Eur J Vasc Endovasc Surg **11**, 429–431 (1996)

The Primary Aortoenteric Fistula in The Netherlands — the Unpublished Cases

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Objective: *Primary aortoenteric fistula is a rare disorder of which only four patients have been reported in the Dutch literature so far. The objective of our study was to obtain more realistic figures on the incidence of this condition, with data on the clinical presentation, diagnostic procedures, treatment and results in a group of patients not previously reported as "case histories".*
Methods: *A questionnaire was sent to all surgical clinics in The Netherlands. Out of 180 questionnaires, 102 have been returned reporting 27 patients to which data of eight others treated in our own institution were added.*
Results: *In all but one of these 29 patients the fistula was caused by an atherosclerotic aneurysm, the one exception being caused by an ingested cocktail pin. Gastrointestinal haemorrhage was the predominant symptom, being present in 28 of the patients, while the complete triad of haemorrhage, pain and a pulsating mass was found in only eight patients. Twenty-seven patients were treated with an* in situ *graft of which 14 are doing well at long term follow-up.*
Conclusions: *Primary aortoenteric fistula is far more common than one would expect from the number of patients reported in literature. A high index of suspicion based on a complete physical examination remains the key to a correct diagnosis. Direct closure of the intestine and* in situ *grafting of the aorta is the treatment of first choice.*

Key Words: Gastrointestinal haemorrhage; Aortic aneurysm; Aortoduodenal fistula; Aortoenteric fistula; Primary aortoenteric fistula.

Introduction

The primary aortoenteral fistula (PAEF), defined as a communication between the native aorta and the intestinal tract, is a rare cause of gastrointestinal haemorrhage. So far, only four cases have been published in The Netherlands.[1–4] World wide 54 patients have been reported in the past 10 years.[5] One might therefore conclude that PAEF occurs once every 10 years in a 12–16 million population, and that the outcome is generally favourable. Neither of these conclusions seems very likely. Being curious to know a more likely estimate of the incidence, the clinical presentation, diagnostic work up, treatment and outcome, we have conducted a survey among the Dutch surgeons.

A questionnaire was sent to all 180 surgical departments in The Netherlands. Among the 102 replies, 68 surgeons reported never to have encountered a patient with PAEF, six misunderstood the questionnaire and reported one or more patients with a secondary fistula. Two surgeons informed us that they had treated three or "several" of these patients but did not supply specific information. Complete data on 27 patients were returned by 26 surgeons. Data on eight cases treated in our own institution are included, but will be reported more in detail separately.[5]

Patients

Data on 35 patients were available for evaluation. The group consisted of 29 men, five women and one patient of unidentified gender, ranging in age from 30–82 years (median 68 years). In all but one of these patients the cause of the fistula was an atherosclerotic aneurysm. The one patient without an aneurysm was 30-year-old mentally retarded man who had swallowed a cocktail pin, that subsequently perforated his duodenal wall.

Gastrointestinal haemorrhage, either haematemesis, melaena or both was the predominant symptom,

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1078–5884/96/040429 + 03 $12.00/0

being present in 28 patients. Fifteen patients complained of pain, either in the loin or in the abdomen. In 19 patients a pulsatile mass was noted to be present on physical examination. Fever or a febrile episode, with or without an increased leucocyte count and sedimentation rate was not included in the questionnaire. Nevertheless it was reported in four patients, and might well be a feature of the syndrome. The complete triad of pain, haemorrhage and a pulsatile mass was present in eight patients.

In 22 patients a (presumptive) diagnosis of PAEF was made preoperatively. On eight occasions the fistula was found as a surprise during laparotomy, while two were first identified at post-mortem examination, and twice the diagnosis was made only after a previous gastrectomy. One patient was not operated because of additional disease with a poor prognosis. Nine patients were operated within 24 h of their initial haemorrhage, in 14 a delay between 1 and 7 days occurred and four were operated more than a week after the onset of their complaints. In the others, the exact time lapse was not documented.

Endoscopy was the most common diagnostic procedure performed, contributing to the diagnosis in 3/17 patients. Two fistulae were positively identified by CT-scan, with characteristic air bubbles around an aneurysm (Fig. 1). Strikingly, a CT-scan was performed in 10 of 17 patients treated in the last 2 years, having been used only once before '91. Ultrasound examination and angiography contributed in identifying or confirming the presence of an aneurysm in several patients, without positively identifying a fistula in any of them. In four patients a laparotomy was performed without additional diagnostic procedures.

As the enquiry was directed to general and vascular, but not to thoracic surgeons, all but two fistulae were located between the abdominal aorta and duodenum or the first part of the jejunum. Twenty-seven patients were treated by in situ grafting, of which eight were covered with omentum, while two prostheses were soaked in Rifampicin. Three patients were primarily treated with an extra-anatomic graft, while aneurysmoraphy was performed in two. Three patients have not been operated. Nine patients expired during or shortly after surgery. Three patients died without having been operated, and one patient died 2 months after extra-anatomic grafting, due to an aortic stump blow out. One of the patients with an *in situ* graft developed a secondary fistula within 2 weeks of the first operation and died during secondary extra-anatomic grafting. Three other patients developed a secondary fistula between 2 months and 6 years after the primary operation. All three eventually died, either from sepsis or aortic stump blow out after secondary extra anatomic grafting.

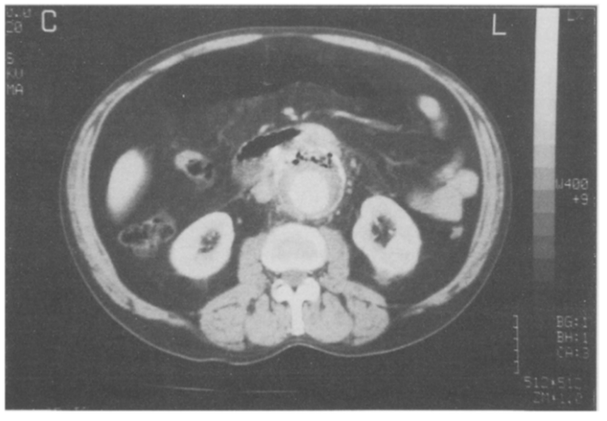

Fig. 1. Gas bubbles in an aneurysm visualised by a CT-scan are pathognomonic for the existence of a fistula. (Courtesy of Dr J de Gruyl).

Discussion

The incidence of PAEF is such that no single surgeon has a broad experience, the largest number of patients having been treated in one institution being 11,[6] with two other reports of eight and five patients each.[5,7] The knowledge on this clinical picture is therefore determined by case histories reported in literature. This image is necessarily distorted because many patients will die from unrecognised PAEF, and published case histories are likely to be biased in favour of successfully treated patients. We have established the occurrence of a primary aortoenteric fistula in at least 35 Dutch patients. This shows that the published cases are likely to be only the tip of the iceberg. The true incidence will of course never be known, but this figure seems to be the best approach of reality. Worthy of note is that 18 of the patients in this study have been treated in the last 3 years, while the other cases are scattered over a period of 15 years. A figure of at least two patients per year can be expected to occur in The Netherlands if we consider the entire period, increasing to six patients per year if we focus on the last 3 years.

As an aneurysm was demonstrated by palpation of the abdomen in 19 patients, this puts a complete physical examinatiion in the front line of our efforts to detect and treat patients with PAEF. Apart from endoscopy,[4] CT-scan appears to be a method that can positively confirm the presence of a fistula.[8,9] In view of the possibility of a sudden exsanguinating haemor-

rhage, an emergency laparotomy seems mandatory as soon as the diagnosis is considered.

The Dutch surgeons have adopted *in situ* grafting as standard treatment for PAEF, a policy that seems to be justified by the results. Out of 27 patients treated with *in situ* grafting, 14 are reported to be alive and well at long term follow-up. Six patients died postoperatively due to cardiac problems, coagulopathy or sepsis. Three patients died years after the operation of an unrelated disease. Four patients developed a secondary fistula, between 2 weeks and 6 years after the initial operation. In comparison, two out of three patients treated with extra-anatomic grafting died, one due to a cardiac arrest, and the other due to aortic stump blow-out. The threat of graft infection is a serious one, but a stump blow-out in patients with an extra-anatomic graft seems to be as likely as graft infection and a secondary fistula in patients with an *in situ* graft. In view of the operative mortality, it seems wise to choose the most simple solution, which is *in situ* grafting. Naturally one should pay appropriate attention to prevention of infection. For this purpose, attention is directed at the intestinal lesion as soon as haemorrhagic control has been achieved. Whenever the lesion is small and the inflammatory reaction limited direct transverse closure will be a safe and expedient procedure. In other cases, especially when the intestinal wall is involved in an inflammatory reaction, a limited resection and end to end anastomosis may be the best solution. After a specimen for culture has been obtained, the field should be thoroughly rinsed while the contents of the aneurysm is curetted as usual. Gowns, gloves and instruments should be changed. A Rifampicin soaked graft should be inserted.[10,11] Instead of closing the aneurysm wall around the graft, an omentum pedicle can be used to cover it. Antibiotics directed by the Gram-stain and culture are given for an arbitrary period of 6 weeks.

Acknowledgements

We wish to express our gratitude to all colleagues who have shared their clinical data with us, and in particular Dr J de Gruyl for providing the CT images of a primary fistula.

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Accepted 28 August 1995