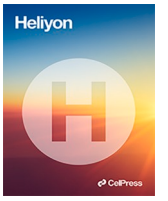
Heliyon 10 (2024) e30410

Contents lists available at ScienceDirect

Heliyon

journal homepage: www.cell.com/heliyon



Case report

Laparoscopy-assisted trans gastric wedge resection: A safe treatment for gastric pyloric liposarcoma: A case report and literature review

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ARTICLE INFO

Keywords:
Gastric tumor
Case report
Liposarcoma
Stomach
Minimally-invasive surgery

ABSTRACT

Background: Gastric liposarcoma (GL) is extremely rare and radical surgery has been the conventional treatment, even in small tumors. Laparoscopic wedge resection has been reported worldwide for subepithelial tumors of the stomach.
Case presentation: The patient was an asymptomatic 63-year-old man presenting with a subepithelial gastric tumor. The esophagogastroduodenoscopy showed a 3 cm ulcerated soft tumor located in the posterior wall of the antrum just above the pylorus. Two preoperative biopsies were performed with a negative result for malignant neoplasm. Dynamic computerized tomography revealed 35 × 35 mm well-defined pyloric mass with fat density. Despite the difficult location of the tumor, function-preserving surgery was performed. Surgery was initiated by a laparoscopic approach with four trocars. After the dissection of the greater omentum, the greater curvature and the posterior wall of the stomach were exposed. A gastrostomy was performed in the anterior wall of the antrum. Due to the difficulty in identifying the tumor location, a mini-laparotomy was conducted. After assessing the pylorus and section parameters, the tumor was extracted by gastrostomy and resected with a linear stapler. The patient was discharged after five days with no complications. The histological diagnosis was a well-differentiated liposarcoma. Resection margins were clear. The tumor cells tested negative for MDM2. No adjuvant therapy was indicated. The patient is alive without recurrence.
Conclusions: Despite its rarity, gastric liposarcoma should be respected for differential diagnosis in submucosal tumors. The main diagnostic method is histological, and surgery is the conventional treatment without yet having a consensus. Minimally invasive wedge resection might be a suitable treatment even if the location is close to the pylorus. Multicenter studies are required to obtain better results in the management of this pathology.

1. Background

Soft tissue sarcomas account for <1 % of all malignancies [1]. Most of them are located in the extremities, especially in the thigh, and intraabdominal location is uncommon [2]. Liposarcoma is one of the most frequent mesenchymal neoplasms in adults and, according to the WHO tumor classification, liposarcomas are categorized into five histological subtypes [3]. Less than 45 cases of gastric

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https://doi.org/10.1016/j.heliyon.2024.e30410
Received 13 December 2022; Received in revised form 24 April 2024; Accepted 25 April 2024
Available online 27 April 2024

liposarcoma have been reported in English literature, which denotes its extreme rarity. These tumors regularly originate in the submucosal layer and are usually located in the lesser curvature of the gastric antrum. Gastric liposarcoma presents nonspecific clinical features, although small tumors are usually asymptomatic. Larger tumors can produce bleeding and gastrointestinal (GI) obstruction, the latter generally those tumors located adjacent to the pylorus [4]. Upper GI endoscopy and abdominal computed tomography (CT) are the primary diagnostic studies. Generally, an endoscopic biopsy is inconclusive, but it must be performed for precise therapeutic planning. Given the limited number of cases, there is no guidance for the management of this disease. Surgical resection with free margins is the primary treatment, however, function-preserving surgery is challenging to achieve [5]. Adjuvant chemotherapy and/or radiotherapy are debatable [6]. The prognosis is established by complete surgical resection and histological subtype. We present an infrequent case of gastric liposarcoma treated with function-preserving surgery and achieving free surgical margins despite its complex location.

1.1. Case presentation

A 63-year-old man with a medical history of diabetes mellitus treated with metformin and, benign prostatic hypertrophy treated with finasteride and terazosin was referred to our hospital with a submucosal gastric tumor of undetermined etiology. The patient did not present specific symptoms. The vital signs were stable and on physical examination, the abdomen was soft, non-tender, non-distended, and without a palpable mass. The screening esophagogastroduodenoscopy (EGD) revealed a 3 cm ulcerated soft tumor in the greater curvature and the posterior wall of the gastric antrum just above the pylorus (Fig. 1A). The pillow or cushion sign was found by applying pressure using closed forceps (Fig. 1B). Two preoperative biopsies were performed, the first at the ulceration area and the second guided by endoscopic ultrasonography (EUS), both were reported as chronic gastritis. Dynamic computerized tomography (CT) showed a 35 × 35 mm subepithelial well-defined mass in the posterior wall of the antrum with fat density and high attenuation. [Fig. 2 (A, B)]. Tumor markers (CEA and CA 19-9) were within normal values. Due to the presence of ulceration, our main diagnosis was liposarcoma, followed by lipoma and gastrointestinal stromal tumor (GIST). Surgical treatment was decided upon due to the complex location of the tumor and the high risk of duodenal obstruction. The patient agrees to surgical treatment and the laparoscopic approach was selected. A 10 mm trocar was placed on the umbilical level, two 12 mm and 5 mm trocars were placed on the right flank, and a single 5 mm trocar on the left flank. The greater omentum was dissected and the omentum transcavity was approached. The infrapyloric area was dissected to approach the greater curvature and the posterior wall of the stomach. Gastrostomy was performed in the proximal antrum. Because of the difficulties of palpation and identification of the tumor, a mini laparotomy was done on the epigastrium. Under direct palpation, the location of the tumor was identified, which was on the posterior wall of the antrum close to the pylorus. The tumor was pulled out through the gastrostomy site and resected with a linear stapler after evaluation of the pylorus and resection margins. The gastrostomy was closed with continuous manual suture. Upper gastrointestinal endoscopy was performed on day 3 postoperative and no evidence of bleeding or leakage was observed. The patient was discharged after five days with no complications. The histological diagnosis was a large 5.0 × 3.4 cm well-differentiated liposarcoma. [Fig. 3 (A, B)]. The mitotic count was less than 1/10 HPF and the tumor was confined to the submucosa. There was no sign of necrosis or angiolymphatic invasion. Resection margins were clear with a safe microscopic margin greater than 10mm. Immunohistochemical studies revealed that tumor cells were positive for S-100 and negative for MDM2. No adjuvant treatment was indicated. Surveillance was carried out every 3 months. After 20 months of follow-up, no recurrence was evidenced in images or endoscopic studies.

Informed consent was obtained for the publication of this case report along with the images of the case. Likewise, this study was accepted by Institutional Review Board (IRB) of the National Cancer Center, Korea (NCC2022-0248).

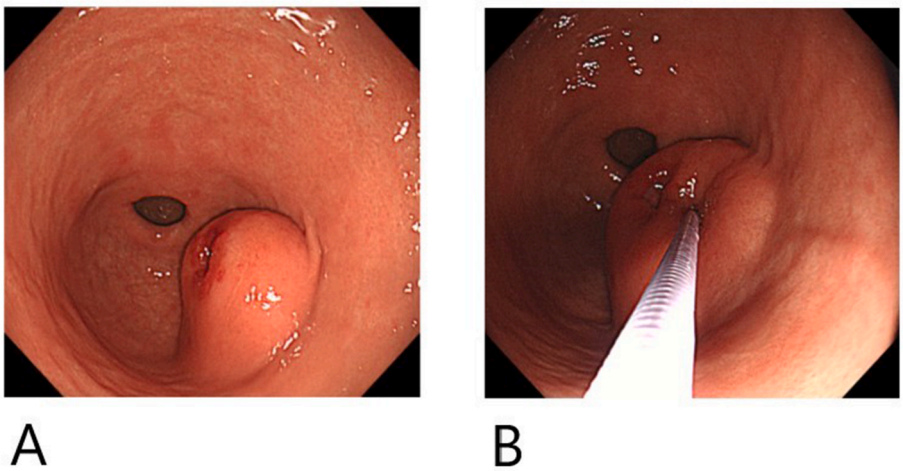

Fig. 1. Esophagogastroduodenoscopy (EGD): **(A)** Submucosal soft tumor located in the greater curvature and the posterior wall of the antrum adjacent to the pylorus; **(B)** Pillow or cushion sign: Indentation of the tumor surface evidenced under the pressure of cold endoscopic forceps.

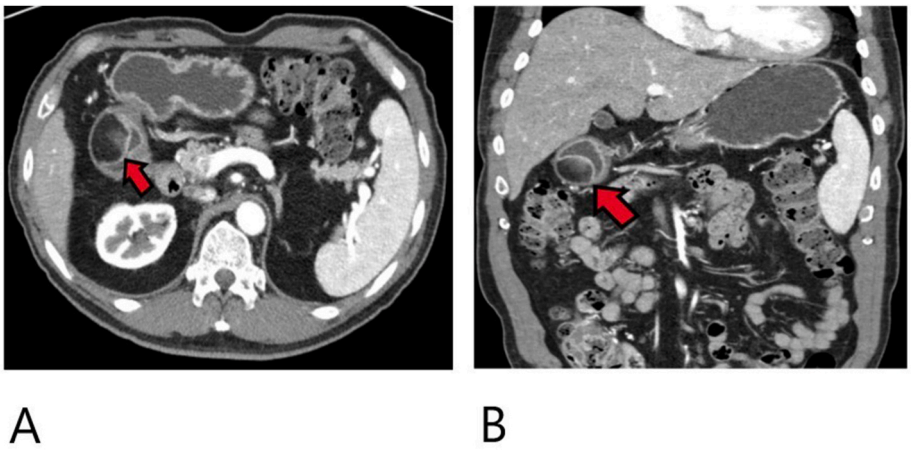

Fig. 2. Dynamic computerized tomography (CT): Subepithelial tumor located in the gastric antrum in close contact with the pylorus. Fat density and high attenuation are evident. The red arrow indicates the tumor. **(A)** Axial view; **(B)** Coronal view. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

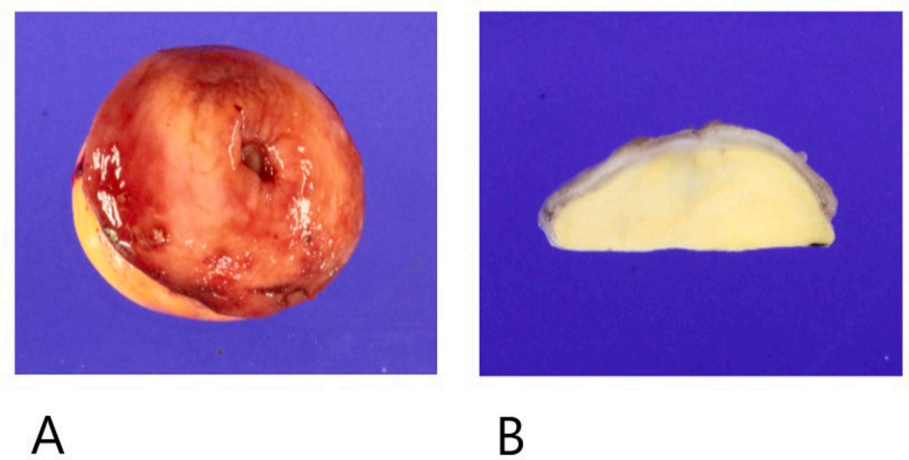

Fig. 3. Well-differentiated liposarcoma. Surgical Specimen (trans gastric wedge resection): Well-defined submucosal tumor composed mainly of fatty tissue with central ulceration located in the gastric antrum. **(A)** Frontal view; **(B)** Transected tumor.

2. Discussion

Gastric liposarcoma was first reported in 1941 by Abrama and Tuberville, since that publication less than 45 cases have been reported over the years [7]. Likewise, soft tissue sarcomas present a low incidence of approximately 50–60 cases per million people and represents 1 % of malignancies [8]. More than 80 sarcoma subtypes have been described and this wide diversity is due to continuous advances in molecular diagnosis [3]. Liposarcoma is the most frequent histological subtype of soft tissue sarcomas, accounting for 50 % and 25 % of retroperitoneal and extremity sarcomas, respectively [6,9]. In histological terms, liposarcomas are divided into 5 subtypes: well-differentiated, myxoid, pleomorphic, mixed-pleomorphic, and dedifferentiated [3]. Well-differentiated liposarcoma (WDLPS) or atypical lipomatous tumor (ALP), is a locally aggressive neoplasm that occurs during the fourth to fifth decade of life. This pathology represents approximately 40–45 % of all liposarcomas and is composed of an atypical proliferation of adipocytes and focal nuclear atypia in both, adipocytes and stromal cells [3]. The mixed subtype is the second most prevalent liposarcoma in adults and frequently occurs in children and adolescents. It presents a myxoid stroma and similarly, round to ovoid cells with a minimum of lipoblasts [4]. Less than 5 % of liposarcomas are of the pleomorphic type, this subtype exhibits a variable number of pleomorphic lipoblasts and denotes a high-grade sarcoma [10]. Usually, dedifferentiated liposarcoma originates from recurrent WDLPS. A transition from an atypical lipomatous tumor to a non-lipogenic high-grade sarcoma is evident in this pathology and, like WDLPS, immunopositivity for CDK4 and MDM2 is used for diagnostic support, although is not confirmatory [11].

Approximately, 30 % of gastric liposarcomas are located in the antrum, especially in the lesser curvature, followed by the gastric fundus, body, and esophagogastric junction [5]. Symptoms are closely related to the tumor size. In small tumors ($\leq$4 cm), as in the

present case, the symptoms are nonspecific, such as diarrhea, abdominal pain, and weight loss [12]. Larger (>4 cm) tumors can present symptoms related to gastrointestinal bleeding, particularly those that graze against the contralateral gastric wall and subsequently ulcerate. Although our patient did not present any symptoms, it could have been complicated by an endoluminal obstruction in the pylorus due to the size and location [4].

EGD presents well-defined, yellowish, submucosal masses that share similar characteristics to gastric lipomas. Likewise, lipomas show typical endoscopic signs such as naked fat, tenting, cushion, or pillow sign [13,14]. It is important to mention that our patient presented the pillow or cushion sign even though it is a malignant tumor, which implies that this finding is not exclusive to lipomas [15]. Computed tomography (CT) allows us to assess the tumor characteristics as well as the possibility of metastatic disease. WDLPS presents as heterogeneous masses with fatty density and is associated with enhanced areas [16]. However, the mixed subtype is related to cystic areas and enhancement in solid components. Furthermore, the finding of hemorrhagic areas and necrosis suggests dedifferentiated liposarcoma [17].

MDM2 and CDK4 immunostainings are typically positive for WDLPS and dedifferentiated liposarcoma (DDLPS) [18]. The amplification of MDM2 is a consequence of the amplification of chromosome 12q13-15, especially DDLPS. The proteasomal degradation of p53 is regulated by MDM2. It has been shown that patients with MDM2 amplification have a decrease in p53 activity, thus presenting a higher recurrence rate and shorter survival [19]. Likewise, it has been reported that patients with retroperitoneal WDLPS who presented CDK-4 amplification in Q-PCR testing had a higher postoperative recurrence rate [20].

Gastric submucosal tumors (SMT) encompass multiple etiologies and for this reason the differential diagnosis is broad and complex. The clinical presentation of the different SMTs is similar and even asymptomatic. Among SMTs of mesenchymal origin, GIST is the most frequent diagnosis but leiomyoma, schwannoma, lipomas and liposarcomas have also been reported. NETs and melanomas are epithelial tumors that are also mentioned as differential diagnoses of SMT [21,22]. Likewise, there have been reports of patients with early gastric cancer who were treated with endoscopic resection who were presented with a typical submucosal tumor [23]. In the specific case of WDLPS, the main differential diagnosis is gastric lipoma since its endoscopic and tomographic characteristics are similar. Given this likeness, conservative surgery should be attempted due to the possibility of benignity.

A complete surgical resection achieving free margins (RO resection) is related to a better prognosis without requiring lymphadenectomy [21]. At the moment, there is no specific surgical technique for the management of this disease; however, most cases were conventionally treated with standard gastrectomy, this being chosen according to the location of the tumor. Most studies report radical surgeries in which the preservation of healthy parenchyma is scarce, even in small tumors (≤4 cm) [5]. Only 43 cases have been reported in the English literature, including 40 patients who received radical surgery (standard gastrectomy). The type of surgery was not described in two patients, and only one treated with endoscopic submucosal dissection (ESD) achieved free surgical margins [24,25]. The main differences between the surgical technique that we present and the conventional surgeries performed in the literature (proximal gastrectomy, distal gastrectomy and total gastrectomy) are the possibility of completely resecting the tumor while preserving the gastric parenchyma and achieving free surgical margins; it also allows improving the quality of life by reducing the possibility of iron deficiency anemia, B12 deficiency, malabsorption syndrome and postgastrectomy syndromes (dumping syndrome, alkaline gastritis, etc.). Compared to the endoscopic procedures already reported, the technique performed in this case allows us to reduce the possibility of presenting positive surgical margins given the greater safety when resecting the tumor under direct palpation. It is important to mention that endoscopic procedures for the resection of sub mucosal tumors also present complications such as bleeding and gastric perforation as the most severe.

Multiple studies have evaluated risk factors related to recurrence in patients diagnosed with liposarcoma, especially those with retroperitoneal location [26–28]. Knebel et al. evaluated 133 patients with a diagnosis of liposarcoma but with different locations; In this study, the histological type, tumor differentiation, tumor size >5 cm and R1 margins were reported to be related to lower disease-specific survival [26]. In a retrospective study conducted in China, 138 patients with recurrent retroperitoneal liposarcoma were evaluated. In the multivariate analysis, R2 resection margin, tumor size >18 cm, histological subtype (myxoid cell liposarcoma, DDLPS), pathological differentiation and local recurrence were related to a lower 5-year overall survival [27].

However, our procedure is not devoid of limitations. First, since this is a function-preserving surgery, the radical nature of the procedure could be minor; However, follow-up of our patient does not show disease recurrence, which demonstrates its safety. Second, lymphadenectomy was not performed in this case, although the literature mentions that it is probably not necessary, we believe that it is important to explore the possibility of lymphatic metastasis in gastric liposarcoma.

Herein, we report a patient with gastric liposarcoma with a complex location successfully treated with minimally-invasive surgery and achieving preservation of gastric parenchyma. Despite the difficult tumor position, from our perspective, this surgical approach was safe and will allow a better quality of life compared to radical surgery.

3. Conclusion

Despite its rarity, gastric liposarcoma should be respected for differential diagnosis in submucosal tumors. The main diagnostic method is histological, and surgery is the conventional treatment without yet having a consensus. Minimally invasive wedge resection might be a suitable treatment even if the location is close to the pylorus. Multicenter studies are required to obtain better results in the management of this pathology.

Data availability statement

The authors confirm that the data reported in this study are available in the article.

CRediT authorship contribution statement

Oscar Paredes: Writing – review & editing, Writing – original draft, Methodology, Investigation. **Sang Soo Eom:** Writing – review & editing, Visualization, Validation, Supervision, Methodology, Investigation. **Sin Hye Park:** Writing – review & editing, Validation, Supervision, Methodology, Investigation, Conceptualization. **Young-Woo Kim:** Writing – review & editing, Visualization, Validation, Supervision, Resources, Project administration, Methodology, Investigation, Funding acquisition, Conceptualization.

Declaration of competing interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests:Young-Woo Kim reports was provided by National Cancer Center, Korea. Young-Woo Kim reports a relationship with National Cancer Center Korea that includes: employment and funding grants. If there are other authors, they declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

The authors would like to thank all the members of the Center for Gastric Cancer of the National Cancer Center, Goyang, Korea.

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