CASE REPORT

Malignant Pleural Mesothelioma and Gastric Metastasis: A Very Rare Case Report and Literature Review

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ABSTRACT

Introduction: Malignant pleural mesothelioma (MPM) is a rare neoplasm. It has closed association with occupational asbestos exposure. Symptoms are commonly due to local invasion of pleura and mediastinal structures. MPM may have local or rarely distant organ metastasis by haematogenous spread in different organs such as liver, adrenal gland, kidney and contralateral lung. However, gastrointestinal involvement is very rare. **Case Report:** We report herein a 58-year-old female patient who was presented with back painand finally was diagnosed as MPM with distant metastasis to the stomach. **Conclusion:** Clinical, imaging and histopathologic findings play an important role in influencing the prognosis as well as treatment.

Nergis Ekmen

INTRODUCTION

Malignant mesothelioma which originates from serosal cells of pleural, peritoneal and pericardial surface, is highly aggressive tumor. Recent epidemiological studies revealed the primary cause of the disease that is asbestos exposure. Local recurrence is not rare, however, gastrointestinal (GIS) metastasis specially for gastric involvement is very unusual condition which has been rarely reported in the literature.[1]

In this article, clinical, imaging findings and literature review of a case with malignant pleural mesothelioma (MPM) and its distant metastasis to the stomach, is presented.

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CASE REPORT

A 58-year-old female patient with complaint of back pain for 3 month duration admitted to the hospital. In routine screening tests, PA x-ray chest film showed right pleural effusion (Figure 1). Biochemical parameters revealed normal except for normochrome normositer anemia (Hb:11,2 mg/dl Htc:33,8 Plt:351.000, Wbc:8700 MCV:84). In thoracal computed tomography (CT) examination, massive pleural effusion, mediastinal shift and compression atelectasia without mediastinal lymphadenomegaly was detected (Figure 2). With known history of asbestos exposure of the patient, marker tests were studied (CEA negative (CEA31), CK 5/6 positive, Calretinin positive, Ber Ep4 Gene Tex negative, TTF-1 (SPM150) Gene tex negative). Regarging to these findings, pleural biopsy, using video-

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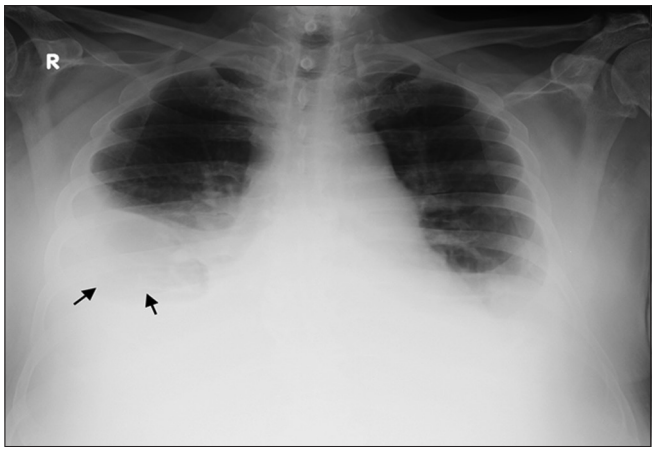

Figure 1: PA X-ray chest graphy showed right pleural effusion (arrows)

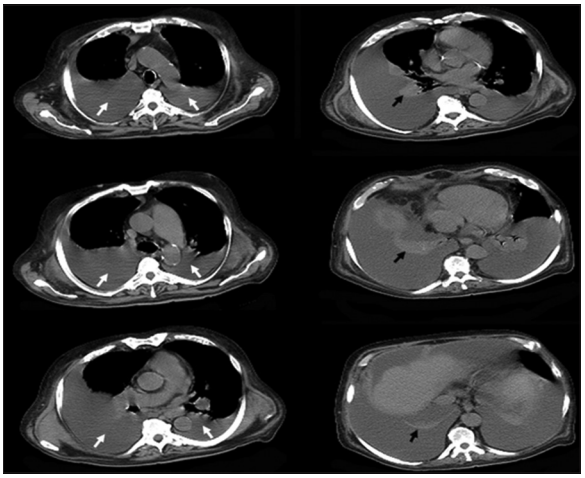

Figure 2: Thorax CT study showed massive pleural effusion (white arrows), mediastinal shift and compression atelectasia (black arrows) without mediastinal lymphadenomegaly

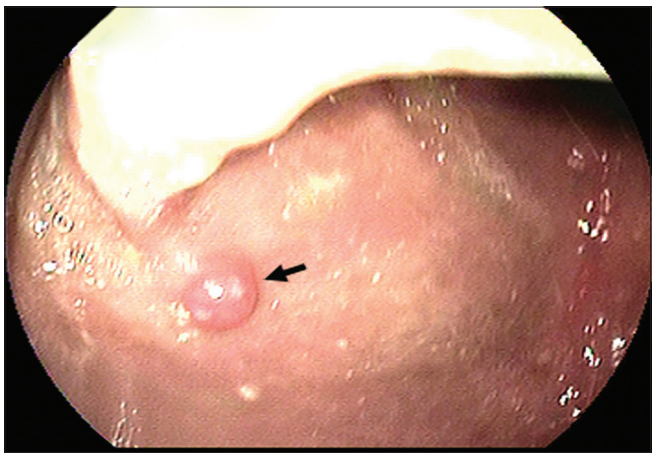

Figure 3: Upper gastrointestinal endoscopy examination showed fndings of esophagitis and 7 mm in diameter size polypoid lesion in the location of distal gastric corpus (arrow)

assisted thoracic surgery (VATS) technique was performed and malign mesothelioma was reported. Morphologic, immunopathologic findings were compatible with epithelioid type malignant mesothelioma. PET-CT scanning demonstrated right hemithorax pleural effusion and low intensity FDG-uptake in the pleura. The patient underwent surgery. In thoracotomy operation, massive thickening of both parietal and visceral pleura was observed, all parietal and visceral pleural surfaces even diaphragmatic surfaces were decorticated. About 80 ml visceral pleura and 100 ml parietal pleura were excised macroscopically, sectional studies revealed nodular pattern area with the largest size of 1 cm diameter. In histochemical studies tumor cells were calretinin (+), cytokeratin 5/6 (+) immunoreactivity, podoplatinine (+); findings were assessed as epithelioid type malign mesothelioma.

Three cure CDDP + Alimta was given. Three month after therapy, control thoracal CT showed no residual tissue or recurrence. Five cure CDDP + Alimta regimen was received totally. Six month after completed therapy, control thoracal CT showed nodular type thickening of the right mediastinal pleura upto 2 cm in size and high FDG- uptake of mediastinal pleura due to mesothelioma recurrence was detected in PET-CT scanning. No FDG-Uptake was detected in abdominopelvic and skeletal system.

Radiotherapy (RT) was planned for right mediastinal recurrence and right posterior thoracic wall lesion. Four month after RT due to dyspepsia, upper gastrointestinal endoscopy was performed. Esophagitis and 7 mm in diameter polypoid lesion was detected, endoscopic polypectomy was applied (Figure 3). Because of being not cost-effective and not changing the course of treatment, and due to have been proven by biopsy, endoscopic ultrasonography (EUS) study was not done. An immunohistochemical study calretinin (+), CD15 (-), CEA (-), cytokeratin 5/6 (-), HBEM-1 (+), panCK (+),S-100 (-), WT-1 (-), histologic grade II was reported. No vascular and perineural invasion was observed. Findings were evaluated as mesothelioma infiltration both histochemically and immunohistochemically which was compatible with gastric metastasis. No lymphadenomegaly or metastasis was detected in control abdominal CT examination. Control thoracal CT revealed right pleural effusion, nodular thickening of mediastinal pleura and fibrotic changes duo to RT.

The patient was followed palliatively for nine month duration. In the last two month follow-up, gradual CRP and leukocyte increasement was detected. However, it was not compatible and explainable with the present clinical condition. CT examination was performed for present abdominal distension and pain, showing peritoneal metastasis which was considered as haematogenous metastasis. Rapid deterioration in general condition occurred and the patient died two years after the initial diagnosis.

Table 1: Reported cases of gastrointestinal metastases of MPM

Case	Author	Year/sex	Metastaticsites	Histologicaltype	Symptoms	Diagnosticmethods
1	Kakukawa et al.	62/M	Multiple (jejunum~ileum)	Unknown	Bloody stool	CE DBE
2	Chen et al.	73/M	Duodenum	SM	Bloody stool	GIF
3	Terashita et al.	63/M	Duodenum	EM	No symptoms	GIF
4	Gocho et al	52/M	Jejunum	BM	Abdominnalpain	Ope
5	Hayashi et al.	70/F	Stomach	BM	Hematemesis	Ope
6	Presentcase	58/F	Stomach	EM	Dispepsia	GIF

EM: Epithelioid mesothelioma, SM: Sarcomatoid mesothelioma, BM: Biphasic mesothelioma, CE: Capsule endoscopy, DBE: Double ballon endoscopy, GIF: Gastrointestinal fber Ope: Operation

DISCUSSION

About 90–95% of mesotheliomas arise in the pleural cavity and 5–10% in the peritoneal cavity. Rarely, mesothelioma originates from the pericardium and tunica vaginalis.[2] However, malignant neoplasm can be locally or rarely occurrence of distant haematogenous metastasis in several different organs. Tumor tend to spread through the pleural cavity and to the chest wall, as well as to the regional lymph nodes in the axillary and supraclavicular area.[3] Distant organ metastases by hematogenous spread occur in more than 10% of cases at late stage of the disease.[4] In contrast, distant metastases from malignant pleural mesothelioma appear relatively late and are frequently discovered at autopsy. The most frequent involved organs are liver, adrenal gland, kidney and contralateral lung. Extensive abdominal involvement is discovered at autopsy in one third of cases. However, involvement of the gastrointestinal lumen alone is exceptional.

MPM predominantly affects men over the age of 50 year (male/female ratio, 3:1). Symptoms are commonly due to local invasion of pleural and mediastinal structures. Chest pain, shortened of breath and cough are the mostly seen presenting symptoms. As reported in 2001, MPM has an extremely poor prognosis (Median post-diagnosis survival time is of <1 year and a 5-year survival rate of <1%).[5] As a result of combination chemotherapy with a platinum agent, antifolates and gemcitabine, the median overall survival has been improved, ranging between 9 and 17 months.[6]

Histologically, mesothelioma is divided into different subgroups including epithelioid, sarcomatoid and mixed or biphasic. According to the study by Fusco et al., the prognosis of epithelial subtype comparing to sarcomatoid variant is significantly improved.[7] The primary diagnosis of our case was epithelioid type, the diagnosis of gastric metastasis was revealed as epithelioid type.

In the literature review, two duodenal, one jejuno-ileal, one jejunal, one gastric metastasis have been reported and all were symptomatically (Table 1).[1,8-10] In our case, MPM metastasis was detected in the biopsy material of the excised polypoid lesion.

Diagnostic criteria for localized malignant mesothelioma[11] including (i) radiological, surgical, or pathological evidence of a localized serosal/subserosal (not organ-centered) tumor mass without evidence of diffuse serosal spread; and (ii) a microscopic pattern similar to that found in diffuse malignant mesothelioma. In our case, at the time of diagnosis it was revealed as diffuse malignant mesothelioma, however, the recurrence form was appeared as local lesion of chest wall and pleural lesion which was diagnosed radiologically. During endoscopic examination polypoid gastric metastasis was detected incidentally. However, no remarkable finding of metastasis was observed in radiologic imaging, it was excised endoscopically and the pathologic diagnosis was done.

In imaging of malignant mesothelioma, CT is primary imaging modality and has a major role in evaluation and staging specially for calcification assessment. On the other hand, magnetic resonance imaging (MRI) and furthermore, positron emission tomography (PET) provide additional important diagnostic information about lesion extension, especially to the chest wall and diaphragm, as well as its characterization. Also PET can provide both anatomic and metabolic data, particularly in those cases of extra thoracic and mediastinal nodal metastases.[12]

Isolated high level of CRP and leukocytes usually has association with inflammatory disease such as bacterial infection. However, leukocytosis sometimes may accompany malignant neoplasms with a high granulocyte colony stimulating factor (G-CSF) level in the absence of infection.[13,14] Katoh et al. showed that the production of G-CSF is the most potent and common cause of tumor-induced leukocytosis.[14] In our case, there was elevation of CRP (50 mg/dl) and leukocytes level (40.000 cells/μl) without presence of infection. Owing to the exitus of the patient, G-CSF level was not measured. Regarding to reported data in literature, tumor cells maybe responsible for the production of the excessive G-CSF.[13,14] Despite absent

G-CSF level in our case, we believe in that CRP elevation and leukocytosis was related to tumoral lesion and its G-CSF production.

REFERENCES

1. Hayashi H, Notohara K, Yoshioka H, et al. Localized malignant pleural mesothelioma showing a thorasic mass and matastasizing to the somach. Inter Med. 2010;49:671-5.
2. Hammar SP. Macroscopic, histologic, histochemical, immunohistochemical,and ultrastructural features of mesothelioma.Ultrastruct Pathol. 2006;30:3-17.
3. Cedrés S, FariñasI L, Stejpanovic N, et al. Bone metastases with nerve root compression as a late complication in patient with epithelial pleural mesothelioma. J Thorac Dis. 2013;5:35-7.
4. Sussman J, Rosai J. Lymph node metastasis as the initial manifestation of malignant mesothelioma. Report of six cases. Am J Surg Pathol. 1990;14:819-28.
5. Ceresoli GL, Locati LD, Ferreri AJ, et al. Therapeutic outcome according to histologic subtype in 121 patients with malignant pleural mesothelioma. Lung Cancer. 2001;34:279-87.
6. Tsao AS, Wistuba I, Roth JA, Kindler HL.Malignant pleural mesothelioma. J Clin Oncol. 2009;27:2081-90.
7. Fusco V, Ardizzoni A, Merlo F, et al. Malignant pleural mesothelioma. Multivariate analysis of prognostic factors on 113 patients. Anticancer Research. 1993;13:683-9.
8. Law MR, Hodson ME, Heard BE. Malignant mesothelioma of the pleura: relation between histological type and clinical behavior. Thorax. 1982;37: 810-15
9. Soltermann A, Pache JC, Vogt P. Metastasis of a pleural mesothelioma to a hyperplastic stomach polyp: an increase of vimentin expression is seen during a gain in deciduoid morphology. Rare Tumors. 2011; 3: e52.
10. Chen H, Tsai K.B, Wang CS, Hsieh TJ, Hsu JS. Duodenal Metastasis of Malignant Pleural Mesothelioma. J Formos Med Assoc. 2008;107:961-4.
11. Allen TC, Cagle PT, Churg AM, et al. Localized malignant mesothelioma. Am J Surg Pathol. 2005;29:866-73.
12. Wang ZJ, Reddy GP, Gotway MB, et al. Malignant Pleural Mesothelioma: Evaluation with CT,MR Imaging, and PET. Radiographics. 2004;24:105-19.
13. Yoshimoto A, Kasahara K, Saito K, Fujimura M, Nakao S. Granulocyte colony-stimulating factor-producing malignant pleural mesothelioma with the expression of other cytokines. Int J Clin Oncol. 2005;10:58-62.
14. Usami M,Uchiyama M, Kawaguchi K, et al. Granulocyte Colony-Stimulating Factor-Producing Malignant Pleural Mesothelioma. J Thorac Oncol. 2007;2:257-8.

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