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Congenital Oesophageal Stenosis Distal to Oesophageal Atresia

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Summary. Two cases with both oesophageal atresia and stenosis in the lower part of the oesophagus are reported. — The stenoses are congenital and most likely can be classified as malformations. — The stenoses had no clinical significance until the children began to eat solid food. Foreign bodies, which had been arrested in the oesophagus above the stenosis had to be removed several times. The importance is stressed of always including the distal oesophagus at the roentgenologic investigation of children with troubles after operation for oesophageal atresia.

Key words: Oesophagus, malformation, atresia and stenosis, atresia, stenosis.

Introduction

The occurrence of both oesophageal atresia and stenosis in the lower oesophagus is considered to be rare and is seldom mentioned in textbooks of pediatric surgery or diagnostic radiology. A total of 11 cases have been reported [2, 4, 5, 7]. Two cases of concomitant oesophageal atresia and stenosis are discussed in this report.

Material and Methods

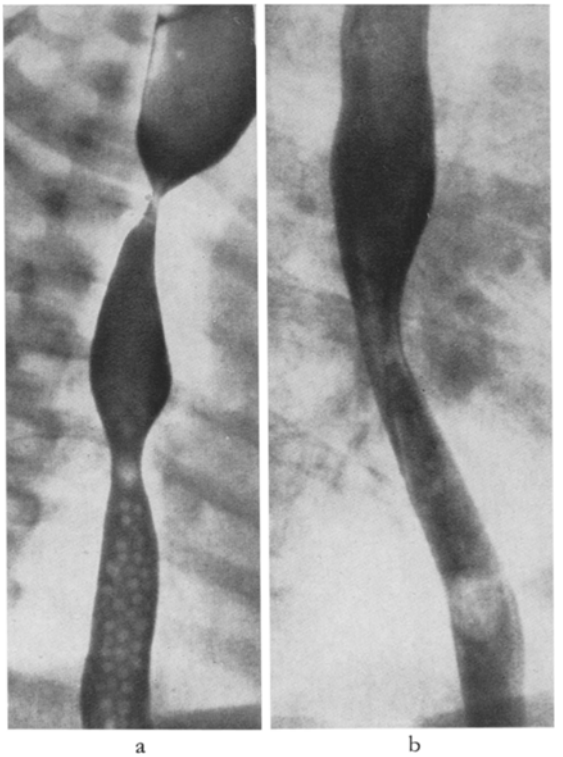
The material consists of 24 consecutive cases of oesophageal atresia in children born between 1969 and 1973. Seven cases were excluded because information on the condition in the distal oesophagus was lacking for various reasons: these included operation performed with colon interposition (2 cases), death of the patients before operation or before postoperative roentgen examination could be performed (4 cases), or because no appropriate roentgen examination was performed (1 case).

In the 17 remaining cases roentgen examination of the oesophagus, including cinefluorography, was carried out 2 weeks postoperatively with further follow-up examinations in all except one case. The follow-up period was 24 days to 3 years.

At roentgen examination attempts were made to provoke hiatal herniation and gastro-oesophageal reflux by successively increasing the intra-abdominal pressure with the aid of an inflatable girdle (6). This manoeuvre was not carried out at the first post-operative examination, however.

Results

A moderate oesophageal stenosis was found in 2 cases (Fig. 1 and 2). It was localised at the level of the left atrium: its length was about 1cm. and it was of hour-glass shape with smooth walls. In both cases the stenosis was demonstrated when the children were 2 weeks old. Hiatal herniation could



a b

Fig. 1a. Roentgen examination of the oesophagus 2 weeks after operation for oesophageal atresia and tracheo-oesophageal fistula. The oedematous swelling at the site of the anastomosis causes hindrance to passage of the contrast medium. The stenosis at the level of the left atrium also causes some difficulties in passage

Fig. 1b. $2^1/_2$ years after the operation. No obstruction at the site of anastomosis. The stenosis in the lower oesophagus causes hindrance to passage of the contrast medium

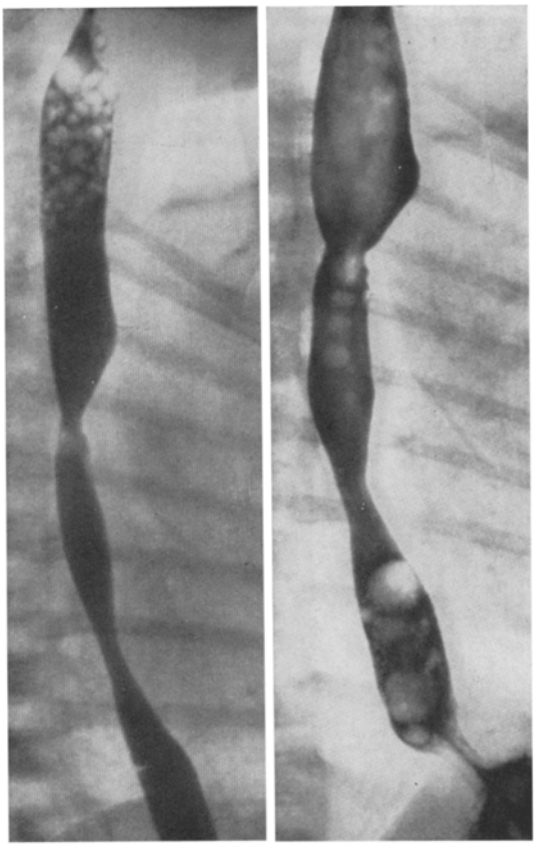


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Fig. 2. Another patient with a stenosis in the lower oesophagus. The roentgen examinations were made 2 weeks (a) and 6 weeks (b), respectively, after operation for oesophageal atresia and tracheo-oesophageal fistula

not be provoked. In a few cases some degree of gastro-oesophageal reflux could be provoked, but not in the 2 cases with stenosis.

The stenosis "grew" with the child during the follow-up period (2.5–3 years).

Discussion

The nature of the stenosis is not completely determined. The early demonstration of the stenoses — in one case the stenosis was seen the very first day of life [7] — strongly suggests that they are congenital. Their time of appearance and localisation and the absence of hernia and reflux all strengthen the idea that the stenosis is a malformation and not an acquired stricture. This is also supported by the fact that the width of the stenosis increased as the child grew and increased in stature, and, in the present 2 cases, no mucosal changes were noticed at oesophagoscopy. No histological examination was performed of the mucosa.

In the present two cases, as in most cases reported in the literature, there was a fistula between the trachea and the lower oesophageal segment. In one case with a long atretic segment, there was no fistula, however [5].

Stenosis of the same character and localisation is also reported in a case with tracheo-oesophageal fistula, but without oesophageal atresia [8], and it is reported as an isolated malformation in a great many cases [1, 3].

The frequency of the combination of oesophageal atresia and stenosis is difficult to assess. It was found by Perreault *et al.* [7] to occur in 3 out of 125 patients and it occurred in 2 out of 17 cases in the present investigation. It would seem not to be too rare. In a questionnaire it was reported in 3 out of 1,058 cases [4], but as information on the detail of the radiological investigation carried out is completely lacking the statement can only suggest a very minimum about incidence. Furthermore, stenosis can be missed if the examination is not skillfully performed.

As long as our two patients were fed with liquid or semi-solid food, the stenosis caused no trouble. After 10 months of age, however, both patients had repeated episodes of vomiting and breathing difficulties: foreign bodies, arrested in the oesophagus just above the stenosis, were found several times at roentgen examination and were subsequently removed at oesophagoscopy (3 and 5 times, respectively). These objects consisted of plastic pieces, stones and food remnants such as grapes or sausage.

There was no hindrance to the passage of the contrast medium through the anastomotic site in these 2 cases.

Children operated upon because of oesophageal atresia not infrequently have respiratory or swallowing difficulties for several months following operation. It is mandatory to include the lower part of the oesophagus in the roentgen examination of such cases.

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