic for 12 years.

On examination. He was thin, pale and slightly dyspnoeic at rest. He was afebrile. There were rhonchi and rales over both lung fields. His abdomen was grossly distended and tympanitic but there was no localized tenderness and bowel sounds and rectal examination were normal.

Investigations. Haemoglobin 13.1 g%, WCC 8400/mm³. Blood urea 41 mg/100 ml. Electrolytes normal. Chest X-ray showed signs of chronic bronchitis and emphysema. Plain X-ray of the abdomen revealed a gas-filled cyst measuring 27 cm (Fig. 1) and barium enema demonstrated moderate diverticular disease of the sigmoid colon which appeared stretched over the cyst (Fig. 2). No barium appeared to enter the cyst cavity.

Treatment. Pre-operatively his chest condition was treated with physiotherapy and because of his dehydration intravenous fluid therapy was necessary. Bowel preparation was carried out with phthalylsulphathiazole and laparotomy performed. Plain X-ray on the day before operation showed the cyst to be smaller (16 cm) and his abdomen was less distended, although there is no evidence that this was the result of therapy.

Operation. (19 October 1970) Through a long mid-line incision a large gas-filled cyst was found arising from a juxtamesenteric diverticulum of the sigmoid colon. This cyst had burrowed laterally and extraperitoneally and then extended forwards across most of the abdominal cavity stripping the parietal peritoneum from the left iliac fossa and anterior abdominal wall (Fig. 3). There were inflammatory adhesions between the cyst and the surrounding

Requests for reprints: Mr D. E. Sibson, Surgical Professorial Unit, St Bartholomew's Hospital, London, E.C.1.

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Table of recorded cases

Author(s)	Sex	Age	Presenting symptoms	Signs	X-ray	Ba enema	Site	Size	Culture of contents	Muscle in wall
Bonvin & Bonte (1946)	M	60	Pain in LIF	Swelling in LIF	Gas-filled cyst	Diverticular disease. Cyst not filled	—	—	—	—
Hughes & Greene (1953)	F	45	2/52 abdominal pain and mass	Mobile mass in RUQ	Gas-filled cyst	Not done	Anti-mesenteric	10 cm	—	Yes
Boijesen (1956)	M	70	Several months lower abdominal discomfort	Mobile mass in RIF	—	Diverticular disease. Cyst not filled	—	15 cm	—	No
Brown (Case 43402) (1957)	M	57	2/52 dull abdominal pain. Mobile mass in abdomen	Smooth, firm resonant fixed mass in RUQ and epigastrium	Gas-filled cyst with small fluid level	Not done	Anti-mesenteric	25 cm	—	No
Frankenfeld <i>et al.</i> (1958)	M	74	1/52 painful abdominal mass. Diminished and recurrent	Tender mass	Gas-filled cyst	Not done	Anti-mesenteric	10 cm	—	No
Dominguez-Rodino & Dominguez-Rodino (1959)	M	64	Recurrent pain and jaundice. Loose motions	None	Gas-filled cyst	Diverticular disease with cyst filled	—	—	—	—
Melamed & Pantone (1960)	M	67	1 year rectal bleeding	None	—	Cyst filled with barium: emptied on evacuation	—	8 cm	—	Yes
Silberman & Thorner (1961)	M	75	6/12 intermittent diarrhoea. Acute abdominal pain	Tender fixed mass in LIF	Gas-filled cyst	Previously diverticular disease with large filled diverticulum	—	15 cm (torsion)	—	No
Marana <i>et al.</i> (1964)	F	58	5/12 abdominal pain and vomiting with increasing mass	Mobile, soft tympanitic mass	Gas-filled cyst	Not done	Anti-mesenteric	20 cm	—	No
Moore & Gold (1964)	F	67	3/52 pain in LIF anorexia and nausea	Large central abdominal mass	Gas-filled cyst	Not done	Anti-mesenteric	10 cm	<i>E. coli</i>	No
Cautiero & Papa (1966)	F	72	Recurrent abdominal pain	Mass in LIF	Gas-filled cyst	Diverticular disease. Cyst not filled	—	20 cm	—	—
Fontaine <i>et al.</i> (1966)	M	42	6/12 mobile abdominal mass	Mass in LIF	Gas-filled cyst	Not done	Anti-mesenteric	10 cm	—	No
Smaltino <i>et al.</i> (1967)	M	44	Severe abdominal pain and vomiting	Indefinite mass in LIF	—	Diverticular disease. Cyst not filled	—	—	—	No
Beauchant <i>et al.</i> (1968)	M	67	15 days abdominal swelling and pain	Resonant round mass	Gas-filled cyst	—	Anti-mesenteric	14 cm	—	—
Asch <i>et al.</i> (1970)	F	44	2/12 enlarging painless abdominal mass	Mobile, non-tender cystic mass	Gas-filled cyst	Diverticular disease. Cyst not filled	Anti-mesenteric	21 cm	Sterile	No
Silbershous & Edwards (1972)	M	69	3 years recurrent abdominal distension with discomfort	Grossly distended tympanitic abdomen	Gas-filled cyst	Diverticular disease. Cyst not filled	Mesenteric	27 cm	<i>E. coli</i> <i>Proteus vulgaris</i>	No

FIG. 1. First plain X-ray of the abdomen showing 27 cm gas-filled cyst.

FIG. 2. Barium enema showing sigmoid colon with diverticular disease stretched over the cyst.

bowel mesentery and omentum. The sigmoid colon showed moderate diverticular disease. The remainder of the abdominal contents was normal. The cyst was mobilized and two 500-ml gas samples taken by water displacement for analysis. A sigmoid colectomy was carried out removing the cyst still attached to the colon, but because the proximal colon still contained barium from 10 days previously, a double-

barrelled colostomy was fashioned in the left iliac fossa. The wound was closed in layers with an intra-peritoneal drain.

FIG. 3. View of cyst (C) from left side showing ascending colon (A) and sigmoid colon (S) not yet mobilized.

Postoperative course. A postoperative chest infection responded slowly but satisfactorily to ampicillin and physiotherapy but on the sixth day he suddenly collapsed and died. Necropsy showed acute left ventricular failure with pulmonary oedema, chronic bronchitis, emphysema and a small pulmonary infarct.

Specimen. The cyst measured 18 cm in diameter and contained a small amount of purulent fluid, culture of which showed a heavy mixed growth of *E. coli* and *Proteus vulgaris*, with some coagulase-negative staphylococci. The wall of the cyst was rough, measured about 3 mm and had no obvious mucosa, and an oblique connection could be demonstrated between the inside of the cyst and a diverticulum of the sigmoid colon.

Microscopical examination. The wall was composed of fibrous tissue with marked inflammatory cell infiltration. The lining was entirely of inflammatory exudate with no mucosa and there was no muscle in the wall.

Gas analysis. Oxygen, 1.2%; nitrogen, 74.4%; carbon dioxide, 13.7%; nitrous oxide, 7.7%; water vapour, 3.0%; traces of unidentified sulphur compounds; no methane detected.

Discussion

Of the sixteen cases now recorded ten are men and six women with an age range of 42–75 years and an average of 61. The reported symptomatology is very variable. Eight patients had noticed a discrete swelling, five of which were painful. Three further patients presented with left iliac fossa pain and one with rectal bleeding. The duration of symptoms varied from days to years. Thirteen out of sixteen patients

had a discrete mass palpable in the abdomen, two of which are recorded as being resonant. Only in two of them were there no abdominal physical signs. The present case would appear to be the only one to exhibit abdominal distension to such a degree that no local mass was detectable. We consider the hyper-resonant distension, in the presence of normal bowel sounds, to be an important diagnostic feature and think that the only useful diagnostic investigations are plain X-ray of the abdomen and barium enema. There is enormous variation in the size of these radiolucent cysts on plain film from 6 cm in the case of Bonvin & Bonte (1946) to 27 cm in the present case, but clearly from the two X-rays taken in this case alteration in the size of a cyst from time to time is possible and may account for the variation in symptomatology described by Frankenfeld, Waters & Schepeler (1959). Barium enema whenever carried out, has always shown diverticular disease of the sigmoid colon but in only three cases could the cyst be filled with barium (Dominguez-Rodino & Dominguez-Rodino, 1959; Melamed & Pantone, 1960; Silberman & Thorner, 1961). The radiological findings are characteristic, but possible confusion arises with gas-filled abscess cavities arising from perforated diverticula as reported by Macbeth & Riddle (1964), and cystitis emphysematosa (Faingold, Hansen & Rigler, 1953). Other conditions showing gas-filled cavities, namely emphysematous cholecystitis and Meckel's diverticulum are not closely similar, having their own clinical and X-ray characteristics, and should not present difficulty (Retterbush *et al.*, 1951; Bischoff & Stampfli, 1955).

In all the previous cases where recorded, the cyst arose from the antimesenteric border of the large bowel. The present case was unusual in that it arose from a diverticulum near the pelvic mesocolon and had burrowed extraperitoneally extending anteriorly and medially.

It is generally accepted that these cysts are inflated diverticula of the colon. It may be that in their genesis there is a small subserosal leak which, becoming loculated, gradually distends and is maintained by a ball valve action at the cyst diverticulum junction. However, in two cases (Hughes & Greene, 1953; Melamed & Pantone, 1960) there was evidence of some mucosal lining and of musculature in the wall of the cyst, suggesting that these might be due to a reduplication of the bowel; these cysts were no greater than 10 cm and may well represent a different entity.

There is no previous report on the nature of the gas in these cysts, although its origin has been assumed to be from the bowel lumen. The possibility exists that fermentation by bacteria may at least play some part in the production or character of this gas, and any entrapped gas will be subjected to

diffusion effects with the blood gases in the cyst wall. Assuming that gas originated from the gut lumen the low oxygen and relatively high carbon dioxide concentrations can be explained on the basis of metabolism by the bacteria present in a walled off cavity with only poor diffusion into the circulation at an inflammatory surface. The organisms grown on culture in this case, *E. coli* and *Proteus vulgaris*, both metabolize carbohydrate to produce carbon dioxide. There are only two reports of culture of gas cyst contents. Moore & Gold (1964) obtained a pure culture of *E. coli* whilst Asch, Milikow & Gump (1970) could obtain no bacterial growth. The concentration of nitrous oxide supports the contention that, in the anaesthetized patient, the wall of the cyst is only slowly permeable to diffusible gases. It is suggested therefore that, whereas some of the gas undoubtedly comes from the bowel as previously suggested via a ball valve mechanism, at least some of it is produced in the cyst by the organisms and both these components are modified by some degree of diffusion with blood gases.

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Primary pneumococcal peritonitis in the nephrotic syndrome

RAMESH KUMAR
M.D., M.R.C.P.

M. G. McGEOWN
M.D., Ph.D., F.R.C.P.(Ed.)

J. McEVOY
M.D., M.R.C.P.

Renal Unit, Belfast City Hospital, Belfast, BT9 7AB

PRIMARY pneumococcal peritonitis appears to have been a frequently seen presenting feature of the nephrotic syndrome in children prior to the advent of antimicrobial therapy (Barnett & Shibuya, 1954; Fowler, 1957).

The present report is of a case of primary pneumococcal peritonitis in a boy who was thought to have acute appendicitis and underwent a laparotomy.

Case report

An 11-year-old boy presented with the complaints of vomiting and abdominal pain of 2 days' duration. Prior to the onset of these symptoms, his parents noticed some puffiness of the face and found the boy rather listless and tired. First, he began vomiting frequently and complained of pain in his abdomen. This pain was constant and aggravated by movement. There was no preceding history of sore throat, pyoderma or chest infection. He had no haematuria, arthralgia, headaches or convulsions. On admission, he had not passed urine for about 18 hr.

On clinical examination, his temperature was 101°F, his eyes were puffy and had slight pretibial oedema. He looked toxic and had an erythematous rash on his neck and upper part of the chest and a few erythematous spots on his buttocks. His heart was regular at 140/min; and blood pressure was 125/70 mmHg. The examination of the abdomen revealed generalized tenderness, most marked in the right iliac fossa with muscle guarding and rebound tenderness.

Correspondence: Dr Ramesh Kumar, Registrar, Renal Unit, Belfast City Hospital, Belfast, BT9 7AB.

His bladder was palpable midway between the symphysis pubis and the umbilicus, and on catheterization the urine showed marked proteinuria. His chest was clear and there was no neurological abnormality. A diagnosis of acute peritonitis with nephrotic syndrome was made, but with localized tenderness in the right iliac fossa and muscle guarding, acute appendicitis with perforation could not be excluded.

On the day of admission a laparotomy was performed through a long right paramedian incision and 2 pints of thin, milky fluid was aspirated and the pelvic sediment of the peritoneal fluid showed marked fibrinous streaking. His appendix and other organs in the abdominal cavity were normal except for marked enlargement of mesenteric, para-aortic and iliac lymph nodes. The peritoneal cavity was cleaned out and a biopsy of a mesenteric gland was taken, which on histological examination showed 'reactive hyperplasia'.

His haemoglobin was 14.1 g/100 ml; WBC 18,200/mm³ with 80% polymorphonuclear cells and an ESR of 85 mm/hr. Urinalysis showed specific gravity 1030; protein + + +, a few erythrocytes, and granular casts, culture was sterile. His blood urea was 43 mg/100 ml and plasma electrolytes were normal. Both peritoneal fluid and a swab from the peritoneal cavity grew pneumococci profusely on culture. Blood cultures were sterile. His throat swab did not grow streptococci and the anti-streptolysin 0 titre was 160 units/ml. The plasma proteins were total 4.1 g/100 ml, albumin 1.2 g/100 ml and on electrophoresis there was increase in alpha₂-globulin