



Long-Term Clinical Outcome of Ischemic Polypectomy in a Patient with Peutz-Jeghers Syndrome: A Case Report

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Abstract

Peutz-Jeghers Syndrome (PJS) is characterized by multiple gastrointestinal hamartomatous polyps, often leading to complications such as small bowel obstruction. We present a case of a 75-year-old male diagnosed with PJS, exhibiting numerous small bowel polyps. We implemented a novel treatment approach known as the Ischemic Polypectomy Technique. The patient underwent Double-Balloon Enteroscopy (DBE) using an underwater technique, involving clipping at the polyp base to induce ischemia and subsequent necrosis. Substantial reductions in both polyp count and size were observed during follow-up small bowel video capsule endoscopies at 5- and 17-months post-procedure. Notably, a decrease from 150 to 35 polyps and a reduction in size from 20 mm to 5 mm, coupled with observed polyp decapitation, demonstrated the efficacy of this method. Compared to conventional DBE polypectomy, the ischemic approach proved more accessible, quicker, and safer. While histopathological confirmation was unavailable due to non-retrieval of polyps, the observed morphological changes strongly indicated therapeutic efficacy. However, uncertainties persist regarding the extensive use of metal clips and associated costs. This case highlights the feasibility and effectiveness of endoscopic ischemic polypectomy for PJS-associated small bowel polyps. Despite potential limitations, this method shows promise in preventing complications associated with PJS, necessitating further case accumulation.

Keywords: Peutz-Jeghers Syndrome; Hamartomatous polyps; Double Balloon Enteroscopy; Ischemic Polypectomy

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Introduction

Peutz-Jeghers Syndrome (PJS) is an inherited cancer predisposition condition primarily attributed to a heterozygous germline pathogenic variant in *STK11*. It is characterized by the development of hamartomatous polyps in the gastrointestinal tract [1-4]. PJS-type polyps are predominantly located in the gastrointestinal tract, particularly in the small bowel (60-90%, especially the jejunum), and frequently in the large bowel (50-64%) and stomach (15-30%) [5].

These polyps can lead to chronic bleeding, recurrent obstruction of gastrointestinal tract, and intussusception. Accordingly, clinical guidelines recommend Double-Balloon Enteroscopy (DBE) with polypectomy as the standard treatment for removing PJS-type polyps, particularly those larger than 15 mm to 20 mm or patients with obstructive symptoms, irrespective of size [6]. However, this procedure carries a complication rate of 6.8% and can be technically challenging and time-consuming [7].

In this report, we introduce a novel technique called "Ischemic Polypectomy" for numerous small bowel polyps in a PJS patient [8]. Our findings indicate long-term clinical outcomes that appears to be safe, effective and straightforward.

Case Presentation

A 75-year-old male was referred to the National Cancer Center Hospital because of multiple gastric polyps detected in a medical health check.

An esophagogastroduodenoscopy revealed multiple gastric polyps measuring less than 5 mm, histologically characterized by foveolar hyperplasia. A colonoscopy identified approximately 50 polyps in colon and rectum, each measuring less than 10 mm. We removed 17 polyps and among them, 14 were pathologically hamartomatous polyps (Figure 1A). Given the endoscopic and histological findings, we clinically diagnosed this patient with PJS.

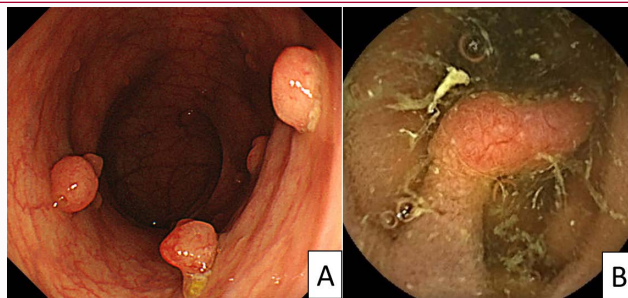


Figure 1: Representative images of gastrointestinal polyps. A) Colonic polyps. B) Small intestinal polyps.

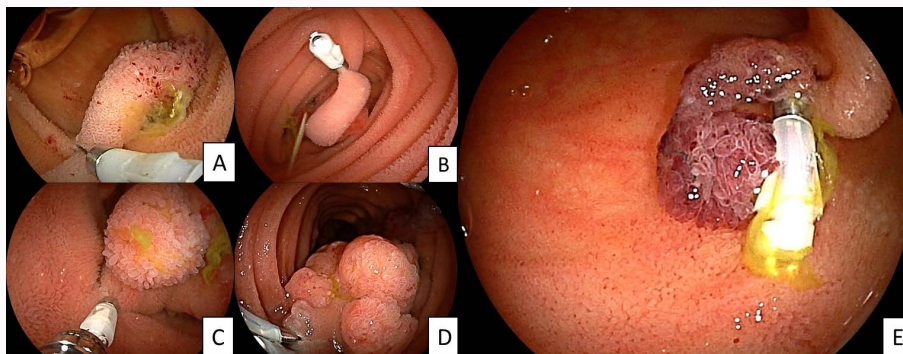


Figure 2: Representative images of ischemic polypectomy. A) Ischemic Polypectomy. B) Ischemic Polypectomy in a polyp located at the 12 o'clock position. C) Ischemic Polypectomy of a polyp with a stalk thicker than 1 cm. D) Placement of an additional clip in the polyp showed in image C due to lack of response. E) Polyp with color changes on its surface after treatment.

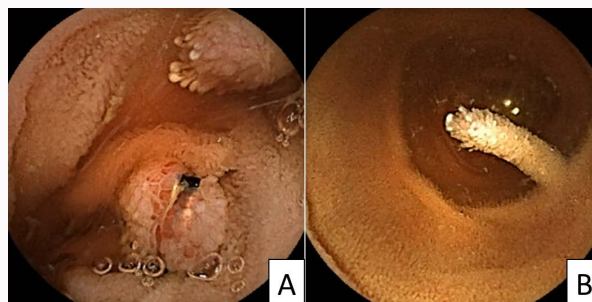


Figure 3: Findings in Small Bowel Video Capsule Endoscopy performed after 17 months. A) 2 decapitated polyps. B) Decapitated polyp, with only the stalk being visible.

A next-generation sequencing multi-gene panel test revealed no pathogenic variants for 147 hereditary cancer genes including *STK11* and a MLPA (Multiple Ligation-dependent Probe Amplification) test showed 2 copies in *STK11*.

A Small Bowel Video Capsule Endoscopy (SBCE) detected about 150 polyps, morphologically sessile or pedunculated (Figure 1B), with a maximum diameter of up to 20 mm. Given the significant number of polyps, a DBE with ischemic polypectomy was planned.

Initially, a retrograde DBE was conducted and small bowel polyps, regardless of their size, were treated using an underwater technique by clipping at their base. The underwater technique allows the polyps to float and their stalks to straighten, facilitating the procedure (Figure 2A-2C and Video). A total of 8 liters of saline solution (NaCl 0.9%), administered via a flushing pump, were used to prevent severe hyponatremia and subsequent neurological complications. We used EZ-CLIP (Olympus, Tokyo, Japan) hemostatic clips, which are easy to reload and orient and come in various sizes, selected based on

the diameter of the stalk. Correct placement of the clip resulted in a noticeable color change from pale to dark purple on the polyp's surface after a few minutes (Figure 2D). Additional clips could be used if there was no response (Figure 2E). After treating all the polyps consecutively, the last clip placed was taken as a reference for the maximum insertion point. The following day, an oral DBE was performed, and the same procedure was repeated until the marking clip. A total of 80 polyps were treated, requiring approximately 100 clips.

To assess the effects of this treatment, two SBCE were performed at 5 and 17 months later, revealing a progressive and significant decrease in the number of polyps. There were 50 polyps in the SBCE at 5 months and 35 in the SBCE at 17 months. Additionally, the maximum diameter of the polyps found in the latter was 5 mm to 6 mm and most of them were decapitated, with only the stalk being visible (Figure 3A, 3B, Video).

Discussion

DBE with conventional polypectomy is generally accepted as the standard treatment for removing Peutz-Jeghers Polyps in the small bowel [6]. This method is known to prevent intussusception and the need for emergency surgery. However, endoscopic treatment with snaring in small intestine is technically more challenging, time-consuming and carries higher risks, with a global complication rate of 6.8%, specifically 2.7% for delayed bleeding, 2.7% for acute pancreatitis and 1.4% for perforation [7].

Polyp Stalk Ligation Technique was first described in 2011 [9] and the use of clips for ischemic treatment has been developed by the Yamamoto group at Jichi Medical University since 2008 [10]. This method offers considerable benefits. It is easier to perform because polyps are more accessible, resulting in shorter procedural times. There is no tissue damage, only ischemia and necrosis, which reduces the risk of bleeding, perforation and post polypectomy coagulation syndrome. In fact, in the Yamamoto series, only one case of acute pancreatitis occurred and was considered a complication; however, it was not directly related to the polypectomy [10].

Our case report demonstrates the feasibility and effectiveness of this treatment based on the long-term clinical outcomes. There were no complications during or after the procedure. Compared to conventional polypectomy, this method is safer and easier to perform, especially using the underwater technique. This approach provides superior exposure of the polyp stalks, simplifying the clipping process. Furthermore, this method allows for the treatment of a greater number of polyps in a single session, reducing the necessity for additional examinations and minimizing inconvenience for the patient. Although we lack histopathological confirmation of the polypectomy due to the non-retrieval of the polyps, SBCE identified notable morphological changes such as polyp decapitation and a significant reduction in their number and size even 17 months after the procedure. Therefore, this treatment may contribute to preventing complications associated with PJS.

However, notable limitations warrant consideration. Firstly, there are uncertainties regarding the safety of placing a large number of metallic clips in the small bowel. Additionally, the cost of the materials used, including clips and saline, is significantly higher than that of conventional polypectomy. Nevertheless, after two years, only approximately 10 clips remain, as confirmed by an abdominal X-ray.

Conclusion

The present case suggests that "Ischemic Polypectomy" could be a valuable and promising technique for managing PJS-type small bowel polyps. Further studies and long-term assessments are warranted to validate its safety, cost-effectiveness, and broader applicability in clinical practice.

Video: https://youtu.be/9Jk-Fhiq_G4

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