Rare Colonic Polyps: A Case of Langerhans Histiocytosis

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Background

- Langerhans Cell Histiocytosis (LCH) is a rare inflammatory myeloid disorder mostly seen in children <5.
- Clinical presentation is heterogeneous with potential for wide-ranging organ involvement, primarily in the bone, skin, and lungs[1].
- Review of literature shows less than twenty adults with incidences of GI tract involvement of LCH and only four cases of adults with LCH colon polyps[2].

Case Presentation

- 66-year-old man presented for screening colonoscopy.
- He had no significant medical history and was in generally good health without any GI or systemic symptoms.
- On colonoscopy, six benign-appearing polyps were removed using cold snare polypectomy
  - Three from the ascending colon (largest 0.5 cm in size)
  - One from the splenic flexure of the transverse colon (0.4 cm in size)
  - Two from the rectum (largest 0.4 cm in size).

Pathology and Management

- The rectal polyps were hyperplastic. Submucosal sections of the ascending and transverse colon polyps stained positive for CD1a, CD68, and S-100, consistent with Langerhans-type cells.
- The LCH diagnosis was further supported by irregular nuclei within focal nuclear grooves and an eosinophilic cytoplasm. Genomic testing of the colon sample revealed BRAFV600E mutation, the most recognized targetable mutation in LCH. He underwent further testing with PET scan and bone marrow biopsy, both of which were negative for LCH.
- Surveillance colonoscopy was recommended in 7 years.
- With his lack of systemic disease and asymptomatic presentation, watchful waiting and Oncology follow-up was recommended.

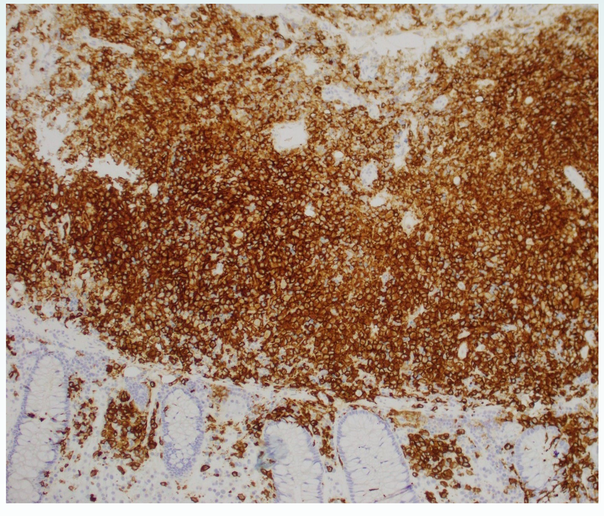

Figure 1. Positive immunostaining for CD1a, classically expressed by Langerhans cells.

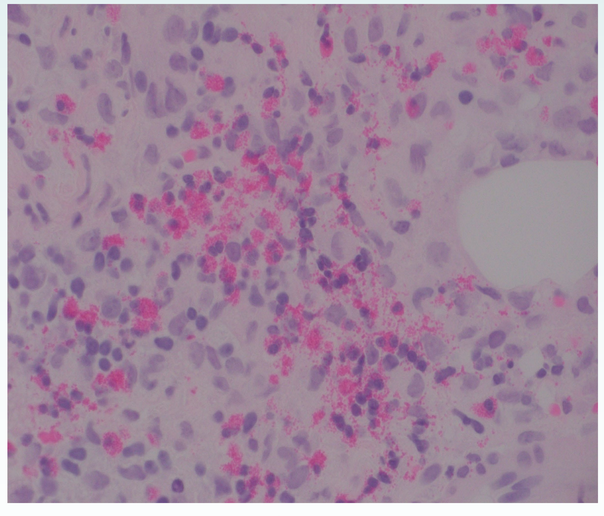

Figure 2. H&E Stain of colonic submucosal infiltration by Langerhans cells under high-power field to showcase eosinophils.

Discussion

- GI tract involvement is very rare in LCH, and in symptomatic patients, etiology could be confused with infectious, allergic, and autoimmune bowel diseases.
- Given the unusual diagnosis of LCH via a colon polyp, it is possible for lesions to be clinically unsuspected and histologically overlooked.
- With increasing rates of screening colonoscopy, more polyps demonstrating LCH on histopathology may be identified.
- Reports of LCH in the GI tract and colon polyps are infrequent and characterization of outcomes will rely on further cases in this population.

References

1.Krooks J, Minkov M, Weatherall AG. Langerhans cell histiocytosis in children: History, classification, pathobiology, clinical manifestations, and prognosis. J Am Acad Dermatol. 2018 Jun;78(6):1035-1044.
2.Matsubara Y, Kobayashi M, Hijikata Y, Ota Y, Hirata Y, Lim LA, Yotsuyanagi H, Tojo A. Gastrointestinal lesion in adult-onset Langerhans cell histiocytosis. Int J Clin Oncol. 2020 Nov;25(11):1945-1950.

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