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Case Report

A rare case of gastric leiomyosarcoma metastasizing to liver and lymph node

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Abstract

Before the advent of specific immunohistochemistry (IHC), many Gastrointestinal Stromal Tumors (GISTs) were misdiagnosed as Leiomyosarcoma (LMS) of the gastrointestinal tract (GIT), given their similar morphology. However, its true incidence is extremely low with only a few reported cases of Leiomyosarcoma of the stomach in literature. Here, we present a rare case of LMS of the stomach with metastasis to the liver and lymph node, discussing the diagnostic challenges faced, based on its clinical, cytological, and histomorphological features.

Keywords: Leiomyosarcoma, Stomach, GIST, Immunohistochemistry, Liver, Lymph node, Metastasis.

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1. Introduction

Only a handful of stromal cancers of the GIT have been diagnosed as Leiomyosarcoma (LMS), with gastric LMS being extremely rare in the post-Kit era.[1] The challenge in diagnosis arises as the morphological features of LMS are similar to the other mesenchymal tumors of the GIT, but differ in treatment modalities and prognosis. This problem arises especially with GIST, a more common soft tissue neoplasm of the GIT.

We present a highly unusual case of Leiomyosarcoma of the Stomach with metastatic nodules to the liver and lymph node, and the diagnostic difficulties associated at different levels including cytology and histopathology.

2. Case Report

A 52-year-old female presented with pain abdomen, vomiting, weakness, and weight loss for 5 months. There was no significant past or medical history. On examination, the patient had pallor and hypotension with an irregular epigastric mass up to the right and left hypochondria. CT scan showed a circumferential mass measuring 5.2 x 2.5 cm involving predominantly the body of the stomach, posterior wall extending to cardia and fundus, along with multiple ill-defined lesions in the enlarged liver and a single nodal mass measuring 2.9 c.m. across, along the celiac axis focally infiltrating the inferior vena cava (IVC), suggesting a primary stomach malignancy (as it was the largest focus of tumor) with liver and lymph node metastasis [**Figure 1**A and B]. Symptomatic management and blood transfusion were done for her complaints and her condition stabilized.

Upper gastrointestinal endoscopy revealed a protruded lesion in the stomach and biopsies were taken [**Figure 1**C]. Histopathological examination (HPE) showed only a small focus of spindle cells in the background of chronic gastritis giving the unclear impression- suspicious for spindle cell lesion [**Figure 2**A]. Consequently, fine needle aspiration cytology (FNAC) liver was attempted which showed sheets of spindle cells with nuclear hyperchromasia and eosinophilic cytoplasm confirming the diagnosis: malignant spindle cell tumor [**Figure 2**B]. Next, a liver ultrasound-guided biopsy was done; HPE of the biopsy on microscopy

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showed circumscribed nodules of spindle cells with frequent mitoses (12 mitoses/10HPF) and no necrosis [**Figure 2**C], and an IHC workup of High-grade sarcoma with the differentials: GIST, LMS, malignant solitary fibrous tumor (SFT), and fibrosarcoma was done. These tumor cells were positive for SMA and Desmin and negative for CD117 and CD34 with a high Ki67 index of ~80% [**Figure 2**D, E, and F].

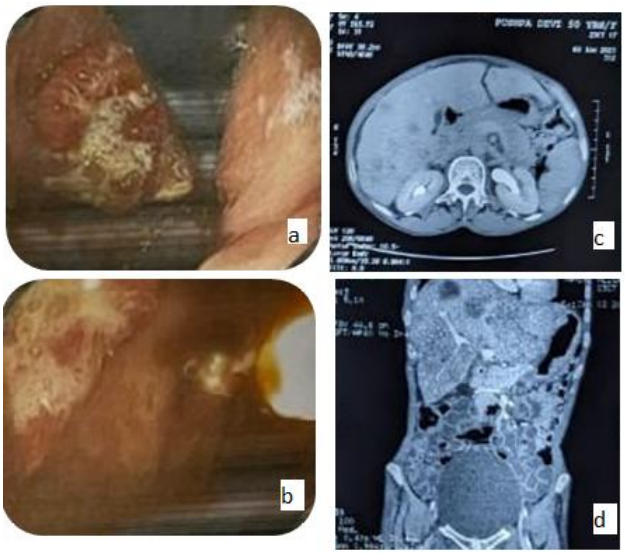

Figure 1: (**a** and **b**) Upper GI endoscopy showing a nodular lesion protruding into the lumen of stomach adjoining the GE junction; (**c** and **d**) CT scan showing heterogeneous mass involving body of stomach with multiple lesions involving both lobes of liver and a nodal mass along celiac axis focally infiltrating IVC.

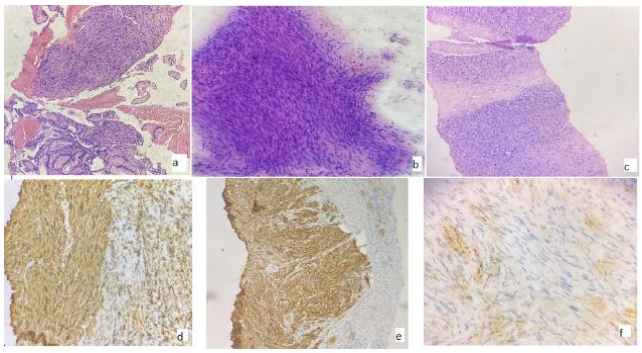

Figure 2: Microscopy (**a**): UGI biopsy shows a focus of spindle cells adjacent to gastric mucosa, H and E stain, 10X; (**b**): Liver FNAC show sheets of pleomorphic spindle cells with nuclear hyperchromasia, PAP stain, 40X; (**c**): Liver biopsy shows nodule of malignant spindle cells below separated from hepatic parenchyma above by a band of fibrosis, H and E stain, 10X; (**d**): Desmin IHC positive in tumor cell, 10x; (**e**): SMA positive in tumor cells, 4X (f) CD117 negative in tumor cells, 40X

Based on these histomorphological features, a histopathological diagnosis of Leiomyosarcoma, FNCLCC grade 2 was given. Finally, based on the CT findings and the stomach and liver biopsy, an overall diagnosis of LMS of the stomach with metastasis to the liver and celiac lymph node was made. The patient was deemed inoperable due to the extensive tumor burden and started chemotherapy (combination of Doxorubicin (45mg), Cisplatin (30mg), and Ifosfamide 1200mg). She was given four cycles of chemotherapy, and passed away at 5 months post diagnosis.

3. Discussion

Since the discovery of C-Kit mutation associated with GISTs, the misdiagnosis of LMS in the stomach has markedly reduced, and now accounts for barely 1% of all gastric malignancies.[2,3] Mere sixteen cases of LMS stomach have been reported in English literature in the post-KIT era, to our knowledge.[3,4,5]

LMS of the stomach generally occurs in elderly with no gender predilection.[5] Patients usually present late with non-specific symptoms, owing to the tumor's predominant extra luminal growth.[7]

Radiological imaging, can facilitate the diagnosis of LMS; however, definitive diagnosis is only possible through tissue biopsy. Our patient's CT abdomen showed a large stomach malignancy with multiple smaller metastatic lesions in the liver and a single nodal mass along the celiac axis focally infiltrating the IVC [Figure 1A]. Metastasis is common in LMS with around 40% presenting with metastasis at the time of diagnosis and lung and liver being the frequent sites. However, in soft tissue tumors, including LMS, lymph node metastasis is rare.[8] Kang et al studied 14 cases of LMS stomach, followed by two other cases being reported later, and none of them exhibited lymph node metastasis.[3,4,5] Two cases of nodal metastases of LMS of other sites have been reported, including a similar case of metastatic lymph node mass infiltrating IVC from a primary adrenal LMS.[8,9]

As LMS arises submucosally, its diagnosis depends on a deeper biopsy. The conventional endoscopic biopsy attempted on our patient also failed to get sufficient deep tissue of the tumor. Subsequently, a liver FNAC and biopsy were done. On microscopy, a spindle cell neoplasm with IHC SMA and Desmin positive and CD117 and CD34 negative was reported, supporting LMS and ruling out GIST (CD117 and CD34 positive), SFT (CD34 positive), Fibrosarcoma (all markers negative). SMA, Desmin, and h-Caldesmon are expressed in more than 70% of cases of LMS while GIST should be positive for any two of CD117, CD34, and Dog-1 markers.[5]

Being a rare entity, the treatment recommendations for metastatic LMS is not yet established. Complete surgical resection in combination with chemotherapy is the preferred option.[9] European Society for Medical Oncology (ESMO)

and the National Comprehensive Cancer Network (NCCN) treatment guidelines recommend anthracycline-based chemotherapy, for soft tissue sarcomas.

4. Conclusion

This is a rare case of gastric LMS with liver and lymph node metastases. The morphological similarity of gastric LMS to other gastrointestinal stromal tumors requires extensive histopathological examination with immunohistochemistry analysis. Because of its rarity, the findings of this case as well as the analysis of additional cases will help us better understand its presentation, prognosis, and outcome.

5. Source of Funding

None.

6. Conflict of Interest

None.

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