

World Journal of *Clinical Cases*

World J Clin Cases 2020 April 6; 8(7): 1188-1342





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The WJCC is now indexed in PubMed, PubMed Central, Science Citation Index Expanded (also known as SciSearch®), and Journal Citation Reports/Science Edition. The 2019 Edition of Journal Citation Reports cites the 2018 impact factor for WJCC as 1.153 (5-year impact factor: N/A), ranking WJCC as 99 among 160 journals in Medicine, General and Internal (quartile in category Q3).

RESPONSIBLE EDITORS FOR THIS ISSUE

Responsible Electronic Editor: *Yan-Xia Xing*

Proofing Production Department Director: *Yun-Xiaojuan Wu*

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Semimonthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Bao-Gan Peng, Sandro Vento

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

EDITORIAL OFFICE

Jin-Lei Wang, Director

PUBLICATION DATE

April 6, 2020

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PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Scaphoid metastasis as the first sign of occult gastroesophageal junction cancer: A case report

Yu-Jie Zhang, Yan-Yan Wang, Qi Yang, Jian-Bing Li

ORCID number: Yu-Jie Zhang (0000-0001-8164-6425); Yan-Yan Wang (0000-0002-9074-9706); Qi Yang (0000-0002-8149-2937); Jian-Bing Li (0000-0001-5409-4805).

Author contributions: Zhang YJ and Li JB were the patient's orthopedists, reviewed the literature, and wrote the manuscript; Wang YY and Yang Q were responsible for providing professional guidance and collecting the medical imaging materials; Li JB revised the manuscript; all authors issued final approval for the version to be submitted.

Supported by Natural Science Foundation of Zhejiang Province, No. LQ19H060002 and No. LQ19H160041; Medical and Health Science and Technology Project of Zhejiang Province, No. 2018KY089.

Informed consent statement: Written informed consent was obtained from the patient and his wife for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016)

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in

Yu-Jie Zhang, Jian-Bing Li, Department of Orthopedic Surgery, The Second Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310009, Zhejiang Province, China

Yan-Yan Wang, Department of Oncology Surgery, The Second Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310009, Zhejiang Province, China

Qi Yang, Department of Pathology, The Second Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310009, Zhejiang Province, China

Corresponding author: Jian-Bing Li, MD, Doctor, Department of Orthopedic Surgery, The Second Affiliated Hospital, Zhejiang University School of Medicine, 88 Jiefang Road, Hangzhou 310009, Zhejiang Province, China. 2505002@zju.edu.cn

Abstract

BACKGROUND

Metastatic tumors of the hand are very rare. They are usually late manifestations of massive advanced malignancies. However, a small portion of acrometastases are indications of occult primary cancer. Here, we report an extremely rare case in which a scaphoid bone lesion was the initial manifestation and was found to be a metastasis from gastroesophageal junction (GEJ) cancer.

CASE SUMMARY

A 57-year-old male patient presented with ongoing left wrist pain and swelling after trauma. He was initially misdiagnosed with infection of the scaphoid bone and treated with antibiotics and anti-inflammatory drugs. Further radiographic investigations showed a scaphoid pathological fracture, indicating a metastatic tumor derived from the GEJ and/or right lung malignancies. Gastroscopy failed to identify the pathology of the mass at the GEJ, which grew in an exophytic pattern. A lung puncture biopsy was not performed because the patient refused the procedure. To relieve his wrist pain and obtain a definite pathology, we resected the scaphoid lesion. Based on the clinical and pathological results, the patient was finally diagnosed with multiple metastases of advanced GEJ adenocarcinoma. He underwent chemotherapy and died 6 mo after his initial presentation.

CONCLUSION

Despite the rareness of the disease, orthopedic surgeons should consider the possibility of metastasis to the bones of the hand when patients complain of persistent and progressive pain in the hand.

Key words: Hand metastasis; Scaphoid; Gastroesophageal junction cancer; Diagnosis;

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Manuscript source: Unsolicited Manuscript

Received: December 20, 2019

Peer-review started: December 20, 2019

First decision: January 7, 2020

Revised: January 13, 2020

Accepted: March 9, 2020

Article in press: March 9, 2020

Published online: April 6, 2020

P-Reviewer: Kikuchi H, Streba LAM

S-Editor: Wang YQ

L-Editor: Wang TQ

E-Editor: Ma YJ



Case report

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Core tip: Hand metastasis is very rare, especially when it is the first sign of an occult primary malignancy. Here, we report a case of a scaphoid bone lesion that had metastasized from a gastroesophageal junction adenocarcinoma; the initial manifestation was persistent wrist pain and swelling. Due to its low incidence and the lack of specific symptoms, hand bone metastasis is easily misdiagnosed, leading to delayed treatment. A review of the literature provides a systematic understanding of the epidemiology, manifestations, diagnosis, treatment, and prognosis of metastatic carcinomas of the hand. Moreover, the early detection and diagnosis of hand metastasis are needed to improve patients' quality of life and prolong their survival.

Citation: Zhang YJ, Wang YY, Yang Q, Li JB. Scaphoid metastasis as the first sign of occult gastroesophageal junction cancer: A case report. *World J Clin Cases* 2020; 8(7): 1287-1294
URL: <https://www.wjnet.com/2307-8960/full/v8/i7/1287.htm>
DOI: <https://dx.doi.org/10.12998/wjcc.v8.i7.1287>

INTRODUCTION

Metastases to the hand bones are rarely encountered in clinical practice and comprise approximately 0.1% of all osseous metastases^[1]. They can be misdiagnosed as rheumatic or infectious diseases due to the lack of specific symptoms. Usually, they are late manifestations of various advanced malignancies; however, in 16% of the affected patients, they are the initial manifestation of an occult cancer^[2]. Here, we report an extremely rare case of a scaphoid bone lesion that was found to be a metastasis from a gastroesophageal junction (GEJ) adenocarcinoma.

CASE PRESENTATION

Chief complaint

A 57-year-old male patient presented with persistent left wrist pain and swelling 2 mo after trauma.

History of present illness

Two months prior to this visit, the patient presented with ongoing left wrist pain and swelling after trauma. Based on an initial computed tomography (CT) scan, the patient was diagnosed with an infection of the scaphoid bone and treated with antibiotics, anti-inflammatory drugs, and rest at a local hospital. However, the pain continued to worsen, and the motion of the wrist was restricted.

History of past illness

The patient had undergone acupotomy therapy several months before the onset of the disease. He had a history of cerebral infarction and intestinal polyps.

Personal and family history

The patient was a regular drinker and heavy cigarette smoker with no relevant family history.

Physical examination upon admission

Physical examination showed left wrist tenderness, swelling, and restriction of motion with regard to radial and ulnar deviation. The swollen area was neither warm nor erythematous.

Laboratory examinations

The C-reactive protein level (49 mg/L) and the erythrocyte sedimentation rate (33 mm/h) were elevated, and the levels of tumor markers, such as carbohydrate antigen (CA)-125 (390 U/mL), carcinoembryonic antigen (CEA) (8 ng/mL), and CA211 (5.4 ng/mL), were significantly elevated.

Imaging examinations

Plain radiographs showed scaphoid bone density reduction and fracture (Figure 1A). CT and magnetic resonance imaging (MRI) scans revealed scaphoid and triangular bone destruction and soft tissue swelling, indicating a pathological fracture (Figure 1B and C). A subsequent chest CT scan suggested a lower esophageal and stomachus cardiacus mass, a right lung mass, and the destruction of multiple ribs (Figure 2A and B). We then performed positron emission tomography-CT with 2-¹⁸F fluoro-2 deoxy-D-glucose that led to a strong suspicion of GEJ and/or right lung malignancies with metastases in multiple locations, including the left lung, lymph nodes, ribs, and scapula (Figure 3). A subsequent gastroscopy did not show any space-occupying lesions (Figure 4), and the biopsy showed only chronic superficial gastritis. A CT-guided percutaneous lung puncture biopsy was refused by the patient. Pathology of the left wrist showed scaphoid bone metastasis of poorly differentiated adenocarcinoma. Immunohistochemistry showed that the sample was positive for caudal type homeobox 2 (CDX2) and negative for thyroid transcription factor 1 (TTF-1) and ALK-Lung (Figure 5A).

FINAL DIAGNOSIS

The patient was diagnosed with GEJ cancer with multiple metastases.

TREATMENT

To relieve wrist pain and obtain a definite pathology, the patient underwent left scaphoid lesion resection (Figure 5B). Based on the pathology results, he was treated with two courses of chemotherapy (paclitaxel and cisplatin). He subsequently discontinued chemotherapy due to intolerance.

OUTCOME AND FOLLOW-UP

The patient died of severe interstitial pneumonia 6 mo after his initial presentation.

DISCUSSION

Bone metastases are very common in various carcinomas and mainly occur in the axial skeleton, such as the spine, pelvis, and ribs. Metastatic involvement of the hand is extremely rare. Kerin^[1] demonstrated that the incidence of hand metastasis is 0.1% among all skeletal metastases. Hematogenous spread is the most common route of tumor metastasis. The hand bones have less red bone marrow, and the remoteness of the anatomical location may account for the low incidence of this disease^[3]. Trauma can contribute to acrometastasis. Repeated trauma may induce the production of chemotactic factors, such as prostaglandins, which promote cell migration and attachment. Additionally, the injury repair process creates an appropriate environment for tumor growth, invasion, and metastatic potential^[4]. The incidence of hand metastasis is higher in males than in females. The lungs, gastrointestinal tract, kidneys, and breasts are the most common primary tumor sites. Within the hand, the distal phalanges are the most frequently involved bones, followed by the metacarpals, proximal phalanges, middle phalanges, and, rarely, the carpal bones^[5]. To the best of our knowledge, metastasis to the carpal scaphoid bone, as was observed in our patient, has rarely been reported^[6], especially as the initial symptom of a primary tumor.

The signs and symptoms of hand metastases include pain, tenderness, enlarged digits, palpable masses, ulcerations, mechanical dysfunctions, and pathological fractures, which lack specificity. Consequently, hand metastases are often misdiagnosed as osteomyelitis, soft tissue infections, gout, tenosynovitis, arthritis, or other inflammatory processes^[7]. Our patient was initially misdiagnosed with a bone infection. Hence, despite its rarity, orthopedic surgeons should consider the possibility of metastases to the hand bone when patients complain of persistent and progressive pain in the hand. Radiographic investigations, such as plain radiography, CT, or MRI, play vital roles in the diagnosis of hand metastases. A positron emission tomography-CT scan can help reveal the primary tumor earlier than other techniques, as it has a sensitivity greater than 90% and a specificity of 78%^[8]. The above imaging

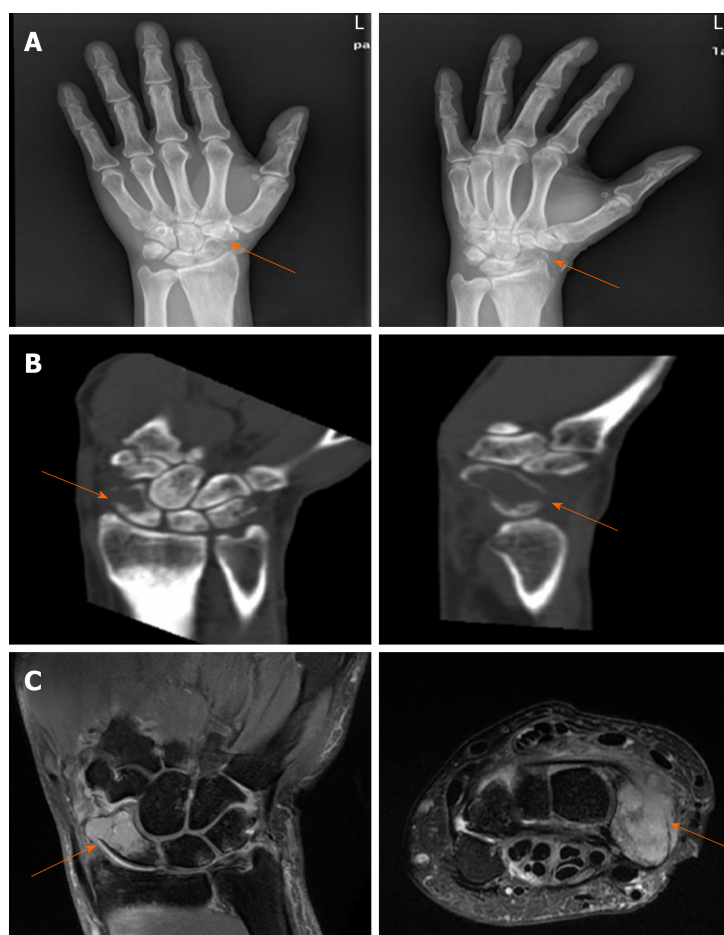


Figure 1 Imaging findings in the left hand. A: Plain X-ray showed a decrease in scaphoid bone density accompanied by a fracture; B and C: Computed tomography and magnetic resonance imaging scans demonstrated scaphoid and triangular bone destruction and soft tissue swelling, indicating a pathological fracture.

examinations can be used for auxiliary diagnosis; however, the final diagnosis should depend on pathology.

To date, there is no standard treatment for hand metastases^[9,10]. Most treatments are intended to alleviate pain and protect hand function. Radiotherapy, amputation, chemotherapy, curettage, cementation, and wide excision are the most commonly used forms of treatment. Hand metastasis often reflects the widespread dissemination of the tumor, which indicates a poor prognosis. The prognosis depends on the aggressiveness of the primary tumor. The median survival is only approximately 6 mo^[1,11].

In our case, we resected the lesion to relieve the patient's discomfort and obtain pathological specimens. The pathology indicated scaphoid bone metastasis of poorly differentiated adenocarcinoma, with the characteristics of CDX2 (+), TTF-1 (-), and ALK-Lung (-). CDX2 is a pathological marker of gastrointestinal adenocarcinoma, and TTF-1 and ALK-Lung indicate lung metastatic tumors^[12,13]. The exophytic growth pattern of the digestive tract tumor observed on the CT scan might explain the negative results on endoscopy and biopsy. Hence, according to the clinical and pathological findings, we concluded that the scaphoid metastasis originated from a cancer of the GEJ. The GEJ is defined as the junction between the esophagus and the stomach. In this location, esophageal squamous epithelial cells gradually transition to gastric gland cells, which can be easily damaged by gastric acid reflux and are associated with a higher risk of cancer transformation^[14]. The incidence of GEJ adenocarcinoma has gradually increased over the last 20 years and is associated with obesity, smoking, and gastroesophageal reflux^[15-18]. Dysphagia and retrosternal pain are often the initial manifestations. The majority of GEJ cancer patients are initially diagnosed at an advanced stage and have a poor prognosis^[19]. In addition to the common sites of esophageal cancer metastases, such as the lymph nodes, lungs, liver, and bones, unexpected metastases have been increasingly reported, especially from lower esophageal cancer. The head and neck, thorax, abdominopelvic region, extremities, and multiple skin and muscle sites are the five main unexpected

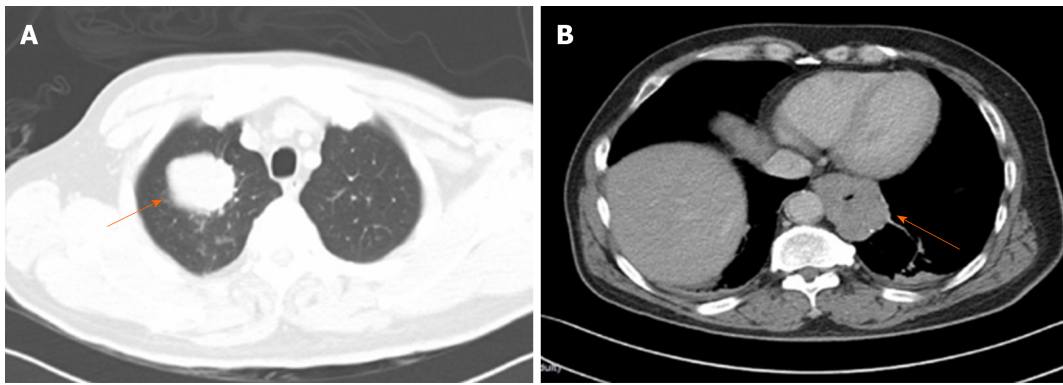


Figure 2 Preoperative chest computed tomography. A: A right upper lobe mass; B: A lower esophageal and stomach cardia mass.

metastatic sites^[20]. Multidisciplinary management, including surgery, chemoradiotherapy, and targeted therapy, is advised^[21]. Surgery is the only curative treatment, while auxiliary perioperative chemoradiotherapy and targeted therapy can improve the survival rate^[22,23]. In this case, the patient had an advanced stage of disease and was not a candidate for surgical treatment. Immunohistochemistry showed negativity for human epidermal growth factor receptor 2; hence, trastuzumab was not used.

In addition, because lung specimens were not available, it was difficult to determine whether the right lung mass was a secondary (metastasis) or primary tumor. After surgery, the patient's quality of life was greatly improved. However, the patient survived for only 6 mo after his initial presentation, potentially due to the delayed diagnosis and the aggressiveness of the primary tumor. Therefore, early diagnosis of metastasis to the hand bone could be helpful in that it allows the initiation of an appropriate course of treatment and prolongs survival time.

CONCLUSION

Hand metastases from solid tumors are clinically rare. Usually, they are late manifestations of advanced malignancies; however, sometimes they can be the initial manifestation of an occult cancer. Because of their low incidence and nonspecific symptoms, hand metastases are often misdiagnosed. Orthopedic surgeons should not ignore the possibility of hand metastases when patients complain of persistent and progressive pain in the hand. The treatment of hand metastasis is individualized and based on the clinical symptoms and characteristics of the primary tumor. The prognosis is generally poor. It is therefore important for clinicians to detect and diagnose these cancers early.

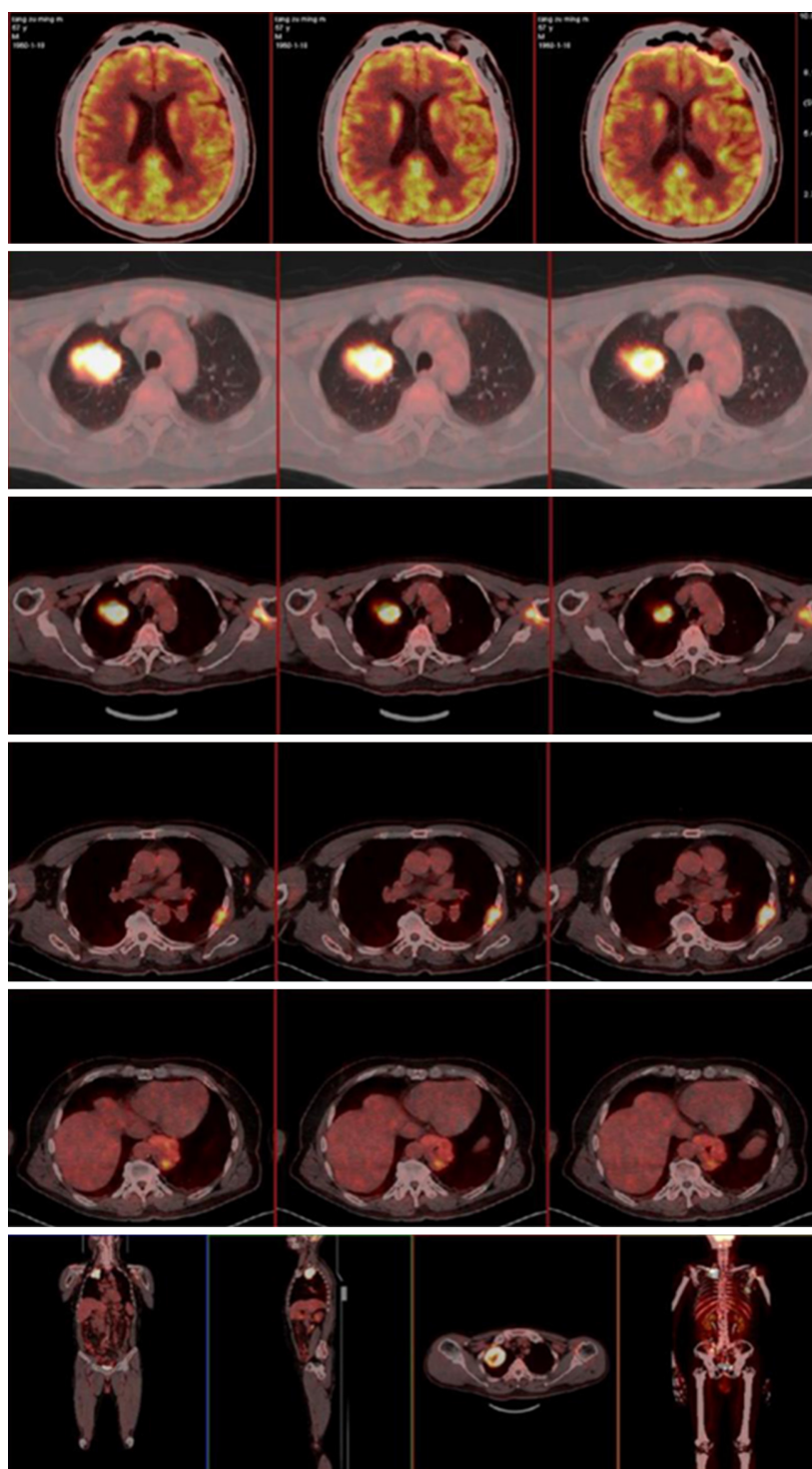


Figure 3 A preoperative 2-¹⁸F fluoro-2 deoxy-D-glucose positron emission tomography-computed tomography scan showed multiple malignant lesions throughout the whole body. This led to a strong suspicion of gastroesophageal junction and/or right lung malignancies with multiple metastases.

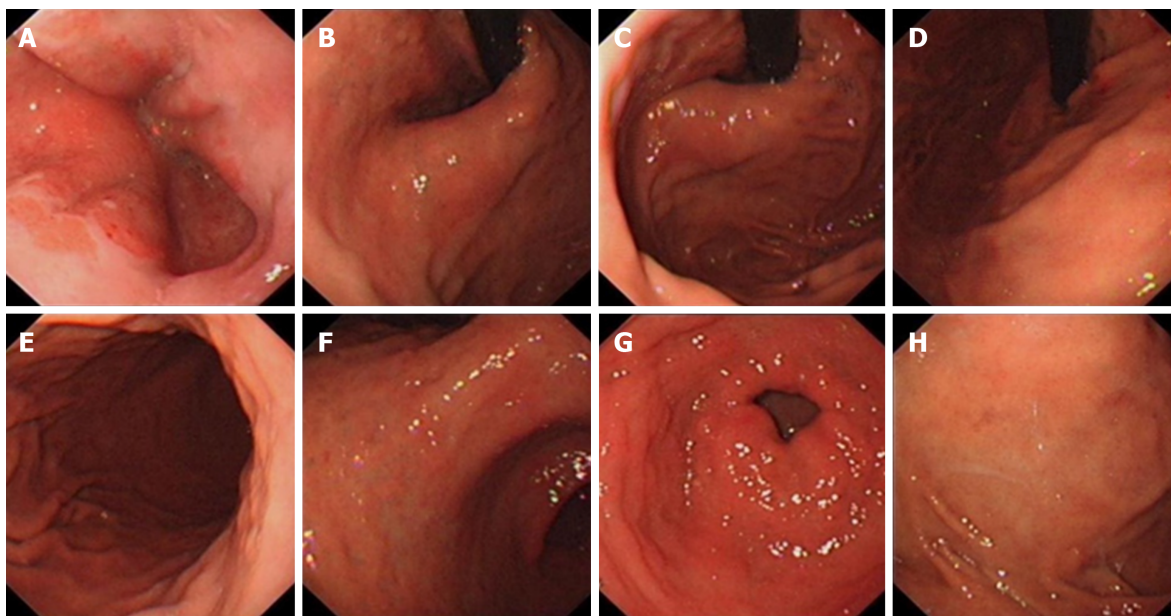


Figure 4 Preoperative gastroscopy did not find any space-occupying lesions.

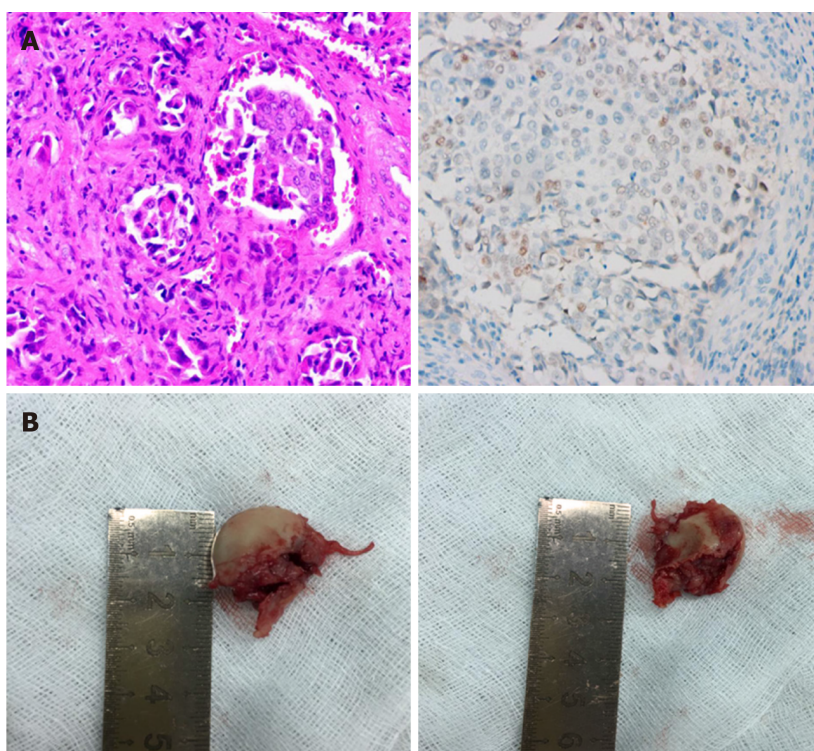


Figure 5 Intraoperative photographs and postoperative pathological results. A: Pathological biopsy confirmed scaphoid bone metastasis of poorly differentiated adenocarcinoma. Immunohistochemistry showed CDX2 positivity (200×); B: The scaphoid bone was completely removed.

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