

Severe Benign Esophageal Stricture Caused by Incidental Radiation Exposure of Gastric Inlet Tissue: Case Report

Gastrik Inlet Dokusunda Rastlantısal Radyasyonun Neden Olduğu Ciddi Özofageal Benign Striktür

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ABSTRACT Gastric heterotopia is defined as ectopic presence of gastric type mucosal tissue in another organ and/or a surface. It's most frequently found in the 1/3 proximal part of the esophagus where it's called (gastric) inlet patch. Although most of these lesions are asymptomatic, they might cause small peptic ulcer or erosion, stricture, stenosis, fistula formation and intestinal metaplasia, high grade metaplasia or adenocancer of the esophagus. Radiation therapy has diverse and sometimes severe effects on gastrointestinal mucosa. We present here a gastric inlet case treated with radio- chemotherapy for breast cancer who developed severe esophageal stricture 1 year after radiation therapy. Because gastric inlet has been found up to 10% in general population, clinicians should consider gastric inlet in every upper endoscopic interventions and should keep in mind that patients who are positive for gastric inlet are at higher risk for development of esophageal complications such as stricture when thoracic radiotherapy is applied.

Key Words: Radiotherapy; esophageal stenosis; deglutition disorders

ÖZET Gastrik heterotopi, gastrik mukozanın diğer organ veya yüzeylerde ektopik bulunması olarak tanımlanır. Sıklıkla özofagusun 1/3 üst bölümünde rastlanır ve (gastrik) inlet patch olarak adlandırılır. Her ne kadar asemptomatik olsa da, bazen peptik ülserden adenokansere kadar değişen lezyonlara zemin hazırlayabilmektedir. Radyasyon tedavisi gastrointestinal mukozada farklı ve bazen ciddi etkilere neden olmaktadır. Bu yazıda, meme kanseri nedeniyle radyo-kemoterapi uygulanan ve radyasyon tedavisinden 1 yıl sonra ciddi özofageal striktür gelişen bir olgu sunduk. Genel popülasyonda %10 oranında bulunabildiği için, klinisyenler üst endoskopi yapılması sırasında gastrik inlet göz önünde bulundurulmalıdır ve torakal bölgeye yapılacak radyoterapilerde striktür gibi özofageal komplikasyonların gelişiminde gastrik inletin katkısının olabileceği akılda tutulmalıdır.

Anahtar Kelimeler: Radyoterapi; özofageal darlık; yutma bozuklukları

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Gastric heterotopia is the presence of gastric mucosa and glands in a tissue or organ other than stomach. These lesions can be seen at any level in the gastrointestinal system.¹ Upper esophagus is the most frequent site to be involved.² Presence of this tissue in the 1/3 upper esophagus has been called *inlet patch (gastric inlet)*. Although it's frequently asymptomatic, it might constitute an area involving different stages of lesions varying from ulcer to dysplasia and adenocarcinoma.³

In this report we presented, a case who has received radio-chemotherapy for breast cancer and admitted to our clinic one year after radiotherapy

with the complaints of dysphagia, odynophagia and who was later diagnosed with *gastric inlet* has been presented. To the best of our knowledge, there is not a similar case in the previous literature.

CASE REPORT

A 68-years-old woman has diagnosed with breast cancer 5 years ago and underwent radical mastectomy. She had recurrence of the disease later on and has received radiotherapy at a dose of 2 Gy/day, 5 days a week for 5 weeks to reach total dose of 50 Gy. Also she received a chemotherapy regimen including Adriamycin and Taxol after radiotherapy at 6 sessions every 3 weeks apart. She stated that her dysphagia started about 1.5 months ago, initially for solid food and progressed to include liquids later on. She also complained about retrosternal chest pain, palpitation, food aspiration, cough and dyspnea all happening during swallowing. She had significant difficulty to thrive and lost about 10 kg in this period.

On physical examination she had fine late inspiratory crackles at bilateral bases. Other system examinations were normal. Laboratory work-up were as the following: Haemoglobin: 9.1 mg/dL, ESR:54 mm/hr, Creatinine: 2.1 mg/dL, Albumin: 3.0 g/dL. Other laboratory tests were normal. An esophagogram was performed for evaluation of dysphagia which showed mucosal irregularity, rigidity and a shoulder sign producing lesion at the 1/3 proximal portion of the esophagus (Figure 1). Further evaluation was done with esophagoscopy. It showed pink-red, abnormal appearing lesion starting few centimeters from the upper sphincter with irregular surface and causing obstruction of the esophagus to the degree that it was impossible to move the scope through. Multiple biopsies were taken from the lesion and sent for pathologic examination.

Although malignancy was the initial suspect, pathology showed columnar gastric epithelial and glandular cells under squamous epithelial of the esophagus. No malignant changes were seen (Figure 2). Therefore, a diagnosis of gastric heterotopia was concluded. A gradual decline in her general condition was noted in follow-up and she died of sepsis.

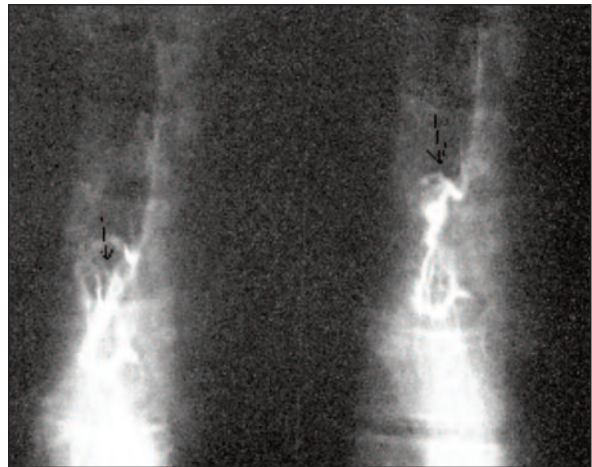


FIGURE 1: Mucosal irregularity, rigidity and a shoulder sign producing lesion (arrow) at the 1/3 proximal portion of the esophagus in esophagography.

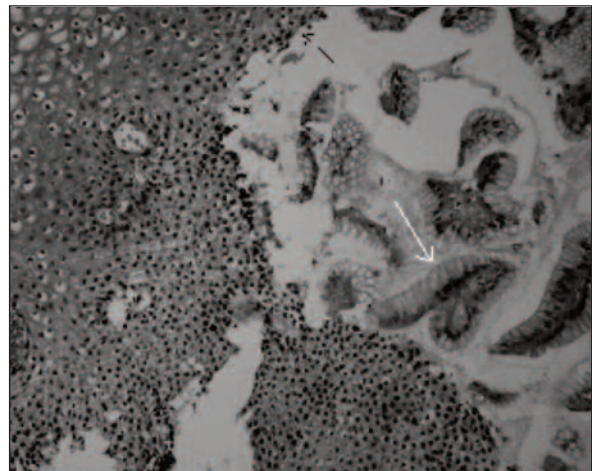


FIGURE 2: Columnar gastric epithelial and glandular cells (white arrow) under squamous epithelial of the esophagus (black arrow) (x200) (HE).

DISCUSSION

Gastric inlet contains gastric type mucosa and it resembles gastric corpus mucosa or cardiac glands.³ *Gastric inlet* is often encountered incidentally during esophago-gastro-duodenoscopy. Its prevalence is 2-4%,⁴ however in a prospective study, prevalence up to 10% has been reported.⁵ A study from our country reported a prevalence rate of 3.6% for gastric inlet and a tendency to occur more frequently in women.⁶ Akbayır et al⁷ reported a lower prevalence rate (1.67%) with a female dominance. These lesions are usually asymptomatic, but as they involve gastric type mucosa, this might cause peptic symptoms or if severe enough complications like peptic

ulcer, erosions, stenosis, fistula and stricture formation as well as perforation, bleeding and intestinal metaplasia.^{2,3} Also high-grade dysplasia and adenocarcinoma of the esophagus might originate from these tissues.³ Radiation has been reported to account for 4% of the esophageal strictures.⁸

Radiotherapy affects mostly the tissues like hematopoietic and gastrointestinal system that have high cell-turnover rates.⁹ Radiation related gastrointestinal damage can be acute or chronic. In acute disease, radiation causes epithelial cell destruction and disturbs cell proliferation. On the other hand, advanced fibrosis and presence of atypical fibroblasts is characteristic of chronic radiation related damage. This progressive ischemic damage and fibrosis might cause stricture and fistula formation as well as more severe complications like perforation. Acute radiation esophagitis is usually seen at the second week of radiation therapy.¹⁰ In our case, the symptoms began about 1 year after completion of radiotherapy. This and the severe stricture formation suggested more chronic rather than acute radiation related damage. Chronic effect of radiation on the esophagus can be seen in 5 years after radiation exposure and usually presents with strictures. Stricture formation is dose related and studies suggest that radiation related esophageal damage takes place in seen 1-5% when total dose of radiation is 60 Gy and 50% when the total dose reaches to 75 Gy. Barium swallow study

is an aid in diagnosis of esophageal stricture and fistula formation. But as malignancy needs to be ruled out, upper GI endoscopic study with brushings and tissue biopsy is usually needed.¹⁰ It was remarkable for our patient to develop such a severe esophageal stricture although she received relatively low dose of radiation.

Combining radiation with chemotherapy has been shown to decrease esophageal tolerance to radiation damage and increase its toxicity. This is more significant with platinum or taxane based chemotherapy.¹¹

It is notable for this stricture forming lesion to be found at the *gastric inlet* tissue. We thought that it might develop due to increased destructive effect of peptic activity secondary to radiation exposure. We also think that chemotherapy has increased the toxic effect of radiotherapy. It is also likely that chronic vascular damage and related ischemia and fibrosis plays a role just like in development of radiation colitis. Because *gastric inlet* has been found up to 10% in general population, clinicians should consider gastric inlet in every upper endoscopic interventions and should keep in mind that patients who are positive for gastric inlet are at higher risk for development of esophageal complications such as stricture when thoracic radiotherapy is applied. By this way, these patients might be treated prophylactically if ever they need to receive radiation therapy in the future.

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