

Case report:

A case of Angiolymphoid hyperplasia with eosinophilia in proximal ascending colon mimicking colon carcinoma

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Abstract:

Angiolymphoid hyperplasia with eosinophilia (ALHE) / Epithelioid hemangioma (EH) is a rare lesion commonly located in skin but arising in colon even rarer. It clinically follows a benign course of uncertain pathogenesis characterized histologically by plenty of proliferation of histiocytoid endothelial cells of blood vessels and infiltration by dense eosinophil, lymphocytes and plasma cells. The lesion may recur after excision but there is no report of metastasis. A case of sixty-four years male with severe pain in right iliac fossa with bloody stool had CT scan and noticed ulceration with bleeding sites on colonoscopy. Histopathological examination showed characteristic features of ALHE /EH in common location like skin. The lesion very often mimic as bleeding colonic carcinoma which necessitate colectomy. This is a rare case of ALHE /EH in proximal ascending colon.

Keywords: angiolymphoid hyperplasia with eosinophilia, colon.

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Introduction:

Angiolymphoid hyperplasia with eosinophilia (ALHE) was termed as epithelioid hemangioma (EH) by Weiss and Enzinger in 1982¹, is a rare condition presented mostly in the skin surrounding head & neck area and also documented unusual sites in hand, tongue, breast and vulva². This lesion occurring in gastrointestinal tract is very rare. Here we are presenting a case of ALHE arising in ascending colon of ilio cecal region because of rarity that very often misdiagnosed as malignancy.

Case Report:

A sixty four years male had occasional severe pain in right iliac fossa for five months, change of bowel habits having passage of stool mixed with blood and mucus for last two weeks and attended in outside surgical clinic in the month of August 2015.

Clinical examination: Loss of weight by three kg,

pallor and moderately diffuse pain and slightly firm ill-defined fullness in the right iliac fossa without any abnormalities of other systems.

Routine laboratory investigation: TLC $11 \times 10^9 / l$, $N_{78} / L_{19} / E_2 M_1 B_0$ and ESR 18mm 1st hr. Blood biochemistry and chest X-ray was within normal limit.

CT Scan of whole abdomen: Concentric gut wall thickening at right iliac fossa involving cecum and proximal ascending colon. Reported a case suspected malignancy. Colonoscopy and biopsy was advised urgently.

Colonoscopy Report: Tiny hemorrhagic nodular ulcer and stricture formation surrounding the ulcer. Rest of the mucosa was normal. The case was reported as intestinal tuberculosis and urgent biopsy was taken to exclude malignancy.

Endoscopic biopsy report: Histopathological features of ulceration of colonic mucosa and inflammatory

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granulation tissue formation, infiltration of lymphocytes, plasma cells, eosinophil and histiocytes in the lamina propria. No granuloma and malignancy seen.

The case was reviewed in the department of pathology Bankura Sammilani Medical College, West Bengal for evaluation and further proper management .

Review report: Several serial tissue sections from paraffin block, hematoxylin & eosin stain , PAS stain and reticulin stain showed histopathological features of colonic tissue with mucosal ulceration. The lamina propria show dense infiltration of lymphocytes, plasma cell, histiocytes, neutrophil and plenty of eosinophil surrounding small to medium size blood vessels having plump hobnailed endothelial cell lining. No granuloma, parasites, increased mitosis and malignant transformation were seen. The case was diagnosed as Angiolymphoid hyperplasia with eosinophilia of colon (fig 1, 2, 3).

Discussion:

ALHE was first noted in 1969 by Wells and Whimster³. The lesion usually occurs in adult with female preponderance. The Histopathological features of this case in colonic location are similar finding seen in skin and lymph node. ALHE in large intestine first reported by Berney DM et al in 1997³ and later on Nonose et al (2008)⁴ and Bui MM et al (2010)⁵ also reported such entity in large intestine. ALHE mostly have benign inflammatory vascular disease with characteristic vascular endothelial changes and infiltration of plasma cell, eosinophil and lymphoid proliferation of unknown pathogenesis. Some authors consider as a reactive changes and other as a neoplastic lesions. The exact etiopathogenesis is not known. Some proliferative vascular stimuli associated inflammation and immunological reaction may have a role. Local vascular injury, arteriovenous anastomosis and mutation of TEK gene -endothelial receptor tyrosine kinase receptor Tie-2, T-cell receptor gene rearrangement are possible associated pathogenic factors⁶. Study with larger sample needed for exact evaluation.

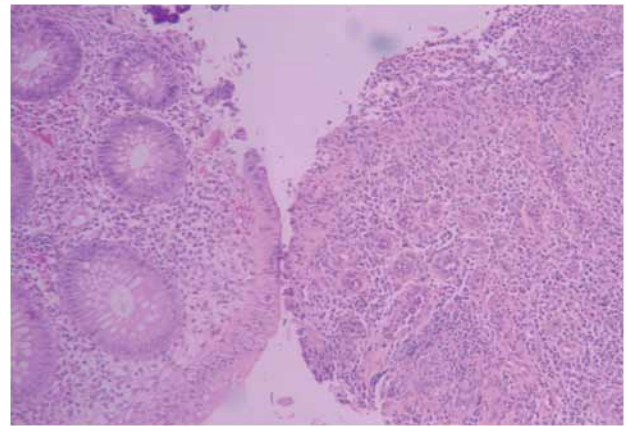
From the Histopathological features, history of intestinal bleeding and anatomical location the most important differential diagnosis of ALHE in ascending colon are Kimura disease, inflammatory fibroid polyp, malignancy eosinophilic colitis, inflammatory bowel disease, tuberculosis, parasitic colitis and rarely

vascular ectasia, vascular malformation- Dieulafoy's disease. The Kimura's disease typically presented with subcutaneous nodular mass, lymphadenopathy and peripheral eosinophilia. Histologically extensive lymphoid follicles with prominent germinal center, eosinophilic infiltration, folliculolysis, micro abscess and attenuation of blood vessels are common features⁷. Absence of such findings with characteristics vascular endothelial hob nailing features excludes kimura's disease in this presenting case. Eosinophilic colitis is characterized by peripheral eosinophilia, transmural dense infiltration of eosinophil absence vascular changes excludes in our case. Absence of spindle cell proliferation mixed with inflammation also excludes inflammatory fibroid polyp. Absence of granuloma with or without caseation, presence of parasites, also excludes other differential diagnosis. Rare entity of vascular malformation Dieulafoy's disease clinically presented with gastric mucosal bleeding. CT scan, endoscopic examination and biopsy confirm this rare entity. Due to bleeding nature of this case clinically colonic carcinoma is an important differential diagnosis in which absence of any increase mitosis and nuclear atypia excludes such possibility.

Colectomy is the treatment of choice of such case. The lesion may recur after excision but there was no report of metastasis. This patient had right hemicolectomy and was symptom free after six months follow up.

Conclusion:

Though ALHE /EH is rare in large intestine but it should be kept in mind as a differential diagnosis of common lesion of this particular site during histopathological evaluation.



Figur-1. Angiolymphoid lymphoid hyperplasia with eosinophilia (ALHE) in colonic mucosa. H&E x 100

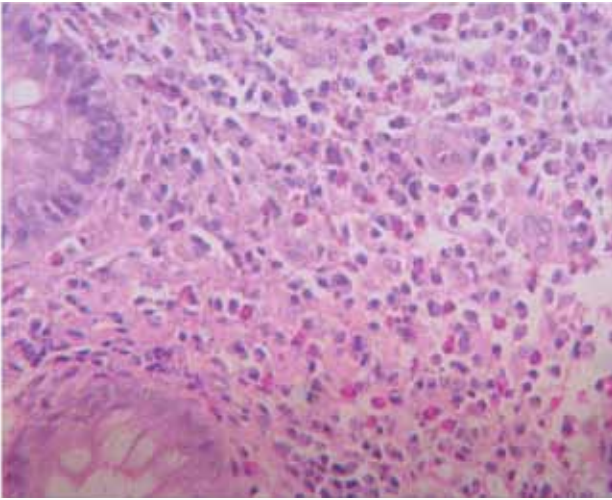


Fig 2. Plenty of lymphocyte, plasma cells, eosinophil with epithelioid blood vessels.H&E x 400.

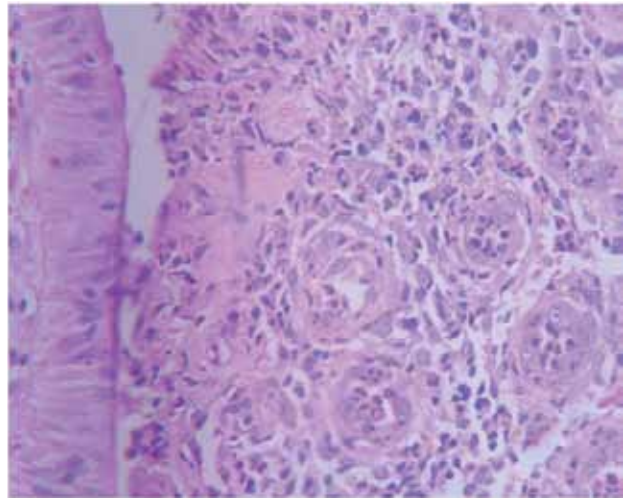


Fig 3.Characteristic hob-nailed changes of endothelial cells of blood vessels in ALHE.H & E x 400

References:

1. Weiss SW, Enzinger FM. Epithelioid hemangioendothelioma: a vascular tumor often mistaken for a carcinoma. *Cancer*. 1982;50(5):970-981.
2. Nair M, Aron M, and Sharma MC. Angiolymphoid hyperplasia with eosinophilia (Epithelioid Hemangioma) of breast : Report of a case. *Surg Today*. (2000); 30: 747-749.
3. Well GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol*. 1969;81 (1): 1-14.
4. Nonose R, Priolli DG, Cardinali IA, et al. Epithelioid hemangioma of the colon: a case report. *Sao Paulo Med J*. 2008; 126(5): 294-296.
5. Bui MM, Draper NL, Dessureault S, Nasir NA, Cooper H, Nasir A, Coppola D. Colonic angiolymphoid hyperplasia with eosinophilia masquerading as malignancy: a case report and review of literature. *Clin Colorectal Cancer*. 2010;9(3): 179-182.
6. Ruifeng Guo, Alde carlo P. Gavino. Angiolymphoid Hyperplasia with Eosinophilia. *Arch Pathol Lab Med*. 2015; 139:683-686.
7. Googe PB, Harris NL, Mihim MC. Kimura's disease and angiolymphoid hyperplasia with eosinophils. Two distinct Histopathological entities. *J Cutan Pathol*. 1987; 14: 263-71.