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Jejunal Plasmablastic Lymphoma in disguise-unmasking a rare entity

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Abstract
Plasmablastic lymphoma (PBL) is a rare variant of diffuse large B cell lymphoma that is aggressive in nature. Initially described in oral cavity with human immunodeficiency virus infection. Extraoral sites include lungs, nasal cavity, lymph nodes and skin with small intestine being a rare site. PBL does not express common lymphoid markers so can be easily misinterpreted as poorly differentiated carcinoma or melanoma or sarcoma. A delayed diagnosis might impact both treatment and survival of patient. This case report underscores the rarity of PBL presenting in the form of intestinal polyposis.

Keywords: Plasmablastic lymphoma, HIV, Small Intestine

Introduction

Plasmablastic lymphoma (PBL) is a rare variant of diffuse large B cell lymphoma that is aggressive in nature. Initially described in oral cavity with human immunodeficiency virus infection. Extraoral sites include lungs, nasal cavity, lymph nodes and skin with small intestine being a rare site [1].

PBL remains a diagnostic dilemma due to morphology and immunohistochemistry profile similar to plasma cell myeloma. It accounts for 2.6% of all AIDS related lymphomas. Due to its aggressive nature and recurrence it poses high rate of fatality despite recent treatment modalities [2].

Here we present a rare case of jejunal plasmablastic lymphoma in a 32-year-old male HIV negative patient presenting as multiple polypoidal growths.

Case Report

A 32-year-old male was apparently normal 15 days back now came with complaints of bloody tarry stools. No history of abdominal pain, distension, fever or hematemesis. Patient had similar complaints 10 months back with generalized weakness for which he was admitted in a nearby private hospital. Ultrasound abdomen showed small bowel intussusception with necrotic retroperitoneal lymphadenopathy. Upper GI scopy showed reflux esophagitis, portal hypertensive gastropathy, H pylori induced pangastritis and hiatus hernia. MDCT Scan abdomen showed enteroenteric intussusception probably secondary to neoplastic etiology. PET CT showed hypermetabolic polypoidal wall thickening in jejunum, hypermetabolic mesenteric, and periportal, small retroperitoneal, mediastinal and right hilar nodes enlarged.

On routine blood investigations patient had low hemoglobin with non-reactive serology status for HIV. Peripheral smear showed microcytic hypochromic blood picture. Surgical resection of jejunum was done and sent for histopathological examination.

Grossly we received a segment of jejunum measuring 9cm in length. Mucosal surface showed presence of three polyps. Largest polyp was close to one of the resected margins of the jejunum measuring 4.5x3.5x2. 5cm.Second polyp measuring 2.5x2.1x2cm at a distance of 4cm from one end. Third polyp was 3.7x2.1x1cm at a distance of 5cm from one end. Cut surface of all polyps showed a grey white homogenous glistening appearance. (Figure 1)

Fig 1: Gross image of the the jejunal polyps

Microscopy of the lesion revealed diffuse sheets of tumor cells having pleomorphic nucleus, vesicular chromatin, prominent nucleoli and scant cytoplasm. Occasional binucleate cells and large, bizarre cells noted. Mitotic figures were 4-5/HPF. Histopathologically a diagnosis of high-grade Non-Hodgkins Lymphoma was given. (Figure 2, 3).

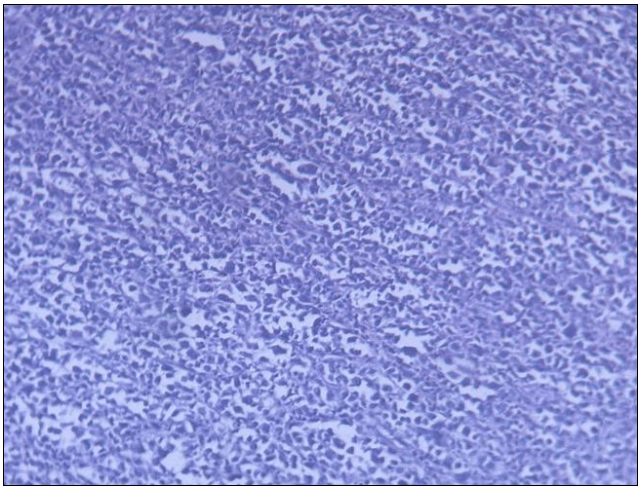

Fig 2: Diffuse sheets of tumor cells (H & E, 100X)

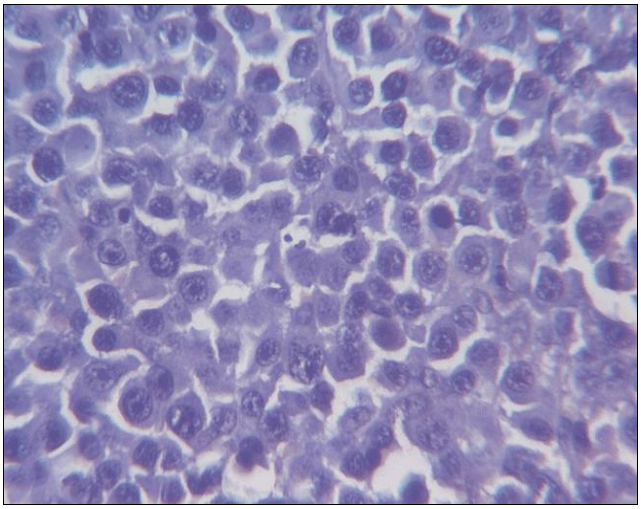

Fig 3: Tumor cells are pleomorphic with vesicular nuclei, prominent nucleoli (H & E, 400X)

Immunohistochemistry (IHC) showed atypical cells positive for LCA, CD10, MUM1(Figure 4), EMA and patchy expression of CD 138(Figure 5). Tumour cells were negative for CD3, CD 20, CD 4, CK, CD5, CD 30, S100 and Vimentin.Ki 67 proliferation index was 80%. Based on IHC findings the diagnosis of Plasmablastic lymphoma was confirmed.

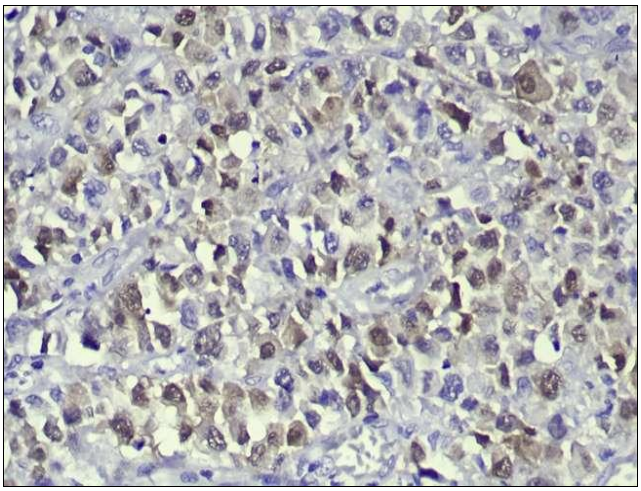

Fig 4: MUM 1 Positive on IHC (400X)

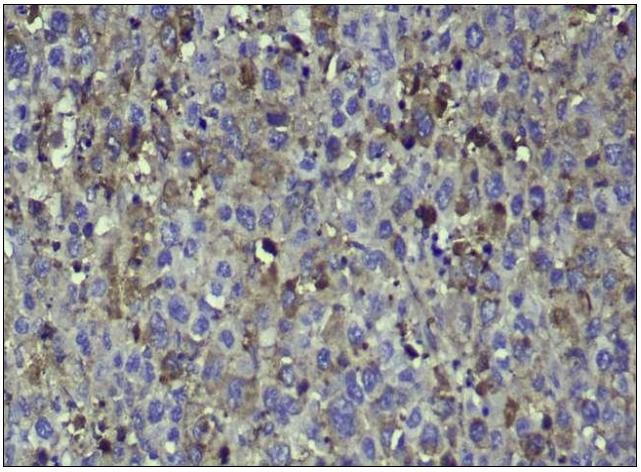

Fig 5: CD138 Patchy positivity on IHC (400X)

Discussion

Plasmablastic lymphoma was first described in 1997 which is known to have a high association with EBV and human herpes virus 8. Although the pathogenesis is uncertain it represents a neoplastic process stemming from post germinal centre B cells [3].

In 2016 PBL was classified by the WHO as an independent subtype of large B cell lymphoma.PBL expresses CD38, CD138, MUM1, EMA with absence of CD20, CD 45.PBL must be differentiated from plasmablastic myeloma (PBM) due to divergent therapeutic approaches.PBM is a variant of multiple myeloma exhibiting proliferation of plasmablasts with clear nuclei, prominent nucleoli, scant cytoplasm and expression of CD38, CD138 with light chain restriction [4].

Gastrointestinal lymphoma is a heterogenous entity which constitute 5-20% of all Non Hodgkins lymphoma and comprises 30-40% of all extranodal lymphomas. Gastrointestinal lymphomas represent 1-10% of all gastrointestinal malignancies. Multiple lymphomatous polyposis is a type of primary gastrointestinal lymphoma with rare involvement of the intestinal segment [5]. The present case also displayed multiple polyps in the small intestine which is a rare possibility.

The tumour usually presents as an annular growth or stricture or intramural lesion. But the present case displayed a rare polypoidal presentation. Clinical symptoms include fatigue, abdominal pain, diarrhea, gastrointestinal tract obstruction, hematochezia and weight loss [6].

PBL is a neoplasm of mature B cells which shows diffuse proliferation of large neoplastic cells most resemble B immunoblasts and have immunophenotype of plasma cells. Valera *et al* opined IG/MYC rearrangement maybe a major cytogenetic alteration in PBL. There is a strong association with EBV in a setting of HIV infection. This case had negative serology status for HIV.EBV plays a role in preventing apoptosis via induction of NF-kB through syk/src mediation and induction of BAX/BAK [7].

The age distribution has been estimated to be 64% with non-HIV PBL over 60 years and 43% between 30-60years.Old age might play a role secondary to age related decreased immune function. PBL has an unfavourable prognosis with HIV positive individuals and are associated with a better response to chemotherapy and longer survival [8].

Similar cases of plasmablastic lymphoma have been reported in the past in literature. Cha *et al* described 60-year-old male with ulcero fungating mass in jejunum.

Bahari *et al* presented a case of small intestinal polyposis in a 17-year-old male. Khera *et al* presented diffuse wall thickening of the jejunal loop and enlarged mesenteric lymph nodes in a 48-year-old male [9].

Conclusion

Plasmablastic lymphoma is a rare aggressive entity commonly seen in immunocompromised patients. PBL does not express common lymphoid markers so can be easily misinterpreted as poorly differentiated carcinoma or melanoma or sarcoma. Awareness of this entity is needed as delayed diagnosis might impact both treatment and survival of patient. PBL of intestine with multiple jejunal polyposis is a rare presentation.

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