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Coexistent Kaposi sarcoma and post-transplant lymphoproliferative disorder in the same lymph nodes after pediatric liver transplantation: A case report

Shu-Hong Zhang, Guang-Yong Chen, Zhi-Jun Zhu, Lin Wei, Ying Liu, Jing-Yi Liu

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Abstract

BACKGROUND

Kaposi sarcoma and post-transplant lymphoproliferative disorder have been occasionally reported in post-liver transplant patients. However, the simultaneous occurrence of these two diseases in the same lymph nodes is very rare.

CASE SUMMARY

We report the case of a 19-mo-old boy, who presented with intermittent fever and enlarged cervical lymph nodes after liver transplantation. Six cervical lymph nodes were biopsied, and the histopathological examinations revealed multifocal hyperplasia of spindle cells around small blood vessels, extravasated erythrocytes, and heavy infiltration of plasma cells in the cortex and medulla of the lymph nodes. The immunohistochemical analyses of spindle cells revealed positive expression of CD34, CD31, erythroblast transformation-specific-related gene, friend leukemia integration 1, and human herpesvirus-8. The lymphoproliferative lesions expressed CD38, CD138, and multiple myeloma 1. Epstein-Barr encoded RNA *in situ* hybridization demonstrated Epstein-Barr virus-positive lymphoid cells. Finally, we diagnosed the coexistence of Kaposi sarcoma and post-transplant lymphoproliferative disorder (plasmacytic hyperplasia) in the same lymph nodes. Treatment strategy included anti-CD20 monoclonal antibody (rituximab) and

discontinuation of the immunosuppressant therapies. Lymph node biopsies during follow-up examinations revealed lymphoid hyperplasia.

CONCLUSION

The rare coexistence of Kaposi sarcoma and post-transplant lymphoproliferative disorder in the same lymph nodes post-liver transplantation possibly associates with immunodeficiency and Epstein-Barr virus and human herpesvirus-8 coinfection.

Key Words: Kaposi sarcoma; Post-transplant lymphoproliferative disorder; Liver transplantation; Epstein-Barr virus infections; Human herpesvirus-8; Case report

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Core Tip: We report a rare case of coexistent Kaposi sarcoma and post-transplant lymphoproliferative disorder in the same lymph nodes after a pediatric liver transplant. The definitive diagnosis was based on histopathological examination of the lymph nodes. The patient recovered after discontinuation of the immunosuppressants and application of anti-CD20 monoclonal antibody (rituximab) therapy. In conclusion, the concurrent occurrence of these two disorders may be associated with immunodeficiency as well as Epstein-Barr virus and human herpesvirus-8 coinfection.

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INTRODUCTION

Post-liver transplantation patients suffer from an increased risk of developing various lesions, including Kaposi sarcoma (KS)[1,2] and post-transplant lymphoproliferative disorder (PTLD)[3-6]. KS mainly involves the skin and occasionally the lymph nodes and stomach[1,2], while PTLT can occur in multiple organs, including the lymph nodes and the gastrointestinal tract[5] after liver transplantation. However, these two different disorders rarely coexist in the same lymph node. In fact, to date, there have been only 7 case reports[7-12], particularly of acquired immunodeficiency syndrome (AIDS), non-AIDS, and kidney transplantation patients, describing the co-occurrence of KS and lymphoid tissue lesions in the same lymph node. These cases are summarized in Table 1. To the best of our knowledge, there is no case report regarding the concurrent occurrence of KS and PTLT in the same lymph nodes of a pediatric liver transplant patient.

In our hospital, the overall incidence of PTLT and KS in post-pediatric liver transplant recipients was 5.4% (43/789) and 0.13% (1/789), respectively, from 2013 to 2021. Only 1 of these cases had KS complicated with PTLT in the same lymph nodes. Herein, we report this unusual case of KS and PTLT coexistence in the same lymph nodes of a 19-mo-old boy after liver transplantation.

CASE PRESENTATION

Chief complaints

A 19-mo-old Asian boy with intermittent fever for 2 mo was admitted to the hospital.

History of present illness

The patient presented with facial edema and diarrhea after 6 mo of undergoing liver transplant surgery.

History of past illness

The patient had a history of past illness, including congenital biliary atresia, cholestatic cirrhosis, and hepatic encephalopathy. He was diagnosed with congenital biliary atresia at the age of 1.5 mo. A Kasai procedure had been performed on the patient at the age of 2 mo to treat his congenital biliary atresia. However, the procedure failed to reach the desired outcome, and he ultimately received a living donor liver transplant at the age of 13 mo. After liver transplantation, the young boy was treated with

Table 1 Previously reported cases of concurrent Kaposi sarcoma and lymphoid tissue lesions in the same lymph node

Case	Ref.	Age (yr)	Sex	Past illness	Type of lymphoid tissue lesion	Treatment	Outcome
1	Licci <i>et al</i> [7]	59	Male	AIDS	DLBCL	ND	ND
2	Licci <i>et al</i> [7]	58	Male	None	TCRLBCL	ND	ND
3	Fernandes <i>et al</i> [8]	18	Female	None	TCRLBCL	ND	ND
4	Paksoy[9]	30	Male	AIDS	DLBCL	ND	ND
5	Kankaya <i>et al</i> [10]	57	Male	None	NLPHL	Without any therapy	Alive (8 yr)
6	Ngan and Kuo[11]	61	Female	None	CHL	Chemotherapy	Alive (13 mo)
7	Sabeel <i>et al</i> [12]	30	Female	ESRD PTLD	B-NHL Subtype: ND	Discontinued cyclosporine; intravenous acyclovir	Deceased (5 wk)

ND: Not available; TCRLBCL: T cell rich large B cell lymphoma; NLPHL: Nodular lymphocyte predominant Hodgkin's lymphoma; CHL: Classical Hodgkin's lymphoma; B-NHL: B-cell non-Hodgkin's lymphoma; ESRD: End-stage renal disease; PTLD: Post-transplant lymphoproliferative disorder; AIDS: Acquired immunodeficiency syndrome; DLBCL: Diffuse large B-cell lymphoma.

intravenous methylprednisolone as induction therapy, followed by an immunosuppressive regimen of tacrolimus (FK506) + methylprednisolone. The methylprednisolone dose was gradually reduced and ultimately discontinued 3 mo after the liver transplantation.

Personal and family history

His father had provided the donor liver. Additionally, while his mother had a history of virus infection (hepatitis B), the virus was not transmitted to the child.

Physical examination

Physical examination upon admission showed several swollen lymph nodes that were palpable in both the anterior and posterior regions of the patient's neck.

Laboratory examinations

Laboratory investigations showed: hemoglobin, 64 g/L; white blood cells, 6.4×10^9 ; platelets, 6.4×10^9 ; alanine transaminase, 42 U/L; aspartate aminotransferase, 62 U/L; alkaline phosphatase, 314 U/L; and gamma-glutamyl transferase, 47 U/L. The patient was serum Epstein-Barr virus (EBV)-DNA positive, which was negative before liver transplantation.

Clinicians suspected PTLD of lymph nodes. To decide the next therapeutic plan, six of the patient's left cervical lymph nodes were excised. The histopathological examinations revealed multifocal hyperplasia of spindle cells around small blood vessels, particularly beneath the capsules of the lymph nodes (Figure 1A). Additionally, extravasated erythrocytes and a relatively scanty amount of inflammatory infiltrate were present. The spindle cells had minimal atypia (Figure 1B), and mitotic figures were absent. The cortical and medullary areas were infiltrated by numerous plasma cells along with small lymphocytes and eosinophils (Figure 1B).

Immunohistochemical staining of the lymph node tissues revealed notable phenotypic features of the spindle cells, such as a diffuse expression of CD34 (Figure 2A), CD31, erythroblast transformation-specific-related gene, and friend leukemia integration 1 transcription factor, as well as a partially-positive nuclear staining for human herpesvirus-8 (HHV8; Figure 2B). In the lymphoproliferative lesions, CD38, CD138 (Figure 2C), and multiple myeloma 1 (Figure 2D) were highly expressed, while CD20 was partially expressed. Moreover, the kappa/lambda ratio was approximately 1:1. Incidentally, the spindle cells and the lymphoproliferative lesions exhibited proliferation rates of approximately 25% and 10%, respectively. The Epstein-Barr encoded RNA *in situ* hybridization revealed EBV-positive lymphoid cells (Figure 3). The B-cell clonality was evaluated using PCR amplification, and the results revealed *IgH*, *IgK*, and *IgL* polyclonal gene rearrangements.

Imaging examinations

Ultrasonography and computed tomography scanning revealed that multiple cervical lymph nodes were enlarged on both sides.

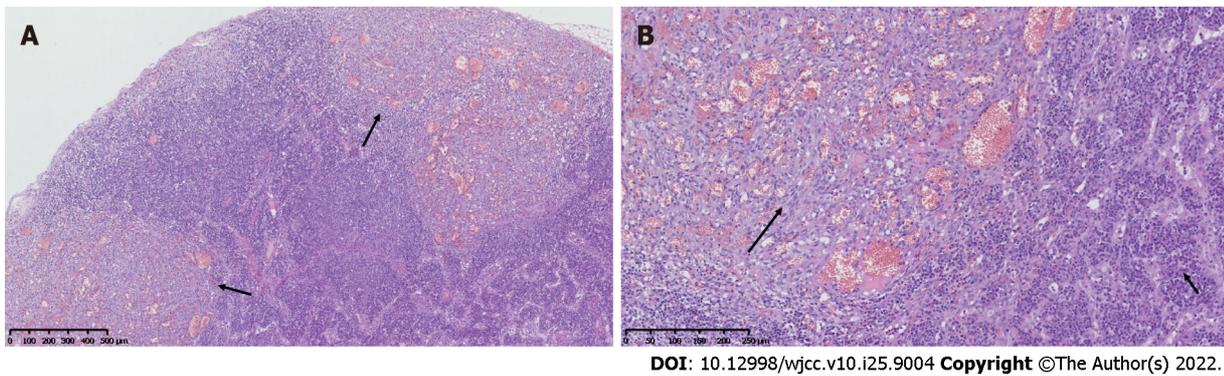


Figure 1 Histopathological changes observed in this case. A: Lymph nodes exhibited multifocal hyperplasia of spindle cells around small blood vessels, mainly beneath the capsule (arrows), and many lymphoid cells in the cortical and medullary regions of the lymph node (hematoxylin and eosin staining, $\times 40$); B: Spindle cells exhibited minimal atypia (large arrow, upper left); heavy infiltration of plasma cells (small arrow, lower right) (hematoxylin and eosin staining, $\times 100$).

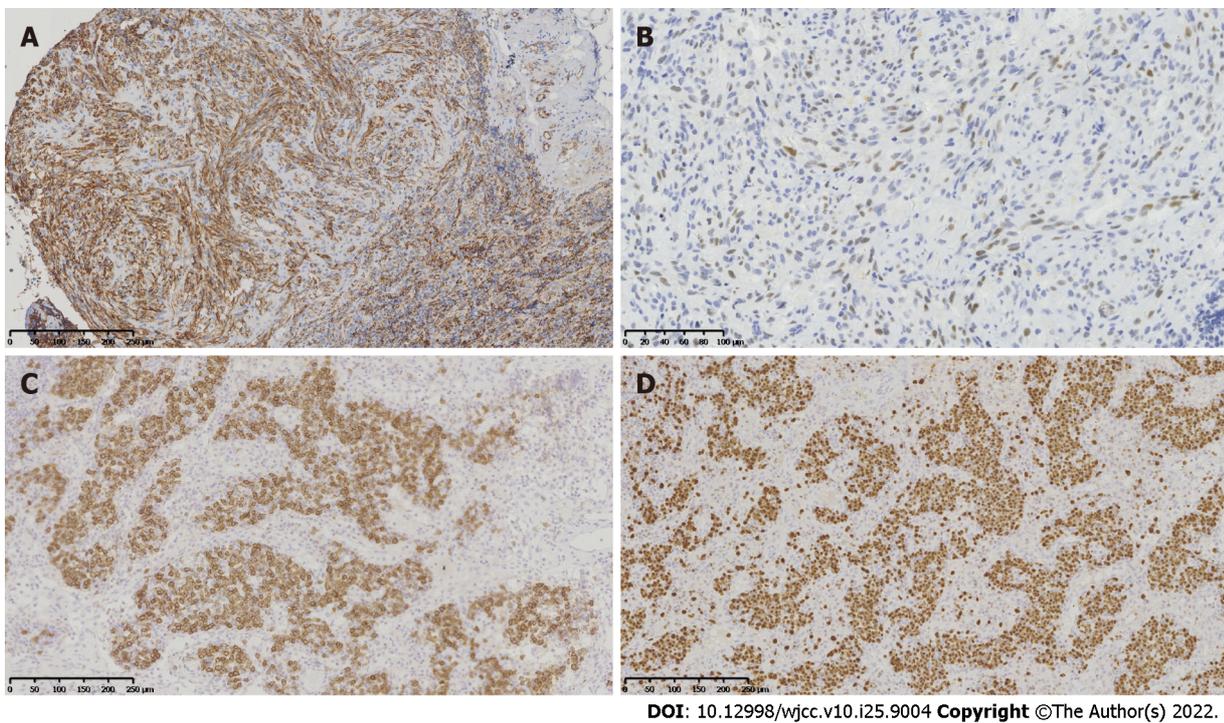


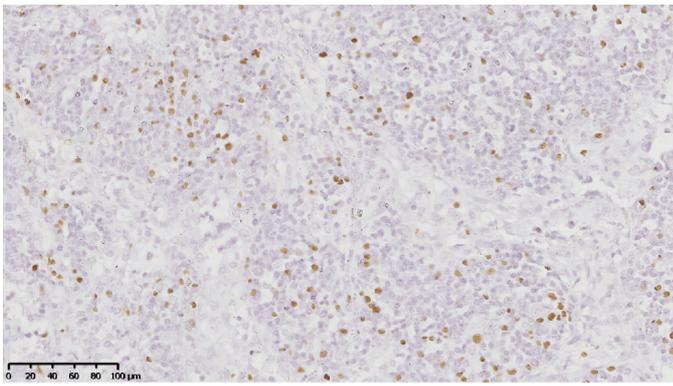
Figure 2 Immunohistochemical features. A: Positive membrane staining for CD34 of spindle tumor cells ($\times 100$); B: Positive nuclear staining for human herpesvirus-8 in parts of spindle tumor cells ($\times 200$); C: Positive staining for CD138 of lymphoid cells ($\times 100$); D: Positive staining for multiple myeloma 1 of lymphoid cells ($\times 100$).

FINAL DIAGNOSIS

The results confirmed concurrent KS and non-destructive PTLD, particularly plasmacytic hyperplasia, within the same lymph nodes of the patient.

TREATMENT

The patient received anti-CD20 monoclonal antibody (rituximab) therapy (1 cycle), and his immunosuppression therapy was discontinued. After 1 cycle of rituximab treatment, the patient's EBV-DNA replication load reduced, but transaminase levels increased. Meanwhile, a liver needle biopsy indicated drug-induced liver injury; therefore, rituximab was not continued. Tacrolimus (FK506) was initiated again 14 d after its discontinuation. The patient's condition improved gradually.



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Figure 3 *In situ* hybridization. Epstein-Barr encoded RNA *in situ* hybridization revealed Epstein-Barr virus-positive lymphoid cells ($\times 200$).

OUTCOME AND FOLLOW-UP

The patient was discharged in a stable condition 33 d after admission. However, at the ages of 60 and 71 mo, fever as well as lymph node enlargement were detected as a result of a telephone follow-up call and subsequent hospitalization. Lymph node biopsies were repeated on both occasions to exclude the relapse of PTLD and/or KS. However, all pathological results indicated lymphoid hyperplasia, and the recurrence of KS and PTLD was discarded as a possibility. The patient (109-mo-old) has been followed up for 8 years; he is in good health and attends school normally. The timeline of the patient diagnosis, treatment, and follow-up are summarized in [Table 2](#).

DISCUSSION

Long-term use of immunosuppressive agents increases the risk of different diseases, including KS[1,2] and PTLD[3-6], in post-liver transplantation patients. However, to date, there are only 7 case reports[7-12], describing the co-occurrence of KS and lymphoid tissue lesions in the same lymph node ([Table 1](#)). The ages of these patients ranged from 18-years-old to 61-years-old, including 4 cases in non-AIDS patients, 2 cases in AIDS patients, and 1 case after kidney transplantation. There were 5 cases of coexistent with non-Hodgkin's lymphoma and 2 cases coexistent with Hodgkin's lymphoma. Our case is different from reported cases mentioned above. Our case describes a liver transplant patient, occurring in a child, with a type of lymphoid tissue lesion associated with PTLD, plasmacytic hyperplasia.

In 1872, Moritz Kaposi[13] first described KS as a type of localized and invasive endothelial cell tumor. Based on clinical and epidemiological characteristics, KS can be divided into four types[14], namely classic, endemic, iatrogenic, and AIDS-related KS. Among them, iatrogenic KS is mainly observed in patients undergoing immunosuppressive therapy after a solid organ transplantation as well as in patients treated with immunosuppressants, notably corticosteroids, for various diseases. In fact, the incidence of iatrogenic KS is 500 times greater among organ transplant recipients as compared to that in the general population[15]. Specifically, KS has been reported in 2.00%-2.16% of adult liver transplant recipients[1,2,16], whereas its occurrence in pediatric liver transplant recipients is rare, with only a few case reports of individual recipients[17-19]. The incidence of KS in our series of liver graft recipients was 0.13% (1/789) from 2013 to 2021 in our department.

Typically, KS lesions occur on the skin, but they can also appear in internal organs and lymph nodes. However, the sole occurrence of KS in the lymph nodes is relatively rare. The KS distribution in lymph nodes can be unifocal or multifocal with a pathomorphology similar to that of KS in the skin, which includes the presence of red blood cells within slit-like spaces formed by spindle cells. The immunophenotype of KS is characterized by the positive expression of vascular endothelial cell markers, including CD31, CD34, erythroblast transformation-specific-related gene, and other relevant antigens.

Incidentally, HHV8 has an etiological role in KS, and all KS cases show almost invariable nuclear expression of HHV8, regardless of their epidemiological subtypes[14]. Hence, HHV8-positivity helps to confirm the diagnosis of KS as well as differentiate it from other vascular lesions. In our case, the immunohistochemical analyses of the spindle cells demonstrated the positive expression of CD34, CD31, erythroblast transformation-specific-related gene, and HHV8, and combined with the pathomorphological features, the diagnosis of KS was confirmed.

Table 2 Timeline of patient diagnosis, treatment, and follow-up

Age (mo)	Patient condition
1.5	Diagnosis: Congenital biliary atresia
2	Treatment: Kasai operation
13	Diagnosis: Cholestatic cirrhosis and hepatic encephalopathy. Treatment: Living donor liver transplantation
19	Diagnosis: Concurrent KS and non-destructive PTLD within the same lymph nodes. Treatment: Anti-CD20 monoclonal antibody (rituximab) therapy (1 cycle), discontinuation of immunosuppression
60	Diagnosis: Lymphoid hyperplasia. Treatment: Lymph node excised
71	Diagnosis: Lymphoid hyperplasia. Treatment: Lymph node excised
109	Follow-up: Alive

KS: Kaposi sarcoma; PTLD: Post-transplant lymphoproliferative disorder.

Incidentally, recipients of solid organ transplant or stem cell allograft can develop PTLD, which includes lymphoid or plasmacytic proliferations, as a consequence of immunosuppression[20]. Approximately 2.3%-4.3% of adult liver transplant recipients develop PTLD[3-5], whereas its incidence is as high as 9.7% in pediatric liver transplant recipients[5]. In our hospital, the overall incidence of PTLD in post-pediatric liver transplant recipients was 5.4% (43/789) from 2013 to 2021. It was slightly higher than that reported in adult liver transplantation patients. The majority of PTLD cases are “early onset PTLD,” *i.e.*, occurring within 12 mo of liver transplantation[21]. On the contrary, “late PTLD” includes those cases that occur > 12 mo after liver transplantation. In our patient, PTLD occurred 6 mo after the liver transplant surgery, thereby making it a case of “early onset PTLD.”

PTLD is most commonly associated with EBV infection since it plays an important etiological role in PTLD. Previous studies have reported that close monitoring of the viral load of EBV in liver transplant patients is helpful to assess the risk of developing PTLD[6]. According to the “World Health Organization Classification of Tumours of Haematopoietic and Lymphoid Tissues” in 2017[20], PTLD can be divided into non-destructive, polymorphic, monomorphic, and classic Hodgkin’s lymphoma-like PTLD. Our case was diagnosed as the non-destructive childhood PTLD, particularly plasmacytic hyperplasia, which tends to occur more in young individuals than the other types of PTLD.

Currently, there is no effective antiviral drug or standard treatment protocol for KS, and its treatment strategy depends on the disease staging, form of disease progression, distribution, clinical type, and patient’s immune status[19]. In cases of post-transplantation KS, if the lesion is confined to one site, then the most appropriate treatment plan is to reduce or discontinue the use of immunosuppressants, followed by a “wait and watch” period. Non-destructive PTLD often regresses spontaneously with discontinuation or reduction of immunosuppression; otherwise, it can be successfully treated by surgical excision. Our patient was followed up for the treatment of KS and was using anti-CD20 monoclonal antibody (rituximab) therapy and discontinuation of immunosuppression for the treatment of PTLD.

The association between two diseases in the same lymph nodes remains undetermined. Whether the two lesions within the same lymph nodes have a common pathogenic mechanism or if they emerged coincidentally is still unclear. We believe that the patient’s immunosuppressed status after liver transplantation and the coincident EBV and HHV8 infections were the causes of the simultaneous occurrence of PTLD and KS in the same lymph nodes.

CONCLUSION

This case portrays a rare coexistence of KS and PTLD in the same lymph nodes of a pediatric post-liver transplant patient. The definitive diagnosis required histopathological analyses. In conclusion, the patient’s immunodeficient status combined with EBV and HHV8 coinfection may be associated with the concurrent occurrence of these two diseases in the same lymph nodes.

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