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**Case Report**

# Serous effusions in Burkitt lymphomas: a report of two paediatric cases with review of literature

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## ABSTRACT

The serosal cavity can be involved in many diseases, both benign and malignant. Such involvement is secondary to occurrence of disease at other sites. Lymphoma, although not a very common cause for serous effusion, is one of the most important one. The reason for serosal involvement in lymphomas has not been fully elucidated. These two cases highlight the importance of pleural and peritoneal fluid examination in the diagnosis of lymphoma. There is correlation between fluid examination and subsequent cytological and histopathological studies, with immunohistochemical confirmation. A comprehensive understanding of the mechanism of serosal involvement in lymphomas may lead to early diagnosis, better patient management and also development of newer treatment modalities for the same.

**Keywords:** Burkitt lymphoma, Peritoneal effusion, Pleural effusion, Serosa

## INTRODUCTION

Numerous benign and malignant conditions, including a variety of lymphoproliferative lesions, can involve the serosa. Usually, the involvement of serosal sites occurs secondary to disease at other sites. Primary involvement of serosal sites, by itself, is rare.[1] Lymphomas can be a cause of serous effusions, involving most frequently the pleural cavity. However, pericardial and peritoneal involvement is not common. The rate of occurrence of pleural effusion in various types of lymphomas ranges from 20 to 30%.[2,3]

Serous effusions can be seen in a variety of diseases, apart from lymphomas. These include infectious pathologies like tuberculosis, systemic diseases involving the kidneys, and various malignancies. Only about 15% of the serous effusions can be attributed to lymphomas.[4]

Different mechanisms are attributed to occurrence of serous effusions in various types of lymphomas. These effusions, when occurring in the pleura, are more common on the left side and show propensity for occurrence in males.[5]

We present two cases of lymphomas, presenting with involvement of pleural and peritoneal cavities, respectively.

## CASE REPORT

A 5-year-old female child presented with complaints of diffuse abdominal pain and distension for 15 days. This was associated with intermittent fever and occasional episodes of sweating. She had visible weight loss since last two months. There was no history of change in bowel habits, vomiting, or discolouration of stools. She did not have cough or any respiratory symptoms. On

examination, an abdominal mass was palpated, which was present mainly in the right lumbar and right hypogastric region. There was no inguinal, axillary or cervical lymphadenopathy.

The patient was evaluated with necessary laboratory and radiological investigations. She was mildly anaemic and had marked thrombocytosis (10,00,000/uL). Computed tomography (CT) of the abdomen showed enhancing, gross circumferential thickening of ileal loops and part of ascending colon. There were multiple enlarged conglomerated retroperitoneal lymph nodes with non-enhancing areas within (likely necrosis). These were encasing the major vessels. Also, multiple discrete, scattered lymph nodal masses were seen in the mesentery and omentum. Mildly enhancing peritoneal thickening and soft tissue nodules involving peritoneum were present. Free fluid was noted in the peritoneal and right pleural cavity.

Peritoneal fluid was sent for cytological examination, and omental and mesenteric biopsies were taken.

Post centrifugation smears from peritoneal fluid were cellular and showed many atypical lymphoid cells, along with plasmacytoid cells having eccentric hyperchromatic nucleus and moderate to abundant amounts of cytoplasm (Figures 1 and 2). Section of cell block prepared from the fluid showed many atypical lymphoid cells, macrophages and apoptotic bodies in a fibrino-haemorrhagic background. The cytological features were reported as suspicious for a lymphoproliferative disorder or lymphoma.

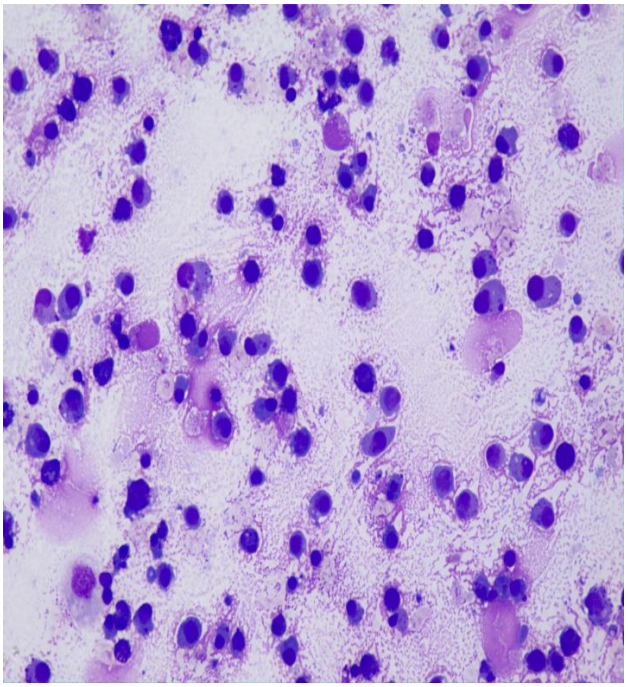

**Figure 1: Post centrifugation smear from peritoneal fluid showing atypical lymphoid cells, along with plasmacytoid cells (Giemsa, x400).**

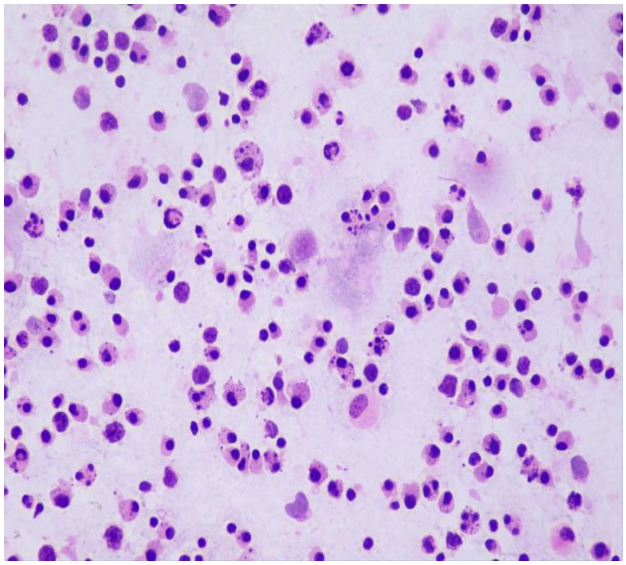

**Figure 2: Post centrifugation smear from peritoneal fluid atypical lymphoid cells, along with plasmacytoid cells (H and E, x400).**

Histopathological examination of omental and mesenteric biopsies revealed tumour that was diffusely infiltrating and composed of medium-sized, round, and uniform atypical lymphoid cells with round nuclei. There was a prominent starry-sky pattern due to the presence of multiple tingible body macrophages. Brisk mitotic activity was noted. Immunohistochemistry was performed and the tumour cells were diffusely immunopositive for CD-79a and negative for CD3 (Figure 3 A-D).

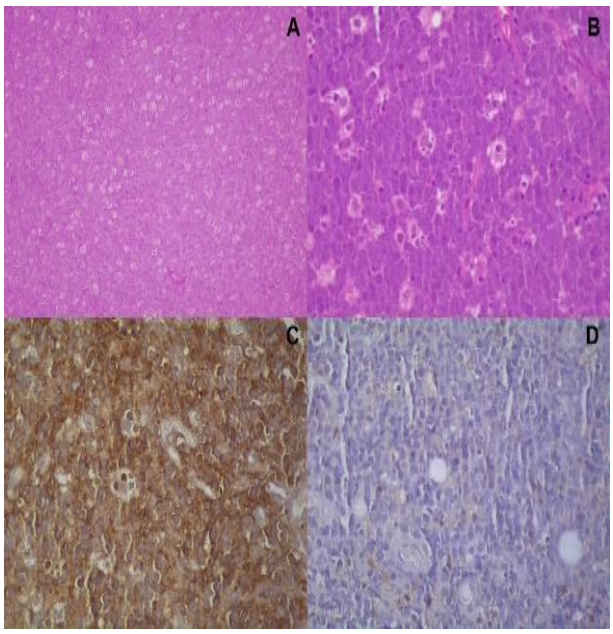


**Figure 3: Biopsy showing prominent starry-sky pattern due to presence of multiple tingible body macrophages (Ax100, Bx400); exhibiting immunopositivity for CD-79a; and immune-negativity for CD3.**

A 13-year-old male patient presented with difficulty in breathing for 10 days, which was associated with high-grade fever and loss of appetite. He also had intermittent non-productive cough. On examination, the patient was poorly built and poorly nourished. He had right sided cervical lymphadenopathy, with largest palpable node measuring 3 cm in largest dimension.

Radiological evaluation of the chest showed presence of a mass lesion on the right side, involving the pleura. Multiple discrete scattered lymph nodes were noted, along with pleural thickening and free fluid in the pleural cavity. Pleural fluid aspiration and direct fine needle aspiration (FNA), from right cervical lymph node, were performed.

Post centrifugation smears from pleural fluid were cellular and showed a few singly dispersed, medium to large cells with moderate amounts of basophilic cytoplasm showing cytoplasmic vacuoles, round to indented nucleus, with many nuclei showing nuclear convolutions and variably prominent nucleoli. These atypical lymphoid cells were also present in the cell block (Figure 4 A-D). Biochemical examination of the aspirated fluid revealed mildly increased protein level, normal sugar level, and there was no growth after 48 hours of aerobic incubation.

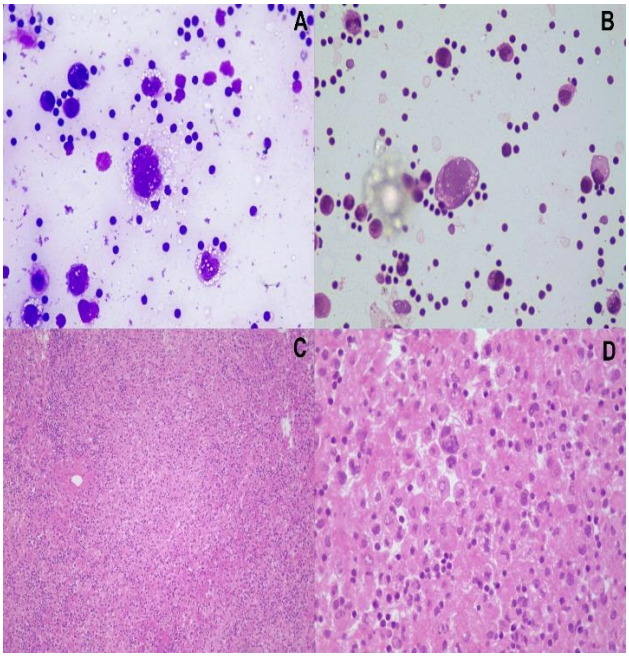


**Figure 4: Post centrifugation smears from pleural fluid showing atypical lymphoid cells (A-Giemsax400, B-H and Ex400); and cell block preparation exhibiting similar features (Cx100, Dx400).**

Direct FNA performed from right sided cervical lymph node was suggestive of high-grade lymphoma, with a possibility of Burkitt lymphoma in view of the atypical lymphoid cells exhibiting cytoplasmic vacuolations (Figure 5).

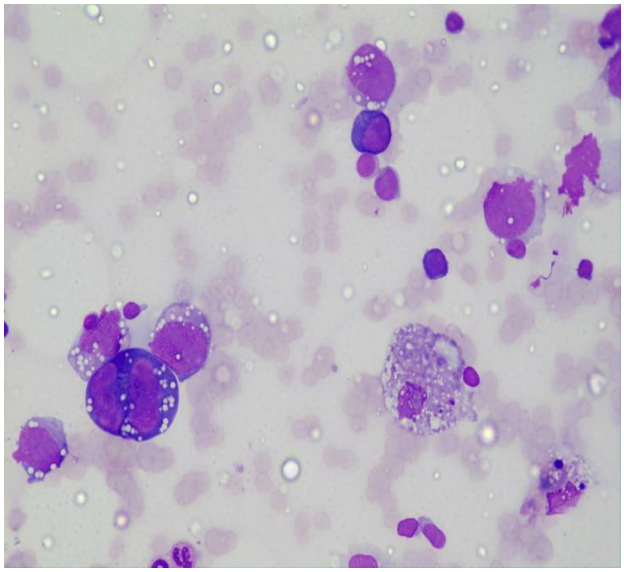

**Figure 5: Direct FNA from right sided cervical lymph node showing atypical lymphoid cells exhibiting cytoplasmic vacuolations (Giemsa, x400).**

## DISCUSSION

Serous effusions, irrespective of site of occurrence, occur more frequently in lymphomas originating from T-cells than B-cell originating lesions. About 25% of the T-cell lymphomas develop pleural effusions.[4] Also, the most common subtype of lymphomas to present with effusions is diffuse large B-cell lymphoma (DLBCL), followed by follicular lymphoma.[5]

In majority of the cases, serous effusions are present at the time of initial diagnosis and occur as a part of disease involving other sites. These effusions can be serous or serosanguinous, and are usually exudative in nature. Chylous effusions can also be seen.[6]

If any obstructive mass is not present, the occurrence of serous effusions in lymphomas can be attributed to vascular leakage which is caused by release or activation of vascular endothelial growth factor (VEGF).[7] The mechanism for the same, in non-Hodgkin lymphoma (NHL) is direct pleural infiltration, whereas, thoracic duct obstruction and impaired lymphatic drainage occur in Hodgkin disease (HD).[4] Chylous effusion is a rare entity which results from accumulation of lymphatic fluid in the serous cavity. The triglyceride level is utilized to make a diagnosis, with value more than 200 mg/dl favouring a diagnosis of chylous effusion. Chylous effusion may occur due to direct leakage due to trauma, exudation of lymph through walls of dilated lymphatic channels, or obstruction of lymph flow due to pressure by an external mass.[8]

The differential diagnoses of serous effusions in lymphomas includes presence of reactive lymphocytes, occurrence of any other small round cell tumour and presence of cells mimicking reed-Sternberg cells. The

lymphocytic infiltrates need to be differentiated on the basis of clinical features, cyto morphology and immunological typing. Diagnosis can be made with sensitivity and specificity of 100%, if immunophenotyping of effusion by flow cytometry is done along with morphological and cytochemical examination.[7]

Paracentesis is recommended as the first line tool for evaluation of any serous effusion. It is a safe and cost-effective procedure which plays both diagnostic and therapeutic roles.[3] However, the sensitivity of cytological examination alone is only about 60 to 70% and other diagnostic modalities should be used to establish or confirm the diagnosis. Florid mesothelial hyperplasia may occur in lymphomas, leading to non-specificity in the cytological report.

The fluid is rarely bloody in cases due to lymphoma, and is usually yellowish and transparent. SAAG, which is serum albumin minus effusion albumin, is usually less than 1.1 g/dl in ascites due to peritoneal malignancies.[9] Lactate dehydrogenase (LDH) level in the effusion can be used, both for diagnosis and prognosis, as it has high sensitivity and low specificity for malignant ascites.[10]

There is a high concordance of cyto morphology of these effusions with fine-needle aspiration cytology (FNAC) of the mass lesion, with rates more than 90%.[11]

## CONCLUSION

There is increasing awareness regarding the involvement of serous cavities in various types of lymphomas. This may be helpful in early diagnosis of cases, leading to initiation of prompt treatment and overall better prognosis. Lymphomas should be considered in the differential diagnoses of serous effusions, and must be used as a stepping stone for further evaluation of the cases.

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