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·临床研究·

巨大十二指肠错构瘤致慢性贫血1例报告并文献复习

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摘　要

背景与目的：十二指肠错构瘤（BGH）是十二指肠良性肿瘤中比较少见的类型，因其临床表现不典型、缺乏特异性，导致术前诊断困难，往往易被漏诊或误诊，主要依靠术后病理确诊。巨大BGH的病例临床更是罕见。本文报告1例巨大BGH致慢性贫血的诊治过程，并结合既往文献对本病特点进行复习，以期为临床工作提供经验借鉴。

方法：回顾性分析西安交通大学附属咸阳市中心医院肝胆外科收治的1例BGH患者的临床资料，结合前期文献资料进行复习总结。

结果：患者，女性，49岁；因腹痛伴恶心、呕吐就诊西安交通大学附属咸阳市中心医院肝胆外科。患者既往有慢性贫血病史，月经规律，偶有柏油样便。入院上腹部B超检查见胰头内侧十二指肠部包块；上腹部CT检查见十二指肠降部及水平部肠壁弥漫性增厚，结构不清；上腹部MRI+MRCP检查见十二指肠降部及水平部与邻近空肠分界不清，肠管明显扩张扭曲，肠壁分界不清，肠壁水肿明显，可见同心圆样改变。胃镜检查考虑十二指肠占位，并镜下活检提示符合胃黏膜异位。上消化道造影提示十二指肠降段与水平段移行区近圆形充盈缺损，考虑良性占位可能性大。最终考虑十二指肠占位性病变并引起梗阻，剖腹探查术后行胰十二指肠切除术，切除标本送病理学检查，最终诊断为BGH，免疫组化MUC5AC（+），术后恢复顺利出院。术后监测血常规，红细胞及血红蛋白逐渐恢复正常，随访12个月，患者一般状况良好，未诉不适，复查各项指标正常，继续随访。

结论：巨大BGH致慢性贫血临床罕见，术前影像学检查往往很难提供有价值的信息，胃镜下活检有助于诊断，但阳性率低，最后确诊需依靠手术切除标本的组织病理学检查，病变较大时外科手术切除是治疗该病的有效手段。

关键词　十二指肠肿瘤；错构瘤；胃肠出血；贫血

中图分类号：R735.3

Giant Brunner's gland hamartoma with associated chronic anemia: a case report and literature review

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Abstract

Background and Aims: Brunner's gland hamartoma (BGH) is a rare type of duodenal benign tumor. It is difficult to diagnose before operation and often easily to be missed or misdiagnosed because of

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atypical clinical manifestations and lack of specificity. The diagnosis mainly depends on postoperative pathology. The cases of giant BGH are rare. This paper reports the diagnosis and treatment of a case of giant BGH with associated chronic anemia, and reviews the characteristics of this disease in combination with the previous literature, so as to provide experience for clinical management of this condition.

Methods: The clinical data of a patient with BGH admitted to the Department of Hepatobiliary Surgery of Xianyang Central Hospital Affiliated to Xi'an Jiaotong University were analyzed retrospectively in combination with a review of the previous literature.

Results: The patient was a 49-year-old female, and was admitted to the Department of Hepatobiliary Surgery of Xianyang Central Hospital Affiliated to Xi'an Jiaotong University because of abdominal pain accompanied by nausea and vomiting. The patient had a history of chronic anemia, regular menstruation, and occasional black stool. On admission, the B-ultrasound examination of the upper abdomen showed the mass of the duodenum overlapped by the head of the pancreas; CT examination of the upper abdomen showed diffuse thickening of the intestinal wall in the descending and horizontal parts of the duodenum, with unclear structure; MRI+MRCP examination of the upper abdomen showed that the descending and horizontal parts of the duodenum and the adjacent jejunum were indistinct, the intestinal tube was obviously distended and twisted, the intestinal wall was indistinct, with obvious intestinal wall edema and presence of concentric circles change. Duodenal space occupying was considered in gastroscopy, and biopsy under endoscopy suggested that it was consistent with the ectopic gastric mucosa. The upper gastrointestinal angiography showed a nearly round filling defect at the junction of the descending and horizontal segments of the duodenum, considered to be a benign occupying lesion. Finally, the obstruction caused by the duodenal space-occupying lesion was considered, and pancreaticoduodenectomy was performed after exploratory laparotomy. The final diagnosis was BGH and immunohistochemical staining for MUC5AC positive after the resected specimen was sent to pathological examination. The patient recovered uneventfully after surgery and was discharged from the hospital. After the operation, the blood routine was monitored, and the red blood cells and hemoglobin gradually returned to normal. The patients were followed up for 12 months. The patients were generally in good condition without complaints of discomfort. All indexes were normal in the reexamination, and the follow-up was continued.

Conclusion: Giant BGH with associated chronic anemia is rare in clinical practice. Preoperative imaging examination is often difficult to provide valuable information. Biopsy under gastroscope is helpful for diagnosis, but the positive rate is low. The final diagnosis depends on histopathological examination of the surgical specimens. Surgical resection is an effective means to treat the disease when the lesion is large.

Key words Duodenal Neoplasms; Hamartoma; Gastrointestinal Hemorrhage; Anemia

CLC number: R735.3

十二指肠错构瘤，也称为十二指肠腺瘤或Brunner腺错构瘤（Brunner's gland hamartoma，BGH），是临床上一种罕见的十二指肠良性肿瘤，在十二指肠良性肿瘤中发病率约为5.0%~10.0%，常规内镜镜检率为0.01%~0.07%[1-3]。通常无临床症状，偶见十二指肠梗阻或上消化道出血，罕见胆瘘、胆汁淤积性黄疸和肠套叠[4]。因临床表现缺乏特异性，诊断相对困难，术前影像学易被误诊为胆道系统疾病，其确诊主要依靠术后病理。目前其病因和发病机制尚不明确，认为可能与胃酸分泌过多、幽门螺杆菌感染、慢性胰腺炎、炎症刺激以及黏膜损伤等因素相关[5-6]。现回顾性分析2021年

1 月 29 日西安交通大学附属咸阳市中心医院肝胆外科收治的 1 例 BGH 患者临床资料，并结合文献进行综合分析，以期为临床诊疗提供参考。

1　资料与方法

1.1　病例资料

患者　女，49 岁。腹痛伴恶心、呕吐 1 d 就诊，患者自诉 1 d 前进食后出现上腹部疼痛，呈持续性，并进行性加重，伴恶心、呕吐，呕吐物为胃内容物，伴发热，体温最高 38.0 ℃。体格检查：贫血貌，右上腹压痛阳性，余未见明显异常。实验室检查：白细胞 14.77×10^9/L，中性粒细胞 13.37×10^9/L，中性粒细胞百分比 90.50%，红细胞 3.37×10^{12}/L，血红蛋白 92 g/L，红细胞压积 29.80%，平均血红蛋白浓度 309 g/L；白蛋白 30.8 g/L，总胆红素 18.3 μmol/L，直接胆红素 14.7 μmol/L，间接胆红素 3.6 μmol/L，丙氨酸氨基转移酶 313 U/L，天门冬氨酸氨基转移酶 588 U/L，碱性磷酸酶 549 U/L，γ–谷氨酰转肽酶 421 U/L；粪隐血试验阳性；肿瘤指标正常。上腹部 B 超检查提示：胰头内侧十二指肠部包块；上腹部 CT 检查提示：⑴ 肝内胆管及胆总管扩张，胆囊积液；⑵ 十二指肠降部及水平部肠壁弥漫性增厚，结构不清（图 1A）。上腹部 MRI+MRCP 检查提示：低位胆道梗阻并胆囊积液，扩张胆总管下段受牵拉向左移位，十二指肠降部及水平部与邻近空肠分界不清，肠管明显扩张扭曲，肠壁分界不清，肠壁水肿明显，可见同心圆样改变，多考虑小肠扭转并局部套叠（图 1B–C）。胃镜检查提示：十二指肠占位、慢性浅表性胃炎，并镜下取 1 块组织活检。活检报告：符合胃黏膜异位（图 2）。上消化道造影提示：十二指肠降段与水平段移行区近圆形充缺，多考虑良性占位可能。既往有慢性贫血病史 2 年余，偶解柏油样便，月经规律。初步诊断：⑴ 十二指肠占位并梗阻；⑵ 慢性贫血。

1.2　治疗过程及随访情况

术前考虑十二指肠占位并引起梗阻，经患者及家属同意，于 2021 年 2 月 3 日在全麻下行剖腹探查术，术中探查腹腔未见明显异常，常规游离十二指肠，切开十二指肠降部，见肿物位于幽门至十二指肠乳头间，大小约 7 cm×6 cm（图 3A），质韧、与周围组织纠集且分界不清，不除外十二指肠恶性肿瘤可能，术中向患者家属充分告知并取得家属同意后，遂决定行胰十二指肠切除术，手术顺利，术后予以抗感染、抑酸、抑酶、营养支持、对症等治疗，术后第 4 天病检报告：BGH，免疫组化 MUC5AC（+）（图 3B）。术后复查上腹部 CT 结果正常（图 3C），术后恢复顺利，于术后第 12 天出院。术后监测血常规，红细胞及血红蛋白逐渐恢复正常。随访 12 个月，患者未诉不适，复查各项指标正常。

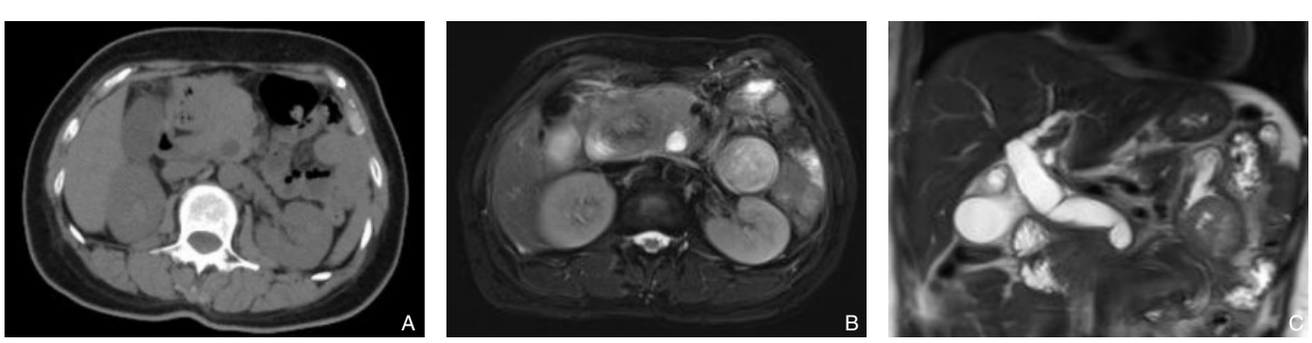

图 1　入院影像学资料　A：CT 示十二指肠降部及水平部肠壁弥漫性增厚，结构不清；B：轴位 MRI 示十二指肠降部及水平部与邻近空场分界不清，肠管明显扩张扭曲，肠壁分界不清，肠壁水肿明显，可见同心圆样改变；C：冠状位 MRI 示低位胆道梗阻并胆囊积液，扩张胆总管下段受牵拉向左移位

Figure 1　Imaging data on admission　A: CT showing diffuse thickening of the intestinal wall in the descending and horizontal parts of the duodenum, with unclear structure; B: Axial MRI imaging showing that indistinct sight of the descending and horizontal parts of the duodenum and the adjacent jejunum, with obviously distended and twisted intestinal tube, indistinct intestinal wall, obvious intestinal wall edema and presence of concentric circles change; C: Coronal CT imaging showing lower biliary obstruction and gallbladder collections, with a left dislocation of the lower common bile duct due to traction

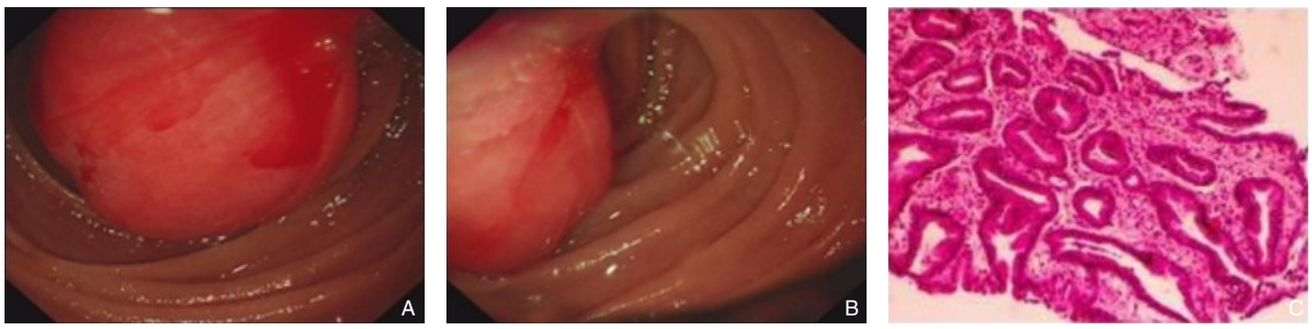

图2 胃镜资料 A：十二指肠降部；B：十二指肠水平部；C：胃镜活检胃型黏膜（HE ×100）

Figure 2 Gastroscopic data A: View of the descending part of the duodenum; B: View of the horizontal part of the duodenum; C: Biopsy showing gastric mucosa (HE ×100)

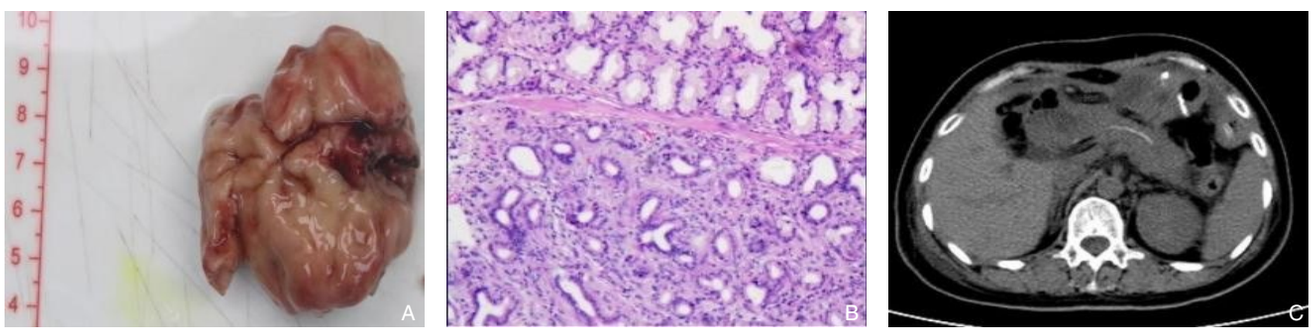

图3 术后资料 A：大体标本；B：组织病理示增生的导管及腺体形成分叶状结构（HE×100）；C：术后复查CT

Figure 3 Postoperative data A: Gross specimen; B: Histopathological examination showing the lobulated structures of the hyperplastic ducts and glands (HE×100); C: Postoperative CT review

2 讨论并文献复习

错构瘤形成的主要原因是机体各种组织成分比例、分布、分化异于正常组织。BGH主要由Brunner腺体、平滑肌、纤维结缔组织及血管构成。其病理表现为十二指肠腺增生伴小叶结构消失，间质内可见淋巴组织、脂肪组织及平滑肌束穿插，有时可见被覆柱状或纤毛柱状上皮的导管，可呈囊性或有黏液潴留[7]。依据病变形态将其分为3型：1型，弥漫性结节增生；2型，局限性结节增生；3型，腺瘤样增生，其中3型最常见，包括有蒂或无蒂两种[8-10]，本例患者属于有蒂的腺瘤样增生。BGH是良性肿瘤，进展为十二指肠腺癌的风险非常低，但具有恶变潜能，随着Brunner腺良性增生性病变的发展，可能会发生黏膜溃疡，从而导致具有乳头状结构的胃小凹化生修复，进而发生恶变[11-15]。因此，当病变增大并且发生形态学改变时，应当警惕恶变可能。

大多数BGH是孤立的带蒂息肉，少数不带蒂，直径大多介于1~2 cm，很少超过5 cm。大多位于十二指肠近端，距幽门越远，发病率越低，十二指肠球部占57.0%；十二指肠降部占27.0%；十二指肠水平部占7.0%[16]。部分BGH患者由于瘤体小，可无临床症状，部分瘤体较大、有临床症状者缺乏特异性（包括消化不良、腹痛、腹胀、恶心、呕吐、消化道出血、梗阻、缺铁性贫血等），故临床诊断困难。消化道出血和梗阻常常是患者就诊的主要原因[4,17-20]。本例患者因消化道梗阻就诊，既往有慢性贫血病史，这可能是由瘤体血管糜烂导致的慢性失血引起。席芸等[21]报道了18例十二指肠腺增生性病变，包括9例BGH，其中有3例体检发现，1例因黑便、5例因消化道梗阻就诊。Peloso等[22]发现，BGH引起出血与瘤体的大小和部位有关，瘤体越大，位置越远，更容易引起消化道出血。有时瘤体过大还会引起胃十二指肠套叠[23-25]。

目前，BGH的诊断常依靠内镜及影像学检查。内镜表现为黏膜下隆起性病变，表面光滑，表面可伴有散在出血点。值得注意的是，内镜活检大多为阴性，这是因为病灶通常被覆正常十二指肠

黏膜，活检深度难以到达黏膜下层的病灶。刘健等[26]报道的 2 例 BGH 均行胃镜下活检，结果均为阴性，最终通过病灶切除后术后病理报告确诊。超声内镜（endoscopic ultrasonograph，EUS）提示病灶为黏膜下受累的混合回声，内部常呈囊性改变，具有特征性的诊断价值[27-28]。EUS 引导下细针穿刺可提高诊断准确率[29]。上消化道造影表现为光滑的无蒂或带蒂状的充盈缺损，边界清楚，可伴有小糜烂或溃疡，无明显浸润及僵硬[30]。CT 及 MRI 显示肿物有蒂且内部呈囊性改变，则提示可能为 BGH，并且可以判断病灶与相邻结构（如胰腺、胆总管和血管）的关系以及有无肠外浸润表现，有助于和其他十二指肠肿块相鉴别，如平滑肌瘤、胃肠道间质瘤、幽门黏膜脱垂或 Peutz-Jeghers 息肉等[31-32]。本文报道的病例 BGH 体积较大（约 7 cm × 6 cm），临床较为罕见，起初误以为胆道系统疾病，这是由于瘤体较大，累及十二指肠壶腹部，导致低位胆道梗阻，引起胆管扩张，容易误诊为胆道相关疾病，需要警惕，注意鉴别。

对于 BGH 较小、无症状的患者，可以保守治疗，如果病灶大、出现症状时，建议切除，可以考虑内镜切除及外科手术。秦榕等[33]报道了 3 例 BGH，均行内镜下切除，术后随访，效果良好，无并发症。内镜治疗的优点是创伤小、安全、费用低，住院时间短。但对于复杂、巨大或无蒂的 BGH，内镜下治疗往往比较困难，需要外科手术切除。本例患者由于瘤体体积大，术中探查不除外恶性可能，最终决定行胰十二指肠切除术，目前随访恢复良好。

本文报道了 1 例巨大 BGH 致慢性贫血病例并回顾性分析相关文献，BGH 患者缺乏特异性临床表现，常见的症状有腹痛、腹胀、恶心、呕吐、黑便等，影像学检查常无阳性发现，并且易误诊为胆道系统疾病，内镜、EUS 以及 EUS 下穿刺活检有助于诊断，但难度较大。当临床上出现消化道梗阻或慢性出血时，胃镜检查发现十二指肠肿物，肿物边界清楚、表面光滑，黏膜呈暗红色、水肿时，应考虑 BGH 可能。BGH 恶性者罕见，但当病变增大并且有形态学改变时，应警惕恶变可能。病灶较大者可考虑外科手术切除。本例患者在手术探查过程中发现瘤体大、且无法排除十二指肠恶性肿瘤可能，故决定行胰十二指肠切除术，手术创伤虽大，但取得较好效果，长期随访，患者恢复良好，未见复发，长期的慢性贫血得到改善，对临床有一定的参考价值。

利益冲突：所有作者均声明不存在利益冲突。

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