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LAPAROSCOPIC SPLENIC BIOPSY – AN UNDERRATED DIAGNOSTIC MODALITY FOR EVALUATING PYREXIA OF UNKNOWN ORIGIN IN CHILDREN

Pediatrics

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ABSTRACT

Introduction: Pyrexia of unknown origin may pose significant diagnostic challenge. We herein report a case of PUO where diagnosis became really difficult. Malignancy can be lurking behind in such cases.

Case description: We present a case 9-year-old boy with pyrexia of unknown origin of 1 year duration. Even after elaborative investigations his diagnosis remained elusive. Eventually laparoscopic splenic biopsy was performed and revealed splenic Hodgkin lymphoma

Conclusion: Laparoscopic splenic biopsy can provide a middle path to reach diagnosis with minimum complications and should be considered early in diagnostic ladder.

KEYWORDS

INTRODUCTION:

Pyrexia of unknown origin (PUO) in may pose significant diagnostic and therapeutic challenge. While a good clinical history, meticulous physical examination and appropriate investigations can provide significant clues, in some situations clinician is faced with a diagnostic challenge. There have been various reports wherein splenic histopathology following splenectomy was able to ascertain the diagnosis especially in occult malignancies (1,2). However, splenectomy is better avoided in children due to risk of fatal infections. Fine needle aspiration cytology (FNAC) of spleen is increasingly being performed but the results are often not satisfactory due to poor yield and risk of bleeding (3). Splenic biopsy and partial splenectomy are other options described in literature and are used as a diagnostic modality in adults but rarely described in pediatric literature. We describe a young boy with PUO, with inconclusive initial investigations including splenic FNAC where laparoscopic splenic biopsy, performed as a diagnostic procedure, helped in ascertaining the diagnosis.

CASE DETAILS:

Y, 9-year-old boy was referred to our unit with fever for 1 year and joint pain for 10 days. He had had recurrent high grade febrile episodes for last 1 year. Each episode lasted 7-10 days followed by afebrile period of 1 to 3 weeks. He also developed pain in multiple joints (right wrist, right knee and both ankle) for 10 days without any swelling or restriction of movements. He had received multiple antimicrobials for these episodes but fever persisted. On examination he had pallor and hepatosplenomegaly. Peripheral lymph nodes were not enlarged. Investigations revealed anemia (haemoglobin- 65 g/L), leucopenia ($3.23 X10^9$/L, P_{48} L_{29} M_{22} E_1) and normal platelet count ($323 X 10^9$/L). Peripheral smear revealed microcytes, macro-ovalocytes, elliptocytes and no atypical cells. Erythrocyte sedimentation rate (ESR-120 mm in 1st hour) and C-reactive protein (CRP-199 mg/L) were elevated. Kidney and liver function tests were normal. Investigations for chronic infections was non-contributory. Considering possibility of lymphoproliferative disorder we performed bone marrow examination that revealed large epithelioid granulomas with few atypical cells. However, immunohistochemistry was not suggestive of malignancy. Though malignancy was a strong clinical possibility, however long duration of illness (1 year) and presence of arthralgia were odd for it. Positron Emission Tomography (PET) scan revealed grossly enlarged spleen with increased uptake (SUV- 10.7) and few enlarged lymph nodes on both sides of diaphragm (SUV- 3.7). Splenic FNAC showed reactive lymphoid cells, polymorphs, eosinophils and immunohistocytic tangles. Few atypical cells were seen but no clonal population identified on flowcytometry. As fever was persisting, with bicytopenia and few atypical cells in splenic FNAC, explorative laparotomy and splenic biopsy were performed (10% splenic tissue removed). Histopathological examination of splenic biopsy tissue revealed binucleate cells against background of mature lymphocytes and eosinophils. Immunohistochemistry showed cells were positive for CD 30, CD 15 and PAX 5 while negative for CD 20, CD 3 and CD 45. These findings were consistent with Hodgkin's lymphoma and thus the diagnosis was made. He was started on chemotherapy that resulted in good outcome.

DISCUSSION:

Occult isolated lesions in spleen due to chronic infections like tuberculosis and lymphoreticular malignancy can present as PUO and can sometimes be difficult to diagnose. Laparoscopic splenic biopsy, though an invasive procedure, may sometimes be decisive in finding etiology in such patients (7). Various procedures for procuring splenic tissue for histopathological examination include splenic FNAC, percutaneous biopsy, laparoscopic biopsy and splenectomy (partial or total). Partial splenectomy involves removal 70-90% of splenic tissue and in laparoscopic biopsy only around 10% of the tissue is removed. Han B et al retrospectively reviewed the advantages of diagnostic splenectomy in adult patients with PUO. They were able to reach diagnosis in significant majority (72%), however 25.9% developed operation related complications and mortality within 1 month postoperative was 16.7% (8). In various series such complications

have been described more in extremes of age group (9,10).To avoid such invasive procedures, percutaneous splenic FNAC and biopsy are other modalities that have been tried as a diagnostic modality in PUO. It can be helpful both in infectious and malignant etiologies (12). However, other studies consider that sample yield is low with this procedure and risk of bleeding and splenic rupture can sometimes be fatal. Total splenectomy has been widely used in adult patients with PUO especially when suspecting splenic malignancy(4,5)

There are hardly any published data on diagnostic utility of laparoscopic splenic biopsy in children with PUO. Splenic biopsy can be safe alternative in children as it preserves a part of spleen and retains its immune function and provides adequate tissue for histological examination. Further perioperative bleeding and complications are also few (7). It is especially useful when image guided procedures do not yield definitive diagnosis.

In the index child, splenic FNAC was uneventful and showed presence of atypical cells but definite diagnosis could not be reached. In such a perplexing scenario laparoscopic splenic biopsy can be a middle path. It can provide adequate tissue for diagnosis and spare some spleen tissue which retains its immune function. In our patient splenic biopsy was uneventful and we were able to reach to diagnosis and start definitive treatment. Thus, laparoscopic splenic biopsy may prove to be important diagnostic tool in children with PUO and splenomegaly. Since early diagnosis is imperative in such patients and risk of overwhelming sepsis after splenic biopsy is low, we advocate early decision of splenic biopsy to avoid unnecessary delay in treatment.

CONCLUSION:

This case highlights that sometimes PUO can become an enigma and reaching to a diagnosis can be really difficult. In cases such as these, malignancy can be lurking behind giving very few clues and causing a delay in diagnosis and treatment. Laparoscopic splenic biopsy can provide a middle path to reach diagnosis with minimum complications and should be considered early in diagnostic ladder.

REFERENCES

1. Sun P-G, Cheng B, Wang J-F, He P. Fever of unknown origin revealed to be primary splenic lymphoma: A rare case report with review of the literature. Mol Clin Oncol. 2017 Feb;6(2):177–81.
2. Zeidman A, Horowitz A, Fradin Z, Cohen A, Wolfson L, Elimelech O. Fulminant Intravascular Lymphoma Presenting as Fever of Unknown Origin. Leukemia & Lymphoma. 2004 Aug;45(8):1691–3.
3. Lishner M, Lang R, Hamlet Y, Halph E, Steiner Z, Radnay J, et al. Fine Needle Aspiration Biopsy in Patients with Diffusely Enlarged Spleens. ACY. 1996;40(2):196–8.
4. Gupta PP, Fotedar S, Agarwal D, Sansanwal P. TUBERCULOSIS OF SPLEEN PRESENTING WITH PYREXIA OF UNKNOWN ORIGIN IN A NON-IMMUNOCOMPROMISED WOMAN. Lung India. 2008;25(1):22–4.
5. Kattepur AK, Rohith S, Shivaswamy BS, Babu R, Santhosh CS. Primary Splenic Lymphoma: A Case Report. Indian J Surg Oncol. 2013 Sep;4(3):287–90.
6. Grosskreutz C, Troy K, Cuttner J. Primary splenic lymphoma: report of 10 cases using the REAL classification. Cancer Invest. 2002;20(5–6):749–53.
7. Dagnini G, Cladironi MW, Marin G, Patella M. Laparoscopic splenic biopsy. Endoscopy. 1984 Mar;16(2):55–8.
8. Han B, Yang Z, Yang T, Gao W, Sang X, Zhao Y, et al. Diagnostic splenectomy in patients with fever of unknown origin and splenomegaly. Acta Haematol. 2008;119(2):83–8.
9. Zhang L, Zhang W, Cai H, Cao X, Chen M, Li J, et al. Patients with Fever of Unknown Origin and Splenomegaly: Diagnostic Value of Splenectomy and Preoperative Risk Factors Suggestive of Underlying Lymphomas. Acta Haematol. 2017;137(4):240–6.
10. Singer DB. Postsplenectomy sepsis. Perspect Pediatr Pathol. 1973;1:285–311.
11. Rajwanshi A, Gupta D, Kapoor S, Kochhar R, Gupta† S, Varma† S, et al. Fine needle aspiration biopsy of the spleen in pyrexia of unknown origin. Cytopathology. 1999 May 1;10(3):195–200.