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Solitary Peutz-Jeghers Type Polyp- A Rare Presentation

Dr. Bhawna Sahu, Pathology Resident
Dr. Sadhana Mahore, Professor and HOD
Dr. Sabiha Maimoon, Professor
Dr. Pragati Karmarkar, Associate professor
Department of Pathology, NKP Salve Institute of Medical Sciences and Research Centre, Digdoh Hills, Nagpur 440019 Maharashtra.

Correspondence Author: Dr Bhawna Sahu, Department of Pathology, NKP Salve Institute of Medical Sciences and Research Centre, Digdoh Hills, Nagpur 440019 Maharashtra.

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Abstract

solitary Peutz-Jeghers type is a hamartomatous polyp without associated mucocutaneous pigmentation or a family history of Peutz-Jeghers Syndrome. Peutz-Jeghers type hamartomatous polyps are associated with a lower risk of cancer and are regarded as a different disorder as compared to peutz-jeghers syndrome. We described a case of 27-year-old young man who had complaint of abdominal pain. Clinically diagnosed as intussusception with large polyp, suspicious of malignancy but histopathological examination revealed PJ type hamartomatous polyp with foci of mild dysplastic changes.

Keywords: Solitary Peutz-Jeghers Polyp, Hamartomatous Polyp, Intestinal Polyp.

Introduction :

Various polypoidal lesions of intestine are inflammatory, hamartomatous and neoplastic (benign or malignant) And they vary considerably in size from less than 1 cm to more than 3.5cm. ususally bigger the polyp, greater the risk that it is cancerous or likely to become cancerous. Hamartomatous polyps are a rare entity. They may be solitary (solitary Peutz-Jeghers polyp or solitary juvenile polyp) or multiple. Peutz-Jeghers syndrome (PJS) is characterized by the familial occurrence of gastrointestinal hamartomatous polyps in association with mucocutaneous hyperpigmentation. A hamartomatous polyp without associated mucocutaneous pigmentation or a family history of PJS is a solitary Peutz-Jeghers type hamartomatous polyp[8]. As compared with PJS, Peutz-Jeghers type hamartomatous polyps are known to be associated with lower risk of cancer[1]. Here we are reporting a case of large polypoidal mass of jejunum with intussusception which later diagnosed as hamartomatous polyp.

Case Report

27-year-old young male came with complaints of abdominal pain. Abdominal examination revealed epigastric tenderness with mass like feeling. Abdominal ultrasonography suggestive of intussusception. Other lab investigations were unremarkable. Patient underwent an exploratory laparotomy, jejuna-jejunal bowel intussusception was noted 10 cm from duodenojejunal junction with single large polyp as a leading mass.

Reduction of jejuna- jejunal bowel intussusception, bowel resection (along with polyp) was done. No other polyps were seen. Clinical diagnosis was kept as intussusception with large polypoidal mass. Considering large size, clinical suspicion of adenocarcinoma was also made. Specimen composed of part of jejunum measuring - 12x5.5x2.5. cm along with pedunculated lobulated polyp measuring 5x4x4cm. polyp was seen occluding the jejunal lumen. Cut surface was pale and showed muscularis layer invaginating into the villi on magnification. Rest of the small intestine was slightly congested. Histological examination of specimen obtained from the lesion revealed polypoidal mass with arborizing pattern of smooth muscle, covered by mucin secreting columnar epithelium with round to oval uniform nuclei. Focal area showed depletion of mucin, hyperchromatic nuclei and mild anisonucleosis. Underlying fibromuscular tissue showed inflammatory infiltrate composed of neutrophils, lymphocytes and eosinophil. Other part of intestine showed unremarkable histology.

Discussion

Polyps of the intestine or rectum, may be benign, precancerous (adenomatous), or cancerous (malignant carcinoma). Usually hamartomatous polyps are 0.5cm to 4cm and benign in nature. The larger the polyp, the greater the potential for malignancy. it may be solitary or multiple. Peutz-Jeghers syndrome (PJS) is a rare autosomal dominant disorder characterized by hamartomatous polyps throughout the gastrointestinal tract and characteristic mucocutaneous pigmentation, primarily of the lips, oral and gingival mucosae. Polyps are found throughout the gastrointestinal tract but most common to the small bowel (60% to 90%) and the colon (50% to 64%)[3] . As compared with PJS, Peutz-Jeghers type hamartomatous polyps are diagnosed at a more advanced age, in the absence of mutation of the STK11/LKB-1 gene, and without familial history or mucocutaneous pigmentation [1]. PJ polyps are generally Hamartomatous polyps are generally considered to have very low malignant potential [4,7,9]. Solitary hamartomatous polyp has been considered a variant or a separate disease entity[6]. Peutz Jeghers (PJ) type of polyp is characterized histologically by tree-like branching of smooth muscle fibres, with a core of smooth muscle covered by mucosal tissue of near normal appearance. Solitary PJ type polyps have been described as having a lobular or nodular surface, whitish colour and whitish spots on the surface. Microscopically, extensive smooth muscle proliferation, with an elongated, arborizing pattern of polyp formation is seen. This characteristic microscopic appearance of PJS polyps enables experienced gastrointestinal pathologists to confirm the clinical diagnosis[5]. In our case as the polyp was with arborizing smooth muscle with focal dysplastic changes not involving submucosa and muscularis propria. however because of low malignant potential, a solitary PJ hamartomatous polyp should be removed. A variety of polypoidal lesions can be found in the jejunum and these lesions can be distinguished based on endoscopic appearance, Endoscopic Ultrasound and histology. Biopsy is recommended to determine histology, prognosis and thus guide management [10].

Conclusion

Usually peutz jeghers hamartomatous polyps are small in size but solitary hamartomatous polyp can be present as large polypoidal mass giving suspicion of adenocarcinoma that is why It is important to examine histopathologically as solitary hamartomatous polyps are of low malignant potential and have different treatment modality. So all the polyps should undergo gastrointestinal endoscopy, biopsy and histopathology to rule out malignancy. These polyps can be treated with endoscopy or surgery depending on size, depth of lesion and evidence of malignant transformation on biopsy.

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Figure:

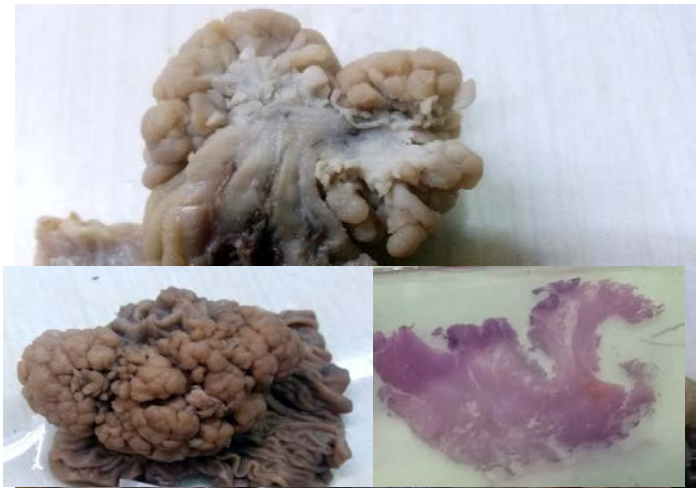

Figure 1 : Photograph **a**-pedunculated polypoidal mass from mucosal surface **b** and **c** cut surface of polyp showing arborising muscular layer.

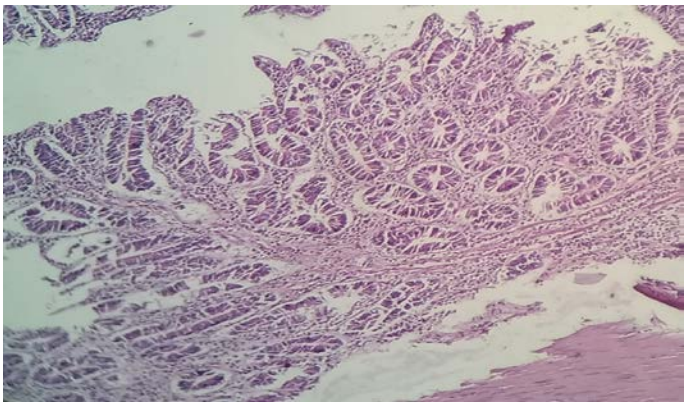

Figure 2: Microphotograph H & E stain showing arborising smooth muscles in polypoidal mass and covering. Epithelium with mild dysplasia highlighted in inset.

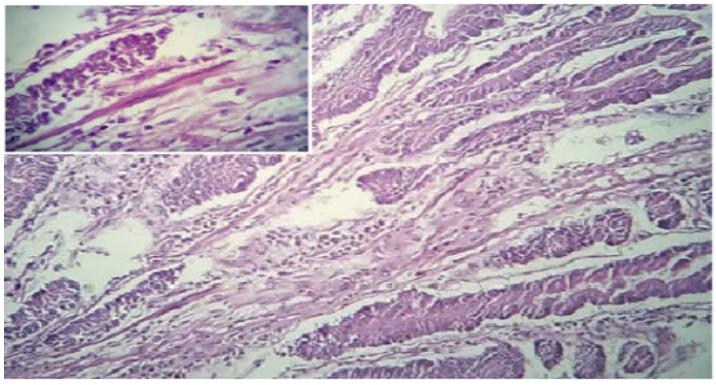

Figure 3: Microphotograph -H&E stain-(10x) arborizing villi showing muscularis layer in stalk. Inset- (40x) muscle fiber of stalk