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A rare case of esophageal lymphangioma associated with gastric adenocarcinoma

Mide adenokarsinomu ile birliktelik gösteren özofagus yerleşimli bir lenfanjioma olgusu

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ÖZET: *Lenfanjioma; lenf damarlarının benign tümörüdür. Genellikle çoçukluk çağında karşımıza çıkar. Bugüne kadar sadece 7 adet özefagus yerleşimli lenfanjioma olgusu bildirilmiş olup, özefagial yerleşimli lenfanjioma ile mide adenokarsinomunun birlikteliği bildirilmemiştir. Bu makalede mide adenokarsinomu ile birliktelik gösteren özefagus yerleşimli bir lenfanjioma olgusu sunulmuştur.*

Bu olgu zaman zaman ortaya çıkan karın ağrısı, yutma güçlüğü, oral alımın olmaması, kilo kaybı ve genel durum bozukluğu ile yatırıldı. Kliniğe yatışını takiben yapılan üst gastrointestinal sistem endoskopisinde dişlerden itibaren 32-34 cm'lerde lümeni tamamen kapatan kitle saptandı. Alınan biopsinin sonucu lenfangioma olarak rapor edildi. Hastanın tamamen durmuş olan beslenmesini sağlamak amacıyla cerrahi gastrostomi uygulandı. Gastrostomi yolundan beslenmede zorluk olması ve verilen gavaj mayiinin geri gelmesi üzerine gastrostomi ağzından baryum verilerek çekilen grafilerde pilor kanalından distale geçiş olmadığı görüldü. Gastrostomiden fiberoskopla girildi. Mide içerisinde bol miktarda hemorajik mayii ve antrumu tutan kitle lezyonu görüldü. Buradan alınan biopsiler grade I adenokarsinoma olarak rapor edildi. Hasta bir hafta sonra kaybedildi.

Anahtar Kelimeler: **Özofagus, lenfanjioma, gastrik adenokarsinoma**

SUMMARY: *Lymphangioma is the benign tumour of lymphatic vessels. It is usually observed in childhood. Only 7 cases with esophageal localization have been reported until now, but neither of them had been with gastric adenocarcinoma as the case reported below.*

The case was hospitalized with complaints of intermittent pain in the abdomen, dysphagia, no oral intake and weight loss. An esophagoscopy was performed to the patient and a mass that obstructed the entire esophageal lumen was observed at 32-34 cm from the teeth and multiple biopsies were taken. The biopsy material was reported as lymphangioma. Surgical gastrostomy was performed in order to feed the patient. When all feeding material came back, barium meal was given from the gastrostomy and X-ray studies showed that there was no passage through the pylorus. Profuse haemorrhagic material and a mass in the antrum was observed during endoscopy through the gastrostomy. Biopsies were taken and reported as adenocarcinoma grade I. The patient died after a week.

Key Words: **Esophagus, lymphangioma, gastric adenocarcinoma**

LYMPHANGIOMA is the benign tumour of lymphatic vessels (1). It is usually observed in childhood. Ninety percent of cases are manifested in the second year of life (2). It is rare in adulthood and usually located in head, neck and axilla (3). The most common type in adulthood is superficial cutaneous lymphangioma (3-6). Until now, only 7 cases with esophageal localization have been reported but neither of them had been associated with gastric adenocarcinoma (7). In this article a rare case of benign lymphangioma associated with gastric adenocarcinoma has been reported.

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CASE REPORT

A 69-year-old woman, with complaints of intermittent pain in the abdomen since June, 1993, dysphagia with solids first thereafter with liquids since July, 1995 and finally no more oral intake during the last 15 days and weight loss of 15 kg. within 45 days of admition to the GATA Haydarpaşa Training Hospital Internal Medicine Clinic. Physical examination revealed that her activity was stage 4 according to Eastern Cooperative Oncology Group (ECOG) with difficult cooperation and no orientation. Her appearance was cachectic with weak turgor and tonus. The pulse rate was 110/min. Expiratory rhonchi in lower lobes of the lungs were noted on auscultation. There was epigastric tenderness on palpation of the abdomen and bowel sounds were decreased.

Laboratory results were as follous; white blood cell count was 10.400/mm^3, erythrocyte count was 4.330.000/mm^3, haemoglobin 9.9 g/dl, haematocrit

Figure 1. The histopathologic appearance of esophageal lymphangioma (Haematoxylin-eozin X20)

was 28.1%, MCV was 63.8 fl, albumin was 3 g/dl, erythrocyte sedimentation rate was 22mm/h. Glycemia, blood urea nitrogen (BUN), creatinine, bilirubinemia, Na, K, ALP, ALT, cholesterol, triglyceride, LDH and total protein levels were all within normal levels. In blood smear, there was hypochromia and microcytosis with PMN 84%, lymphocyte 12% and monocyte 4%. Occult blood in stool was (++). Serum iron (Fe^{++}) was 40 mcg/dl (N: 50-150), serum iron binding capacity was 452mcg/dl (N: 150-450). CEA (Carcino Embryonic Antigen) was 1.6 ng/ml (N: 0-3 ng/dl).

The patient underwent esophagoscopy which revealed that the esophageal lumen was almost obstructed by a vegetative mass at 32-34 cm from the teeth. After multiple biopsies were taken from the proximal parts of the mass, endoscopy was ended. The biopsy material was reported as lymphangioma (Figure 1).

Gastric wall thickness was increased and gastric luminal appearance was hypoechoic on abdominal ultrasound. Abdominal CT (computed tomography) revealed a mass lesion with hydro-aeric levels that occlude the esophagus lumen from the aorta-pulmonary window to the cardia and another mass of 7x5x3 cm located on the lesser curvature of stomach (Figure 2).

In order to feed the patient, surgical gastrostomy was performed using 36F Pezzer uretheral catheter. The patient was given 2000 kcal/day enteral feeding solutions for 3 days with difficulty. When feeding material refluxed, barium meal was given from the gastrostomy and X-ray studies showed that there was no passage through the pylorus. A guide was inserted into stomach and the gastrostomy tube was extracted. By way of the guide gastroscopy (Fujinon UGI-FT7) was performed and profuse haemorrhagic material and an antral mass were observed. Biopsies were taken and reported as adenocarcinoma grade I. Because the patient was old and her general status was poor, a palliative operation to supply a passage could not be performed and TPN (Total Parenteral Nutrition) was started but the patient died after a week.

DISCUSSION

Lymphangioma is usually observed in childhood and located in head, neck and axilla (3). Ninety percent of cases are manifested in the second year of life (2). It is rare in adulthood and the most common type in adulthood is superficial cutaneous lymphangioma (3-6). According to Anderson's study lasted for fifteen years, only 48 of 768 benign tumours were lymphangiomas (1). The cases of gastrointestinal lymphangioma have also been reported in the literature. But only 7 cases with esophageal localization have been reported (7). In the trial of Kori, 2 of 38 mediastinal mass were lymphangiomas and one of them was with esophageal localization (7). Cases with gastric, jejunal and gallbladder localization have also been reported (8-10). None of these cases has been associated with gastric adenocarcinoma. Since no case of benign lymphangioma associated with gastric adenocarcinoma has been reported in the literature, this case would be an unusual one. We also believe that the diagnostic procedure for the mass that occlude the antropyloric passage makes the case report interesting. But if fine needle aspiration bi-

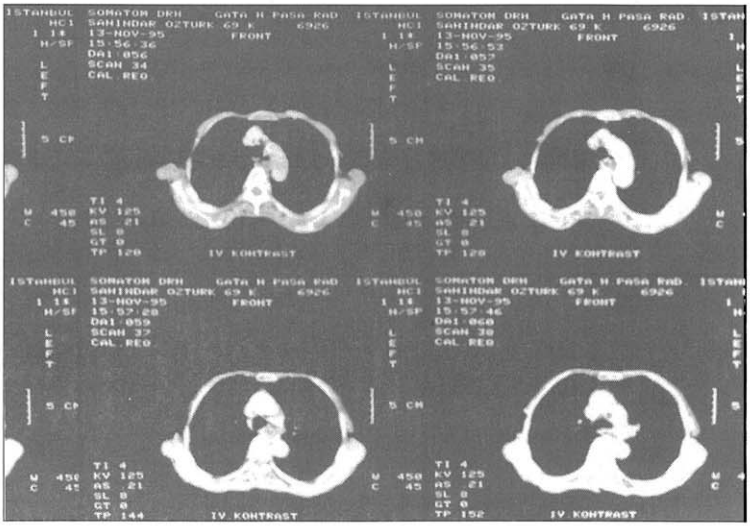

Figure 2. The appearance of esophageal lymphangioma in Computed Tomography

opsy would have been done after computed tomography, the surgical gastrostomy operation had not been performed.

As a conclusion, in cases with benign lymphangioma, the possibility of malignancies should be kept in mind as well.

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