A solitary rectal juvenile polyp with chicken skin-like changes in the surrounding mucosa in an adult: A case report

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Abstract. The majority of colorectal polyps in adults are adenomatous polyps, while hamartoma polyps are rare. Juvenile polyps are the most common type of polyp in children; however, they are rare in adults. Fecal calprotectin (FCP) is commonly elevated in inflammatory bowel disease and is rarely studied in juvenile rectal polyps. Reports of elevated FCP in solitary juvenile rectal polyps of adults are rare. A 57-year-old female was admitted to The Affiliated Hospital of Qingdao University (Qingdao, China) for treatment due to intermittent stool with mucus and blood. Colonoscopy revealed a solitary polyp in the rectum with a diameter of ~2.0 cm, a short and wide subpedicle, with congested and swollen mucosa on the surface and chicken skin-like changes in the surrounding mucosa. The patient had no family history of colorectal polyps or cancer. Endoscopic submucosal dissection was used to remove the polyp. Histopathological examination indicated that the polyp was a juvenile polyp and no signs of malignancy were found. The present case report describes details on this case of an adult patient with a solitary juvenile rectal polyp with chicken skin-like changes in the surrounding mucosa and high FCP.

Introduction

Juvenile polyposis syndrome (JPS) is an autosomal dominant genetic disease with multiple hamartoma polyps in the gastrointestinal tract and the estimated incidence of the syndrome ranges between 1 in 16,000 and 1 in 100,000 worldwide (1). JPS is closely related to the risk of gastric cancer and colorectal cancer (2). JPS is clinically diagnosed in an individual with any of the following: i) ≥5 Colorectal juvenile polyps; ii) juvenile polyps in other parts of the gastrointestinal tract; or iii) any number of juvenile polyps and ≥1 affected family member (1). A total of ≤60% of individuals with clinically defined JPS carry variants in SMAD4 or bone morphogenetic protein receptor type 1A genes, and ~20-50% of JPS cases have no family history (3-5). The pathological mechanism underlying JPS frequently involves the epithelium of the polyps becoming ulcerated, which leads to the infiltration of inflammatory cells-this is the first step in a series of sequential events (2). As the glands and crypts of the juvenile polyp begin to fill with mucus, the polyp becomes inflamed and enlarged, progressing to the classic hamartomatous juvenile polyp (4,5). Finding an early, effective method of identifying JPS is becoming an urgent clinical issue. Patients with JPS are usually asymptomatic, but in the case of symptoms, rectal bleeding with concurrent anemia is most common, followed by abdominal pain, diarrhea and eventually intussusception at the site of the largest polyps (6). Colonoscopy should start at the age of 15 years (or sooner if symptomatic) and then repeated at a 1-3-yearly interval, and genotype-specific recommendations may be required (6). The majority of cases may be managed with therapeutic endoscopy with polypectomy; however, surgery is necessary for patients who develop cancer or where endoscopic management is not possible (7).

By contrast, solitary JPS is frequently less predictive of a malignant tumor tendency (8). Juvenile polyps of the colorectum are the most common type of polyp in children but are rare in adults (9,10). The present case report provides details on the diagnosis and treatment process of an adult solitary juvenile rectal polyp with elevated fecal calprotectin (FCP).

Case report

Chief complaints. A 57-year-old female was admitted to The Affiliated Hospital of Qingdao University (Qingdao, China) for treatment, presenting with intermittent stool with mucus and blood on December 30, 2021.

History of present illness. The patient complained of intermittent excretion of mucus in the past month, covered with a small amount of dark red blood stains, and the initial time of appearance was unknown. The patient did not have any

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Abbreviations: JPS, juvenile polyposis syndrome; FCP, fecal calprotectin; IBD, inflammatory bowel disease; ESD, endoscopic submucosal dissection; NBI, narrow-band imaging; H&E, hematoxylin and eosin

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symptoms of abdominal pain or diarrhea, and colonoscopy indicated that the colonic mucosa was smooth, the submucosal vascular network was clear and the colonic folds were regular. The colonoscopy result excluded a diagnosis of enteritis.

History of past illness. The patient had a history of hypertension and had not been given standardized treatment.

Family history. The patient had no family history of colorectal polyps or cancer.

Physical examination upon admission. The physical examination revealed no positive signs of the heart, lungs and abdomen.

Laboratory examinations. Blood analysis indicated red blood cell, white blood cell and platelet counts within the normal ranges. Prothrombin time, partial thromboplastin time and D-dimer level were normal. A fecal occult blood test had a positive '+' result and FCP was elevated to 805 mg/kg (normal range, 0-49 mg/kg). Indicators of thyroid function, rheumatism-related indices, serum tumor markers, urinalysis and electrocardiogram were all normal.

Imaging examinations. The abdominal ultrasound indicated mild fatty liver (data not shown). Echocardiography was normal. Chest CT revealed multiple small nodules in both lungs and regular follow-up was recommended. The scan parameters were as follows: Tube voltage, 100 kV; tube current modulation, 200-480 mAsec; spiral pitch factor, 0.98; collimation width, 0.625; and gantry rotation time, 0.6 sec.

Further diagnostic work-up. Colonoscopy revealed a solitary polyp in the rectum, ~2.0 cm in diameter, with a short and wide pedicle, hyperemia and edema of the mucosa on the surface of the polyp, plus mild lobulation (Fig. 1). Observations using narrow-band imaging (NBI) indicated irregular expansion of glands on the lesion surface, and blood vessels were unclear (Fig. 1).

Treatment. Polyps were successfully removed by endoscopic submucosal dissection (ESD; Fig. 1).

Hematoxylin and eosin (H&E) staining. A 5-mm section was prepared from the paraffin-embedded polyp and H&E staining was performed for histological analysis. In brief, fresh tissue was fixed with 4% paraformaldehyde for 24 h at room temperature, then dehydrated with a graded alcohol series (75% alcohol for 4 h, 85% alcohol for 2 h, 90% alcohol for 2 h and 95% alcohol for 1 h), absolute ethanol II for 30 min, alcohol benzene for 5-10 min, xylene for 5-10 min and wax for 3 h. The wax-soaked tissue was embedded and stored at -20˚C. Following the solidification of the wax, the tissue was paraffin-embedded and sliced. At room temperature, the sliced tissue was stained the hematoxylin for 5 min, then washed with water for 10-30 sec, and with 0.5% eosin added for 1-3 min, then washed with water again for 1-2 sec, followed by absolute ethanol II and xylene washed for 2 min. Finally, the slide was sealed with neutral gum at room temperature. The H&E-stained sections were imaged with a light microscope (model, BX51; Olympus Corporation).

Outcome and follow-up. Histopathological examination revealed that the polyp was an inflammatory hyperplastic polyp with surface erosion and granulation tissue hyperplasia, surface ulceration (arrow 1), slightly cystic expansion of glands in polyps (arrow 2), abundant polyp stroma (arrow 3), a large number of inflammatory cells (arrow 4) and proliferation of capillaries (arrow 5), and it was diagnosed as a juvenile polyp (Fig. 2A and B). The histological appearance of chicken skin-like changes surrounded this juvenile polyp, including densely deposited foamy macrophages located in the lamina propria (arrow 1), interstitial edema (arrow 2), telangiectasia (arrow 3), glandular papillary hyperplasia (arrow 4) and massive inflammatory cell infiltration [e.g., eosinophils (arrow 5), neutrophils (arrow 6), lymphocytes (arrow 7) and plasma cells (arrow 8)] (Fig. 2C and D). Following the Chinese ESD guidelines (11), endoscopic polyp removal was deemed appropriate, since the patient was middle-aged, had no other comorbidities and exhibited only one polyp. ESD is recommended because it takes a short time (~10 min), does not damage the muscular layer and there is no risk of perforation. The patient did not use any other drugs, such as antibiotics, to alter the number of intestinal neutrophils or intestinal flora after polypectomy. Following polypectomy, food intake was forbidden for 12 h, followed by a liquid diet, and the patient was discharged without any discomfort. At the clinical review 2 weeks later, the patient was no longer experiencing the above symptoms and the FCP level had returned to 36.2 mg/kg. Repeat colonoscopy 6 months later indicated that the local mucosa was well-healed after rectal ESD. Repeat abdominal CT indicated mild fatty liver.

Discussion

JPS is a relatively rare autosomal dominant genetic disease characterized by scattered juvenile polyps in the gastrointestinal tract and is closely related to the risk of gastrointestinal tumors (2). By contrast, colorectal sporadic juvenile polyps are not associated with an increased risk of tumors (12). Sporadic juvenile polyps may also undergo dysplastic changes (13,14). Previous research indicated that juvenile polyps occur in 2% of children and adolescents, accounting for the majority of childhood polyps (12,15). The most common types of polyps in adults are hyperplastic polyps and adenomas. Most sporadic juvenile polyps occur in the sigmoid colon, followed by the rectum (14). Patients with juvenile polyps have symptoms, such as rectal bleeding and abdominal pain, which are similar to those of other types of polyp (14,16). The size of juvenile polyps ranges from mm for sessile nodules to cm. Larger polyps frequently appear leafy, and even a small amount may be observed and is frequently accompanied by erosion and granulation tissue hyperplasia (16). In the present case, chicken skin-like mucosal changes were found to be distributed around the juvenile rectal polyp; however, chicken skin-like changes most often appear in advanced colorectal adenomas (17,18). Chicken skin-like changes with adenocarcinoma or high-grade dysplasia exhibit much higher infiltration of lipid-laden macrophages (17,18). It is assumed that lipid-laden macrophages move toward the tumor and infiltrate the area around it. This phenomenon may explain the extent of inflammatory activity

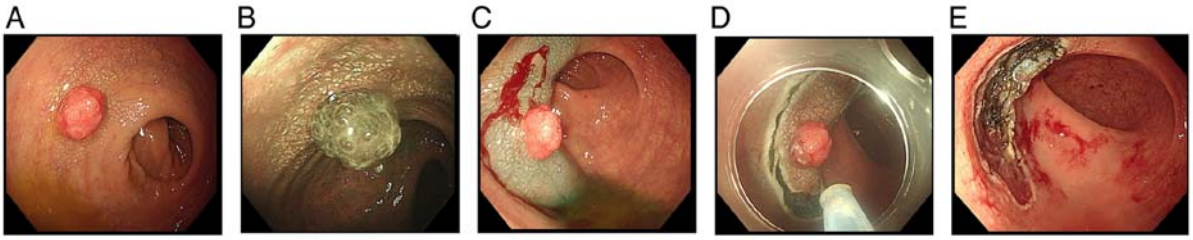

Figure 1. Endoscopic images. (A) White light endoscopy. (B) Narrow-band imaging endoscopy. (C) Submucosal injection. (D) Cut around the polyp and dissection. (E) The site after dissection.

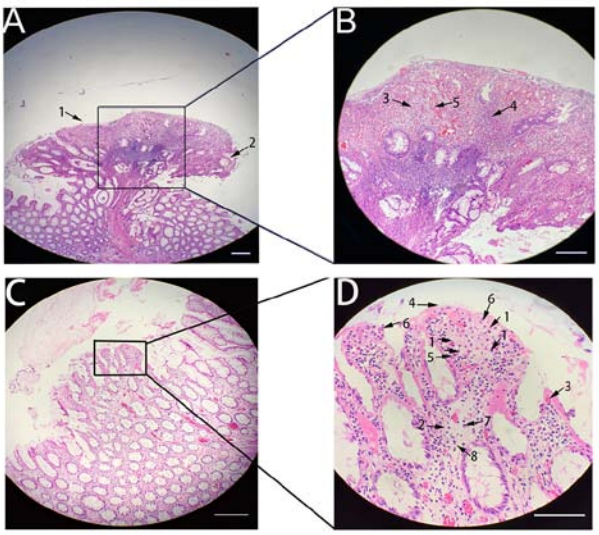

Figure 2. Histological images of the polyp and the surrounding area. (A) H&E staining of the juvenile polyp showed an inflammatory hyperplastic polyp with surface erosion and granulation tissue hyperplasia, surface ulceration (arrow 1), slightly cystic expansion of glands in polyps (arrow 2) (magnification, x10). (B) In the magnified window from A, abundant polyp stroma (arrow 3), a large number of inflammatory cells (arrow 4) and the proliferation of capillaries (arrow 5) were observed (magnification, x40). (C) Histological appearance of chicken skin-like changes surrounding juvenile polyps (magnification, x40). (D) The magnified window indicates densely deposited foamy macrophages located in the lamina propria (arrow 1), interstitial oporotic edema (arrow 2), telangiectasia (arrow 3), glandular papillary hyperplasia (arrow 4) and massive inflammatory cell infiltration [e.g., eosinophils (arrow 5), neutrophils (arrow 6), lymphocytes (arrow 7) and plasma cells (arrow 8)] (magnification, x200; scale bars, 50 μm).

and carcinogenetic progression of the tumor (18). In the present case report, the juvenile polyp was a benign polyp, and a large number of inflammatory cells, including macrophages, were observed in the pathological tissue. Therefore, it was hypothesized that its appearance may have been caused by repeated long-term inflammatory activity of the juvenile polyp. A juvenile polyp was diagnosed mainly by histopathology. In the present case, the types of macrophage, lymphocyte and neutrophil cells in the pathology specimen was able to be separated under the microscope and immunohistochemistry was to be considered if a combination of lymphatic hematopoietic system diseases was indicated by microscopy; however, the cell distribution of this specimen was loose and diverse under the microscope and did not support lymphatic hematopoietic system diseases. The mucosal epithelium and glands of the specimen were not found to be dysplastic under the microscope, and thus, performing immunohistochemistry to detect potential markers of colorectal cancer was not deemed necessary.

In the present case report, under white light endoscopy, a subpedicle-like polyp with a size of ~2.0 cm was observed in the rectum, with hyperemia, nodules and unevenness on the surface and chicken skin-like changes in the surrounding mucosa. Similarly, observations using NBI indicated irregular expansion of glands on the lesion surface, and blood vessels were unclear. Based on the aforementioned considerations and Kudo classification as suspicious V_i type (19), it may be hypothesized that these appearances provide most doctors with the initial impression of potential early rectal cancer, whereas the final pathology results suggest juvenile polyps. Therefore, an important finding of the present case report is the indication that for adult colorectal lesions, in addition to the common adenomatous polyps, there is also a possibility for diagnosis of the relatively rare lesion that is a hamartomatous polyp. A key feature of juvenile polyps is that the surface may have congestion, swelling and nodular changes. The glandular dilatation indicated by NBI is inflammatory dilation rather than tumor glandular dilatation, and the unclear display of blood vessels may be due to severe swelling of the inflammatory blood vessels and not the disappearance of tumor blood vessels.

Calprotectin is a nonglycosylated human protein with a molecular weight of 36.5 kDa found in high concentrations in the cytosol of neutrophil granulocytes (20,21). Currently, FCP is recommended as an aid in distinguishing between inflammatory bowel disease (IBD), such as Crohn's disease or ulcerative colitis, and non-IBD, such as irritable bowel syndrome (20). The present case study suggests that elevated FCP levels may be related to the presence of juvenile polyps in adults, since after endoscopic resection, FCP was found to be within the normal range. This is consistent with a previous report by Hodgson-Parnell *et al* (21). In the present case, elevated FCP was considered to be associated with repeated inflammatory stimuli of juvenile polyps, where neutrophils aggregated and were released. However, to the best of our knowledge, reports on the relationship between FCP and adult juvenile polyps are currently scarce (21), and further research is required to determine this relationship and the underlying mechanism.

In conclusion, the case of an adult patient with a solitary juvenile polyp of the rectum was reported, with chicken skin-like changes in the surrounding mucosa and a high FCP level. The polyp was ~2.0 cm in diameter and had a short and wide pedicle. This polyp was successfully removed by ESD without any complications and the patient will be thoroughly followed up after one year. The correlation between juvenile polyps and FCP requires further study.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

XS and TS designed and supervised the study. XS, YH and XS performed the data collection and analysis. XS, MS and XS conducted data analysis and interpretation. All authors confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

The study procedures were performed in accordance with protocols approved by the Medical Ethics Committee of The Affiliated Hospital of Qingdao University (Qingdao, China).

Patient consent for publication

The patient provided written informed consent for publication of this case report and all the accompanying images.

Competing interests

The authors declare that they have no competing interests.

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