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A Case with Serrated Polyposis Syndrome Controlled by Multiple Applications of Endoscopic Mucosal Resection and Endoscopic Submucosal Dissection

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Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 66
Final Diagnosis: Serrated polyposis syndrome
Symptoms: Positive fecal occult blood test
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology

Objective: Unusual setting of medical care

Background: Serrated polyposis syndrome (SPS) is characterized by numerous hyperplastic polyps and sessile serrated adenoma/polyp (SSA/P) in the large intestine. SSA/P is known to transform into malignant lesions through the serrated pathway instead of the adenoma-carcinoma sequence. Early diagnosis with lower gastrointestinal endoscopy and early treatment are now considered to be essential.

Case Report: We had an experience with a case of SPS to which endoscopic treatment was applied in multiple sessions. Endoscopic treatment was performed for 16 lesions in total, and the pathological findings were SSA/P for 15 and adenoma for the other lesion. We intend to continue performing endoscopic surveillance for any newly developing lesions.

Conclusions: SPS has a potential for malignant transformation, and issues, such as long-term prognosis and optimal therapeutic strategies, await resolution. However, multiple endoscopic treatments are useful for cases with lesions that are controllable employing this modality.

MeSH Keywords: Endoscopy, Gastrointestinal • Intestinal Polyposis • Treatment Outcome

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Background

Serrated polyposis syndrome (SPS) is characterized by numerous hyperplastic polyps and sessile serrated adenoma/polyp (SSA/P) in the large intestine [1]. Although hyperplastic polyps have not been regarded as a target for treatment in the past, it has been revealed that lesions similar to hyperplastic polyps include tumors containing dysplastic cells. Such lesions have been referred to as SSA/P [2]. The diagnostic criteria for SPS were first proposed by Burt et al. for the World Health Organization (WHO) in 2000. In recent years, these diagnostic criteria have been redefined, and the condition is now generally referred to as serrated polyposis. According to the WHO clinical diagnostic criteria, SPS is diagnosed when at least one of the following criteria is satisfied: (1) at least five serrated tumors are detected in the area proximal to the sigmoid colon, and two of them measure more than 10 mm in diameter; (2) many serrated tumors are located in the area proximal to the sigmoid colon, and a patient has a first-degree family history of SPS; and (3) there are 20 or more serrated tumors distributed throughout the large intestine [3]. SSA/P is known to transform into malignant lesions through the serrated pathway instead of the adenoma-carcinoma sequence [4]. Thus, early diagnosis with lower gastrointestinal endoscopy and early treatment are now considered to be essential.

We herein report our experience with a case of SPS to which endoscopic treatment was applied in multiple sessions. We successfully cleared all polyps. SPS is a relatively new disease entity and we have to be aware of this syndrome, because SPS has malignant potential.

Case Report

At a routine health checkup, a 66-year-old man was found to have a positive fecal occult blood test, and subsequent lower gastrointestinal endoscopy detected polyps exceeding 10 mm in diameter at more than 10 sites in the right hemi-colon. The largest lesion had a diameter of 30 mm. Although his older sister had a history of colorectal cancer, he had no relatives with SPS. We planned multiple applications of endoscopic treatment and then obtained informed consent from him. We first performed endoscopic mucosal resection (EMR) for five lesions in the ascending colon. The pathological diagnosis was sessile serrated adenoma/polyp (SSA/P) for four lesions and adenoma for the other. At this time, EMR was also attempted for the 30-mm type IIa+IIc lesion (Paris classification [5]). However, resection had to be abandoned due to swelling of the center of the lesion being insufficient even after local injection of physiologic saline. Six months later, endoscopic submucosal dissection (ESD) was performed for the 30-mm type IIa+IIc lesion, for which EMR had earlier been abandoned (Figure 1A, 1B). The histopathological examination of the resected specimen revealed proliferation of serrated epithelium in the mucosal layer, an enlarged and irregularly branched crypt, and deformation of the crypt base into the shape of an inverted T. The lesion was diagnosed as SSA/P (Figure 1C, 1D).

EMR was performed for two lesions in the ascending colon 14 months later, followed by ESD performed for two other lesions in the ascending and transverse colons 16 months later, and ESD for three lesions in the transverse colon 22 months later and for three more lesions in the transverse colon 28 months later. The pathological diagnosis was SSA/P for all of these lesions. Finally, endoscopic treatment had been applied to 16 lesions in total, and the pathological diagnosis was SSA/P for 15 lesions and adenoma for the other lesion. This case fulfilled the following WHO classification of SPS: at least five serrated tumors are detected in the area proximal to the sigmoid colon, and two of them measure more than 10 mm in diameter. Clearing of all polyps was completed; we recommended colonoscopy surveillance every one to two years afterward.

Discussion

SPS is characterized by malignant transformation of hyperplastic polyps into serrated carcinoma through the serrated pathway. As for epidemiology, there is no sex difference, the mean age at the time of diagnosis is 55 years, and 10% to 50% of SPS patients have a family history of colorectal cancer [6].

Despite the small number of reported SPS cases, there are sporadic reports on the risk of developing colorectal cancer and concomitant tumors in other organs. These polyps usually occur on the right side of the colon, and right-sided lesions have malignant potential [7]. It is estimated that 25% to 70% of SPS patients develop colorectal cancer [3], and those with more polyps are more likely to develop this malignancy [8]. Moreover, a retrospective cohort study showed that patients with a first-degree family history of SPS are more likely to develop colorectal cancer [9]. Thus, we consider meticulous follow-up and medical care to be necessary not only for SPS patients but also their relatives.

Immunological fecal occult blood testing has a high diagnostic accuracy for the detection of colorectal cancer [10]. However, because serrated polyps are unlikely to cause bleeding, fecal occult blood testing is reportedly less appropriate for screening of early colorectal cancer in SPS patients than in those at risk for other forms of this cancer [6]. Although neither a screening method nor an optimal therapeutic strategy for SPS has as yet been established, the following regimens have been proposed. (1) After clearing of all polyps ≥5 mm, colonoscopy should be performed every one to three years depending on number and

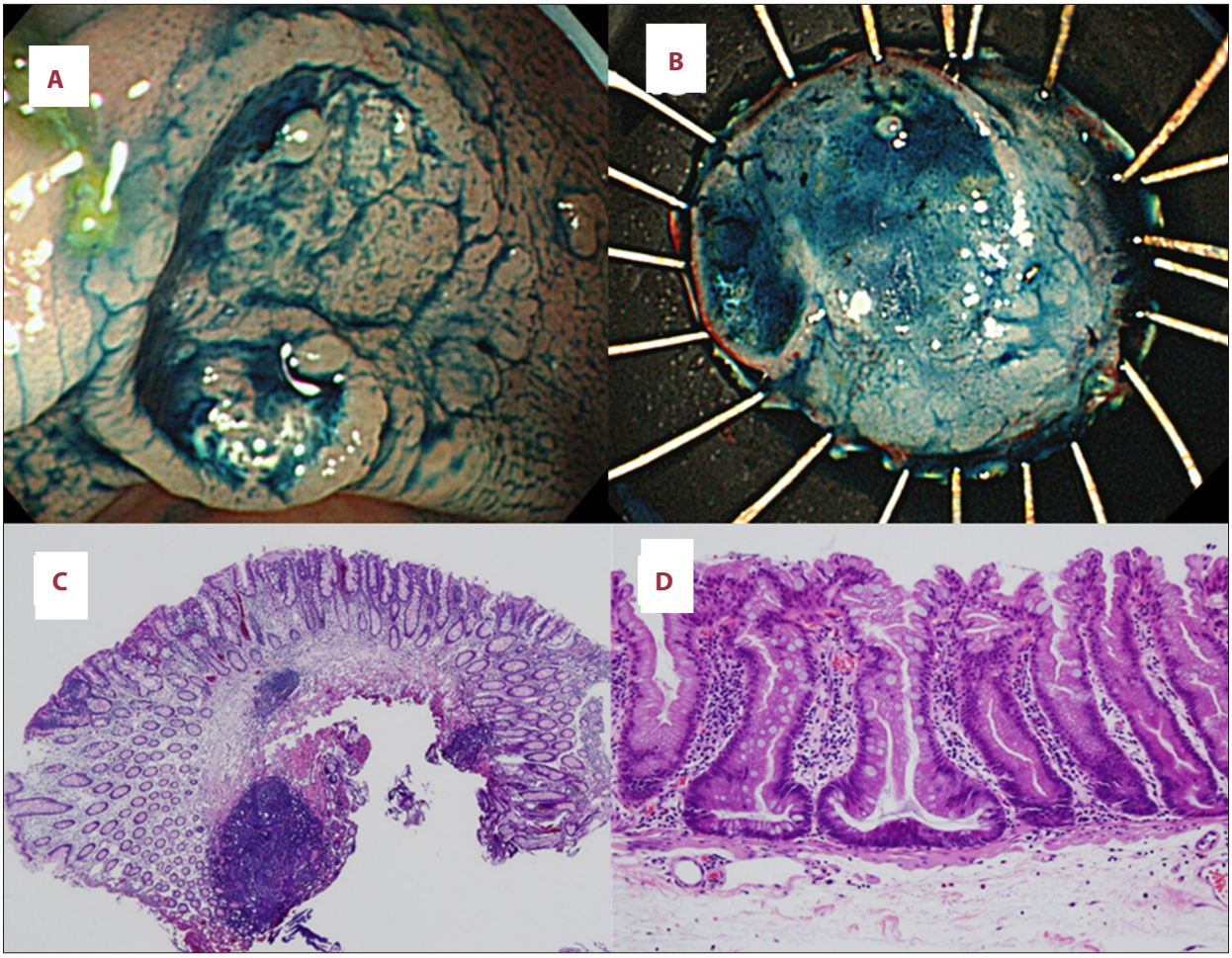

Figure 1. Endoscopic submucosal dissection of a sessile serrated adenoma/polyp. (**A, B**): A 30-mm type IIa+IIc lesion in the ascending colon. (**C, D**): The histopathological examination of the resected specimen revealed proliferation of serrated epithelium in the mucosal layer, an enlarged and irregularly branched crypt, and deformation of the crypt base into the shape of an inverted T. The histopathological examination of the resected specimen diagnosed as SSA/P.

size of polyps. (2) In patients with a first-degree family history of SPS, screening colonoscopy should be started at the earliest of the following: age 40, same age as youngest diagnosis of serrated polyposis if uncomplicated by colon cancer, or ten years earlier than earliest diagnosis in family of colorectal cancer complicating serrated polyposis. Following baseline examination, colonoscopy should be repeated every five years if no polyps are found or every one to three years if proximal serrated polyps or multiple adenomas are found. (3) When control by endoscopic treatment is difficult because of large or multiple tumors, or if high grade dysplasia occurs, colectomy should be considered [11]. Because colorectal polyps in SPS are accompanied by genetic abnormalities [5] and may occur metachronously at multiple sites, continuous endoscopic surveillance is essential. However, the diagnosis of SPS is often missed because flat lesions are not easy to recognize, SPS is a relatively new disease entity, many endoscopists are unaware of this syndrome, and the definition is usually reached only after multiple colonoscopies and histology reports, performed by different people. Therefore, we should keep in mind that we need to observe carefully and check previous colonoscopy findings and histology reports when we perform colonoscopy.

Conclusions

In our case, although multiple applications of endoscopic treatment were required for a relatively long period of time, the lesions were controllable employing this modality. In addition, our patient preferred endoscopic treatment. While he undergoes strict endoscopic surveillance follow-up, we intended to endoscopically treat all indicated lesions in the future. Although several issues, such as long-term prognosis and optimal therapeutic strategies for SPS, await resolution, multiple and continuous applications of endoscopic treatment also appears to be a useful option for managing SPS in cases such

as our patient with lesions that appear to be controllable employing this modality to avoid colectomy. We have to be aware of this syndrome, because patients with SPS are prone to develop colorectal cancer.

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Conflicts of interest

The authors have no conflicts of interest to disclose.