Epiphrenic Esophageal Diverticulum*

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A case of congenital pulsion diverticulum of the lower esophagus with a long history of about 22 years duration is presented. Resection was carried out with successful end result. The literature on the subject is reviewed.

Lahey and Warren,¹ in their study of 365 cases of pharyngo-esophageal diverticula and an unstated number of traction diverticula of the esophagus, found only nine cases of epiphrenic diverticulum. Effler and co-workers² found that only seven cases were recorded over a period of eight years at the Cleveland Clinic. Such an infrequency appears to justify the presentation of one case of epiphrenic diverticulum seen at the Department of Surgery of Sheth Vadilal Sarabhai General Hospital, Ahmedabad.

CASE REPORT

A Hindu man about 36 years old was seen on October 24, 1965, in the Department of Surgery of the Sheth Vadilal Sarabhai General Hospital, Ahmedabad, with the chief complaint of dysphagia to solid food for the past year and a half associated with hiccough. For the last three months he also developed difficulty in swallowing liquids. As the skigram of chest revealed tuberculous lesion in the right upper lobe, he was put on anti-tuberculous drugs, as a result of which he gained weight and became symptom free for some time. At the age of 14 years, he had developed pain in the chest and midsternal discomfort, after having taken two to three mouthfuls of meal. This subsided after vomiting. Such attacks used to recur at gradually diminishing intervals. The dysphagia recurred with a feeling of obstruction at the midsternal level; but he did not have any discomfort or pain in the interscapular region. At the time of his admission to the hospital, he could only swallow fluids, part of which he vomited.

On examination, he was moderately built and nourished. His conjunctiva and nails were of normal color and he had no palpable glands in the neck. His tongue was moist and coated. His pulse was 82 per minute, temperature was normal, and blood pressure was 120/80 mm of Hg.

Liver and spleen were not palpable. Cardiovascular, respiratory, and central nervous systems were normal.

Investigations

Results of urinalysis were normal. Hemoglobin, 68 percent (11.6 gm per 100 ml); blood group B III., RH positive; bleeding time, 2 minutes, 48 seconds; clotting time, 2 minutes, 20 seconds. Screening of chest-lung fields was clear with no evidence of bronchiectasis. The heart size was within normal limits.

Esophagogram

There is a large (about 6.5 cm. diameter) "pulsion" type of diverticulum with a narrow neck along the right side and posterior aspect of the lower part of thoracic esophagus, situated about 5 cm above the diaphragm. The upper portion of the esophagus is dilated. No hiatus hernia is seen (Fig 1).

On November 12th, thoracotomy was carried out through the bed of left eighth rib. The diverticulum was of a congenital pulsion type, covered with muscular layer, about 2½ inches × 2½ inches in size, with a 1-inch wide neck. The proximal esophagus was dilated while the distal part was normal. The diverticulum was excised and the mucosa was sutured with interrupted 000 chromic catgut while the muscular layer was closed with interrupted 0000 silk sutures. The chest was closed with intercostal underwater seal drainage. The postoperative course was uneventful and at the time of discharge, he had no difficulty with swallowing. Barium study of the esophagus on November 29th, showed complete removal of the previously seen epophageal diverticulum. There were irregular waves of contraction in the lower half of the esophagus, giving rise to "corkscrew" appearance.

Biopsy Report

Section shows the wall of esophageal diverticulum lined by hyperplastic squamous epithelium with a collection of chronic nonspecific inflammatory cells underneath, in which a fair number of eosinophils are also seen. In the wall, few fibers of muscularis mucosae are seen but the rest of the wall consists of connective tissue with a fair

Figure 1. Esophagogram outlining a pulsion diverticulum of the lower part of the esophagus.
number of congested blood vessels and scattered small collections of chronic nonspecific inflammatory cells consisting chiefly of lymphocytes and a few plasma cells.

When the patient was contacted at follow-up two years later, he reported that he was symptom free and would come later for radiological studies.

**Discussion**

Zenker and Ziemssen\(^8\) classified esophageal diverticula into (1) traction type, usually found in the midthoracic region, the result of cicatricial contraction of the esophageal wall caused by tuberculous or other forms of chronic lymphadenitis. Occasional-\(\text{ly},\) however, even the pulsion of the esophagus, which is a normal phase in the upper gastrointestinal duplication, is a normal phase in the solid stage of the human embryo during the eighth and ninth week. Alternatively, it may arise from persistent vacuoles, which is a normal phase in the solid stage of the alimentary tract, during the sixth and seventh weeks of fetal life. The diverticulum resulting from the persistence of congenital diverticular phase resembles very much an acquired pulsion diverticulum. Occasionally, it may have all the layers of the esophageal wall, including the muscular layer. The congenital diverticulum resulting from spherical duplications of alimentary tract acquiring a luminal connection will also contain all the layers of the esophageal wall; but it may at times contain, even gastric, colonic mucosa, or pancreatic tissue. At times, the inner or fused part may contain a variable number of layers of the esophagus. D’Abreu\(^8\) reported the first fully documented case of spherical duplication luminally connected with esophagus with all the layers in the wall, as well as islets of gastric and pancreatic tissue in the mucosa. Mendl and Evans,\(^7\) also recorded a similar case, in which they were also able to demonstrate muscular activity in its wall on radiological examination, thus enabling them to make the preoperative diagnosis of a congenital epiphrenic diverticulum. These authors also record two patients with epiphrenic esophageal diverticulum, who also had additional diverticula of the duodenum and jejunum. Such an association supports the hypothesis of upper gastrointestinal duplication as a mechanism of esophageal diverticulum formation.

A number of workers have commented on the additional factors responsible for the formation of acquired pulsion diverticulum. Effler and associates\(^3\) have noted the presence of hypertrophic circular muscle distal to the diverticulum, hiatus hernia, and diverting mucosal flap, opposite the opening into lumen, thus diverting the bolus of food into the diverticulum instead of the esophagus. Habein and co-workers\(^8\) observed associated lesions in 17 out of 24 cases studied by them. These were hiatus hernia in 11, cardiospasm in 2, diffuse spasm in 9 and esophagitis in 6 cases. Some of these conditions co-existed. Belsey\(^10\) has presented a most exhaustive study on the functional diseases of the esophagus other than those due to organic causes. The conditions are upper esophageal spasm, diffuse spasm giving rise to corkscrew esophagus and achalasia cardia. Diffuse esophageal spasm if persistent for any length of time is likely to lead to pulsion diverticula. According to Ellis and Payne,\(^11\) the motility disturbances of the esophagus studied by intraluminal pressure studies using pressure transducers, can be divided into (1) hypomotility disturbances characterized by achalasia and (2) hypermotility disturbances characterized by diffuse esophageal spasm. It is in the latter variety that both hiatus hernia and epiphrenic diverticulum are frequently associated.\(^12\) Goodman and Parnes\(^13\) have demonstrated a diaphragm below the level of the diverticulum.

Johnstone,\(^14\) however, did not find a diverticulum in 200 cases of achalasia studied by him. In the present case, no specific abnormality in the esophagus was demonstrated and it was a congenital type of a pulsion diverticulum.

A large number of diverticula are asymptomatic and they are detected only on routine barium study of the esophagus. Gerard and Sabety\(^15\) observe that more cases of pulsion diverticula are seen with increasing use of barium studies of the esophagus. The more frequent symptoms encountered are retrosternal pain, dysphagia, regurgitation of food, vomiting, epigastric fullness or pain and occasionally gurgling noises in the chest. In longstanding cases there may be inanition and anemia. The duration of symptoms in most of the cases is very long even up to 30 years. In the present case, although the history of dysphagia was of 1½ years only, the minimal symptoms appear to date back 22 years.

In any case definite diagnosis is established only on detailed barium studies of the esophagus. In addition to the demonstration of the diverticulum and its size, radiological examination will also
demonstrate the associated lesions as cardi spasms, hiatus hernia, diffuse spasm, etc. and thus aid the planning of the operative procedure in good time.

One of the conditions for which an epiphrenic diverticulum may be mistaken is the hiatus hernia especially when the sac is large as was considered in the present case. A careful radiological study would, however, remove the doubt.

In the diagnosis of this condition esophagoscopy is usually avoided because of the risk involved. The instrument may be directed into the diverticulum resulting in perforation with its inherent dangers of mediastinitis. It only helps in detecting either esophageitis or a rare occurrence of a carcinoma within the diverticulum, but the danger of perforation outweighs its usefulness.

Lahey and Warren,1 in the early period, had used diverticulopexy for such epiphrenic diverticulum in three cases. The method probably was justified then, in view of the grave risks involved in the thoracic surgical procedures. De Bakey and Creech observed that surgical treatment of epophageal diverticulum had considerably improved. To-day, most authorities believe in resection of the diverticulum, since the risks have minimized considerably and the results are gratifying. Dilatation of the esophagus over a swallowed thread used to be practised at one time, but to-day it is rarely employed except to help relieve tension on the suture line.9 It becomes imperative to correct the associated lesions which are likely to influence the chances of recurrence.2,10 The repair of hiatus hernia and the excision of a diverting mucosal flap appear to be justified. The addition of an adequate esophagomyotomy from the neck of the sac up to the cardia should be an essential part of the procedure as justified by the good results.2 It, however, carries with it the risk of occurrence of reflux esophagitis with resultant stricture formation. In view of this, it would appear more reasonable to carry out esophagomyotomy only in the presence of cardi spasms. In the present case, only the resection of the diverticulum was carried out without any additional procedure on the esophagus and the patient was completely relieved of symptoms. It would, therefore, be proper to conclude that his symptoms were related to the mechanical pressure caused by the large size of the diverticulum. The need for resection and anastomosis arises only in cases with stricture formation resulting from reflux esophagitis. In rare cases with an inanition and severe anemia, however, a preliminary gastrostomy may add considerably to the overall safety of the patient by improving his general condition.

ACKNOWLEDGMENT: Our thanks are due to the Superintendent, Sheth Vadilal Sarabhai General Hospital, Ahmedabad, for permission to use the hospital records.

REFERENCES