Case Report

Endoscopic Resection of a Gastric Glomus Tumour Using a Hybrid Approach of Endoscopic Submucosal Dissection and Endoscopic Full Thickness Resection: A Case Report

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Abstract

Gastric glomus tumour is a rare type of mesenchymal neoplasm arising from arteriovenous shunts in the muscular is propria responsible for thermoregulation, also known as glomus bodies. Due to its indistinguishable characteristics, this usually benign tumour is often resected with surgery to simultaneously diagnose and treat the lesion. We present a case of a 57-year old woman with a submucosal lesion in the angulus of the stomach, which turned out to be a glomus tumour after endoscopic resection using a hybrid approach of endoscopic submucosal dissection and endoscopic full-thickness resection.

Keywords: Glomus Tumour; Endoscopic Resection; Endoscopic Submucosal Dissection; Endoscopic Full Thickness Resection

Abbreviations: EUS: Endoscopic ultrasound; ESD: Endoscopic submucosal dissection; eFTR: Endoscopic full thickness resection.

Introduction

Glomus tumour is a rare type of mesenchymal neoplasm derived from modified smooth muscle cells of normal glomus bodies, which play a role in thermoregulation [1]. Although glomus tumours usually involve areas rich in glomus bodies such as the subungual regions or the dermis of extremities [1], these neoplasms may present anywhere in the body, including the gastrointestinal tract [2]. Here we report a case of a gastric glomus tumour, which was treated curatively with endoscopic resection using a hybrid approach of endoscopic submucosal dissection (ESD) and endoscopic full thickness resection (eFTR).
Case Report

A 57-year old woman was referred for further evaluation of a submucosal mass in the angulus of the stomach, after biopsies of the lesion and a computed tomography scan at the referring hospital were both inconclusive. Endoscopic ultrasonography (EUS) at our centre demonstrated a hypoechoic submucosal lesion at the angulus of approximately 1cm, originating from the fourth echo layer (Figure 1). Although EUS-guided fine needle aspiration showed normal cylindrical epithelium without suspicion of malignancy, the sample was considered superficial and therefore unrepresentative. Due to reservations regarding the lesion’s nature, the patient was kept under strict endoscopic follow-up consisting of 6-monthly upper endoscopy supplemented with EUS.

After two years of endoscopic surveillance, the lesion was biopsied using EUS-guided fine needle biopsy, where only a small amount of tissue was retrieved. In this small fragment, a lesion composed of monomorphous cells with round nuclei and a fine chromatin pattern was seen. As immunohistochemical stains associated with neuro-endocrine tumours (i.e. synaptophysin and CD56) were weakly positive, the suggestion of a neuro-endocrine tumour was made. A subsequent PET-CT scan specifically aimed at the detection of neuro-endocrine tumours (i.e. DOTANOC) could not visualize the suspect lesion, but showed no signs of metastases. Therefore, an attempt was made to endoscopically resect the lesion by means of ESD.

Using a DualKnife (Olympus, Tokyo), the endoscopist initially opted to create a submucosal tunnel towards the lesion, in order to either proceed with Submucosal Tunnelling Endoscopic Resection or ESD, depending on the true layer of origin. This approach was chosen since gastric neuro-endocrine tumours are usually located in the submucosa, which was in contrast to the EUS findings where the lesions appeared to arise from the proper muscle layer. However, the lesion’s location on the angulus complicated both antegrade and inverted approaches. After several unsuccessful attempts, the decision was made to perform a circular mucosal incision instead. Further submucosal dissection

Figure 1: Endoscopic and histopathological images of the gastric glomus tumour. (A) The gastric tumour at the angulus was initially diagnosed upon upper endoscopy. (B) Additional endoscopic ultrasound demonstrated a hypoechoic submucosal mass of approximately 1cm. (C) After endoscopic submucosal dissection was technically unsuccessful, the lesion was removed en-bloc with endoscopic full thickness resection. (D) Histopathological evaluation of the resection specimen showed normal mucosa and underlying smooth muscle with the lesion located in the stomach wall (black rectangle; magnification x20). (E) The lesion was composed of multiple nodules of tumour cells with a solid growth pattern, located around dilated vessels. The tumour cells were uniform, round, with distinct cell borders. The nuclei had a fine granular chromatin pattern (magnification x200). (F) Actin stain was positive in the lesion (magnification x100).
better exposed the lesion, showing that the lesion arose from the muscularis propria, hence protruding into the gastric lumen. The procedure was converted to an eFTR. The lesion was grabbed with a forceps to provide traction, and was subsequently removed en-bloc after firing the clip followed by snaring (Figure 1). One day after the procedure, the patient was discharged without any adverse events.

Histopathological evaluation of the endoscopic resection specimen showed a lesion composed of multiple nodules of tumour cells with a solid growth pattern, located around dilated vessels at the level of the muscularis propria. The tumour cells were monomorphic and round, with distinct cell borders. The nuclei displayed a fine granular chromatin pattern. Although the lateral margins were tumour negative, the tumour cells reached into the vertical resection margin. Further characterization of the lesion showed that immunohistochemical stains for smooth muscle actin and calponin were positive. Stains for keratin, c-KIT (CD117), DOG1, S100 and chromogranin were negative, while stains for CD56 and synaptophysin were weakly positive. The proliferation index (Ki-67) was low. These findings led to the diagnosis of a glomus tumour. Considering the typically benign nature of this tumour type, the endoscopic resection was considered curative and a shared decision was made to withhold the patient from further follow-up.

Discussion

We present a case in which a gastric glomus tumour was diagnosed and treated by means of endoscopic resection using a hybrid approach of ESD and eFTR.

Gastric glomus tumours represent approximately 1% of all gastric mesenchymal tumours [3], and are predominantly benign with malignancy rates reported up to 6% [2,4,5]. Due to the lack of specific clinical and endoscopic characteristics, gastric glomus tumours are difficult to distinguish from other types of gastric submucosal neoplasms with either upper endoscopy, EUS or radiologic imaging modalities [6-8]. Although histopathological sampling may provide a more definite diagnosis, tissue biopsies and fine needle aspiration may be hampered by the submucosal nature of the lesion as well as sampling error [9]. As a consequence of this diagnostic challenge, complete resection of a gastric glomus tumour is considered necessary to simultaneously diagnose and treat the lesion. Thus far, surgical resection has been considered the first-choice treatment, with gastric wedge resection as the most frequently used method followed by distal gastrectomy and subtotal gastrectomy [4,10,11]. However, surgery is an invasive procedure with significant burden for the patient.

Compared to surgery, endoscopic resection holds several potential advantages for the evaluation and treatment of gastric glomus tumours including the preservation of the gastric anatomy and function, less morbidity, shorter procedure times, and shorter hospital stay. ESD in particular has proven a safe and effective alternative to surgery for the resection of early gastric neoplasia [12]. Although a few studies have demonstrated that ESD can be used to accurately characterize and resect gastric glomus tumours, [7,10,11,13] en-bloc resection may be impeded by tumour invasion into the muscularis propria. As demonstrated by our case, eFTR may then be a valid alternative for en-bloc resection of lesions with limited size.

To conclude, complete resection is considered a necessity for gastric glomus tumours, as diagnostic modalities are often insufficient to provide a conclusive diagnosis. Surgical excision is currently the most widely used treatment approach, but this case report demonstrates that endoscopic resection could serve as a less invasive alternative in selected cases.

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References


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