Dysphagia due to multiple esophageal rings

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SN SULLIVAN. Dysphagia due to multiple esophageal rings. Can J Gastroenterol 1995;9(1):10-12. A 27-year-old Saudi man with dysphagia due to multiple esophageal rings is reported and the literature reviewed. Dysphagia due to multiple esophageal rings is very rare. Only 15 cases have been reported. The patient is usually male and has had dysphagia for many years when presenting. The cause of the rings is unknown. Theories to explain dysphagia are that the rings are either congenital or an unusual manifestation of gastroesophageal reflux.

Key Words: Dysphagia, Esophagus, Rings, Webs

Dysphagie due à des anneaux oesophagiens multiples

RÉSUMÉ : Le cas d'un jeune homme de 27 ans, originaire de l'Arabie Saoudite et atteint de dysphagie attribuable à la présence d'anneaux oesophagiens multiples, est présenté ici et la littérature est passée en revue. La dysphagie attribuable à la présence d'anneaux oesophagiens multiples est très rare. Quinze cas seulement ont été répertoriés. Le patient, habituellement de sexe masculin, souffrira déjà de dysphagie depuis plusieurs années au moment de la consultation. La cause de ces anneaux est inconnue. Selon les théories en vigueur, la dysphagie est due à la présence congénitale de tels anneaux ou à une manifestation inhabituelle du reflux gastro-oesophagien.

SINGLE RINGS OR WEBS OF THE proximal or distal esophagus are a well recognized cause of dysphagia. Multiple rings or webs of the esophagus are very rare; only about 21 cases where dysphagia was due to more than one web or ring are reported in the English literature. This paper reports a Saudi patient with dysphagia due to multiple

esophageal rings. The literature on the topic is also reviewed.

CASE PRESENTATION

A 27-year-old Saudi man presented with a one-month history of dysphagia for solids. He denied heartburn or acid regurgitation. He had ingested no caustic substances and had not taken any



Figure 1) Endoscopic photograph showing multiple esophageal rings/webs

medications known to cause esophagitis. He had no history of oral or skin lesions. Nine months previously he had been involved in a motor vehicle accident and sustained chest injuries. He remembered having a nasogastric tube inserted and left in place for one week. Past health and family history were unremarkable. Physical examination was normal.

The patient was endoscoped using an Olympus PQ20 panendoscope. Multiple rings were identified, beginning at 28 cm from the teeth (Figure 1). The endoscope could not be passed through the upper rings so a guidewire was passed under endoscopic control, and a 9 and 11 mm Savary Guillard bougie

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passed with little resistance. The patient was immediately rescoped and several disrupted rings with slight bleeding and many intact rings were seen extending down to 35 cm just above the gastroesophageal junction. There was a small hiatal hernia and lower esophageal sphincter the appeared incompetent, but there were no esophageal ulcers or erosions. The rest of the endoscopy was normal. Biopsy of one of the proximal rings was reported as showing unremarkable squamous epithelium. Three weeks later the patient attended for follow-up endoscopy. Although his dysphagia was much improved, the rings were still very evident. He was again dilated, this time up to 14 mm, with no pain experienced.

DISCUSSION

In 1958 Shamma'a and Benedict (1) reported 58 patients with esophageal webs, two of them having more than one web. These two patients were women with the Plummer-Vinson svndrome; one had two webs and the other had several webs, which were in the upper esophagus (1). Since then there have been a further 19 patients reported with two or more esophageal webs or rings (1-10) (Tables 1,2). In 15 cases the rings/webs were multiple (Table 2). All cases but one were men, ranging from 13 to 84 years old, and all presented with a history of food impaction usually requiring endoscopic intervention. The webs/rings were in the mid- and distal esophagus or involved the entire esophagus.

The cause of the rings is unknown. Except for four patients who had heartburn (3,5,10), there have been no obvious predisposing factors such as iron deficiency, caustic ingestion, pemphigus or epidermolysis bullosa. No consistent abnormality of esophageal motility has been identified that can explain the development of rings (2,4,6,10). It has been suggested that the rings may be an unusual manifestation of otherwise occult gastroesophageal reflux.

This theory is supported by the histological findings of basal zone hyperplasia, intraepithelial eosinophils or

TABLE 1

Case reports of several esophageal webs/rings

Author/year (reference)	Sex	Age (years)	Duration (years)	Location/ number*	Treatment
Shamma'a/1958 (1)	Female	44	10	Upper/two	†
	Female	60	15	Upper/several	†
Kelley/1966 (2)	Male	46	20	Upper and mid/four	Bougienage [‡]
Longstreth/1979 (3)	Male	21	14	Mid/two	Bougienage [‡]
	Male	18	8	Mid/two	Bougienage [‡]
	Male	40	15	Mid/two	Bougienage [‡]

*Number of webs/rings; ¹Treatment consisted of esophagoscopy with divulsion of the web, repeated bougienage, iron therapy or a combination of these; ¹Experienced an unusual degree of pain or bleeding

TABLE 2 Case reports of multiple esophageal webs/rings

Author/year	2	Age	Duration		
(reference)	Sex	(years)	(years)	Location	Treatment
Shiflett/1979 (4)	Male	81	12	Entire	Bougienage
Janisch/1982 (5)	Male	42	5	Upper and mid	Bougienage
	Male	42	17	Upper and mid	Refused
Munitz/1983 (6)	Male	84	10	Entire	None
Carlisle/1984 (7)	Male	14	2.5	Entire	Bougienage*
Agarwal/1990 (8)	Male	37	22	Entire	Unknown
	Male	50	40	Entire	Bougienage*
Harrison/1992 (9)	Male	35	Lifelong	Entire	Bougienage*
	Male	68	Lifelong	Entire	Refused
Bousvaros/1992 (10)	Male	15	ns	Entire	$AA+H_2RA$
	Male	13	ns	Entire	$AA+H_2RA$
	Male	14	ns	Distal third	$AA+H_2RA$
	Male	17	ns	Distal third	$AA+H_2RA$
	Male	18	ns	Entire	$AA + H_2RA$
	Female	9	ns	Entire	t
Present report	Male	27	1 month	Mid	Bougienage

*Experienced an unusual degree of pain or bleeding; † Rings disappeared after fundoplication. AA+H₂RA Antacids and H₂-receptor antagonists; ns Not stated

both on microscopic examination (5,6,8,10) and in abnormal pH studies in the few patients reported (10). However, medical antireflux treatment, even with omeprazole, has not affected the rings. One child bas been reported whose esophagus returned to normal following a fundoplication (10). A congenital origin is suggested in some cases by the very long duration of symptoms. A congenital and possible familial origin is also supported by the report of multiple rings in a father and son. Another male child in that family died at age one week because of "an inability to swallow" (9). The potential etiological factors in the presented patient were his nasogastric intubation

for one week after his motor vehicle accident and chest injury, and the endoscopic impression that his lower esophageal sphincter was incompetent.

The dysphagia caused by the rings is easy to treat by bougienage but some caution must be exercised. Of the 11 patients treated by bougienage seven experienced bleeding or an unusual degree of chest pain after dilation. Since in many of the cases the degree of esophageal narrowing prevented the passage of all but the smallest endoscope, it may be prudent not to dilate the esophagus to a normal diameter too quickly.

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