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ABOUT COVER

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Treatment for CD57-negative $\gamma\delta$ T-cell large granular lymphocytic leukemia with pure red cell aplasia: A case report

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Author contributions: Xiao PP collected the patient's clinical data and drafted and revised the manuscript; Chen XY, Dong ZG, and Huang JM managed the patient in the hospital; Wang QQ, Chen YQ, and Zhang Y collected the laboratory data; all authors approved the final manuscript.

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Abstract

BACKGROUND

T-cell large granular lymphocytic leukemia (T-LGLL) is a rare type of aplastic anemia with diverse clinical manifestations. Concomitant diseases are often present at the first manifestation. We describe the treatment of a patient with CD57-negative $\gamma\delta$ T-LGLL with pure red cell aplasia (PRCA).

CASE SUMMARY

A 34-year-old woman with a 20-year history of anemia visited our hospital owing to severe dizziness and was admitted. Her condition was diagnosed as CD57-negative $\gamma\delta$ T-LGLL with PRCA through bone marrow cytology, bone marrow pathology, bone marrow flow cytometry, bone marrow multiplex polymerase chain reaction combined with fluorescent fragment analysis, and other tests. Treatment with prednisone, methotrexate, and subcutaneous erythropoietin did not significantly change her hemoglobin level. After treatment with oral cyclophosphamide for 3 mo, her hemoglobin level increased to approximately 100 g/L. After 5 mo of treatment, the patient could perform activities of daily living independently.

CONCLUSION

The treatment of CD57-negative $\gamma\delta$ T-LGLL with PRCA with cyclophosphamide helps to improve prognosis.

Key Words: Large granular lymphocytic leukemia; Pure red cell aplasia; Aplastic anemia; $\gamma\delta$ T-cell; Cyclophosphamide; Case report

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Core Tip: This case report presents a rare case of $\gamma\delta$ T-cell large granular lymphocytic leukemia with pure red cell aplasia. A 34-year-old woman was admitted to our hospital with a 20-year history of anemia. Upon investigation, it was discovered that our patient had an atypical immunophenotype. Cyclophosphamide was added to her treatment, and other drugs, such as prednisone, methotrexate, and erythropoietin, were discontinued. Her hemoglobin level increased to 100 g/L within 3 mo. We believe that our study makes a significant contribution to the literature because we report on a favorable prognosis in our patient.

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INTRODUCTION

T-cell large granular lymphocytic leukemia (T-LGLL) is classified into two types, $\alpha\beta$ -LGLL and $\gamma\delta$ -LGLL, based on the T-cell receptors (TCR). Of these, $\alpha\beta$ -LGLL is common and $\gamma\delta$ -LGLL is rare, accounting for approximately 5% of all T-LGLL cases [1]. Approximately 8%–19% of T-LGLL patients have pure red cell aplasia (PRCA), 25% have thrombocytopenia, and 10%–18% have rheumatoid arthritis [2]. The typical immunophenotype of $\gamma\delta$ -LGLL is CD57-positive, which mostly results in indolent disease. However, we present a case of CD57-negative $\gamma\delta$ -LGLL involving progressive aggravation of anemia and describe the treatments used.

CASE PRESENTATION

Chief complaints

A 34-year-old woman with a 20-year history of anemia was admitted to our hospital. She experienced severe dizziness, which led her to visit the hospital.

History of present illness

The patient reported that a diagnosis of mild anemia was made 20 years ago; however, no treatment followed the diagnosis. Gradually, the anemia worsened. Two years ago, the patient visited a hospital for bone marrow cytology and received a diagnosis of PRCA. She was treated with oral cyclosporine A (CSA) 125 mg bid for 2 years. Her condition worsened, and approximately every 2 wk, she received an infusion of red blood cells after her diagnosis (until this admission).

History of past illness

The patient had no other remarkable medical history.

Personal and family history

The patient reported an unremarkable personal and family history.

Physical examination

The patient presented with the features of severe anemia, including pale skin and lips. There was no palpable swelling of superficial lymph nodes and no tenderness in the sternum. The body temperature was 36.7 °C; heart rate, 92 beats/min; blood pressure, 110/76 mmHg; respiratory rate, 20 breaths/min, and oxygen saturation, 98%. The lung sounds were normal, and the spleen and liver were not enlarged.

Laboratory examinations

Complete blood count showed the following results: White blood cell count, 2.56×10^9 /L; neutrophil level, 0.87×10^9 /L; red blood cell count, 1.16×10^{12} /L; hemoglobin (Hb) level, 35 g/L; platelet count, 241×10^9 /L; and reticulocyte count, 0.98%. Biochemical

test results were normal, and direct and indirect antiglobulin test results were negative. Further, the following findings were noted: Antinuclear antibody/extractable nuclear antigen antibody, negative; antineutrophil cytoplasmic antibody, negative; anticentromere antibody, negative; humoral immunity, normal; rheumatoid factor, normal; cyclic citrullinated peptide antibody, negative; human leukocyte antigen-B27, negative; erythrocyte sedimentation rate, 21.0 mm/h; ferritin, > 1500 ng/mL; Vit B12, > 1117.75 pmol/L; C-reactive protein, normal range; Coombs test, negative; flow cytometry for paroxysmal nocturnal hemoglobinuria, negative; thyroid function, normal, and anti-human T-cell lymphotropic virus antibody, negative.

Bone marrow cytology revealed PRCA as well as large granular lymphocytes (Figure 1). Bone marrow pathology revealed the presence of few erythroid cells and a relatively higher T lymphocyte count (Figure 2). A sinus rhythm was noted on performing electrocardiography. The result of chromosome karyotype examination was normal. Erythroid progenitor cell culture resulted in 60/105 bone marrow-derived mononuclear cells. Bone marrow flow cytometry revealed the presence of 13.22% of $\gamma\delta$ T lymphocytes with the immunophenotype CD3⁺⁺, CD8⁺, TCR $\gamma\delta$ ⁺, CD2^{part+}, CD5^{part+}, CD4⁻, CD57⁻, CD34⁻, CD33⁻, CD117⁻, CD56⁻, and TCR $\alpha\beta$ ⁻ (Figure 3). Bone marrow multiplex polymerase chain reaction combined with fluorescent fragment analysis found a D β -J β rearrangement in TCR β (299 bp) at 194 bp and a V δ -J δ rearrangement in TCR δ at 221 bp. The monoclonal rearrangement was detected in the V γ lf interval, V γ 10-J γ interval (238 bp), and V γ 9 and V γ 11-J γ intervals in TCR γ at 174 bp (Figure 4).

Imaging examinations

Chest computed tomography showed two notable findings. First, the patient had decreased heart density, which is in line with the diagnosis of anemia. Second, the density of the liver parenchyma was high, which might signify splenomegaly. Using hepatobiliary and spleen color Doppler ultrasound, splenomegaly was confirmed. The patient showed a normal sinus rhythm on echocardiography.

FINAL DIAGNOSIS

This patient's condition was diagnosed as CD57-negative $\gamma\delta$ T-LGLL combined with PRCA.

TREATMENT

Two units of red blood cells were transfused in our patient after admission. Then, the patient was administered oral methotrexate (MTX; 15 mg, weekly) and prednisone (50 mg, daily) and a subcutaneous injection of erythropoietin (3000 IU, three times weekly). After 1 mo, the Hb level was maintained at 40–50 g/L, and it did not increase. Therefore, the prednisone and MTX doses were gradually tapered until each drug was completely discontinued; erythropoietin was also discontinued. Cyclophosphamide (CTX) 200 mg bid was administered as the only treatment. The Hb level increased gradually up to approximately 100 g/L after 3 mo.

OUTCOME AND FOLLOW-UP

With the current diagnosis and 5 mo of treatment, the patient has been able to perform activities of daily living independently.

DISCUSSION

T-LGLL is a relatively rare disorder; only a few clinical trials have been conducted, and few experiences have been published regarding this disease. Therefore, simply relying on traditional diagnostic methods, such as examination of bone marrow morphology, may lead to misdiagnosis and missed diagnosis. Liu *et al*[3] retrospectively analyzed 10 cases of T-LGLL. Among them, two cases of T-LGLL involved PRCA. Zhu *et al*[4] analyzed 17 cases of $\gamma\delta$ T-LGLL, of which 11 cases

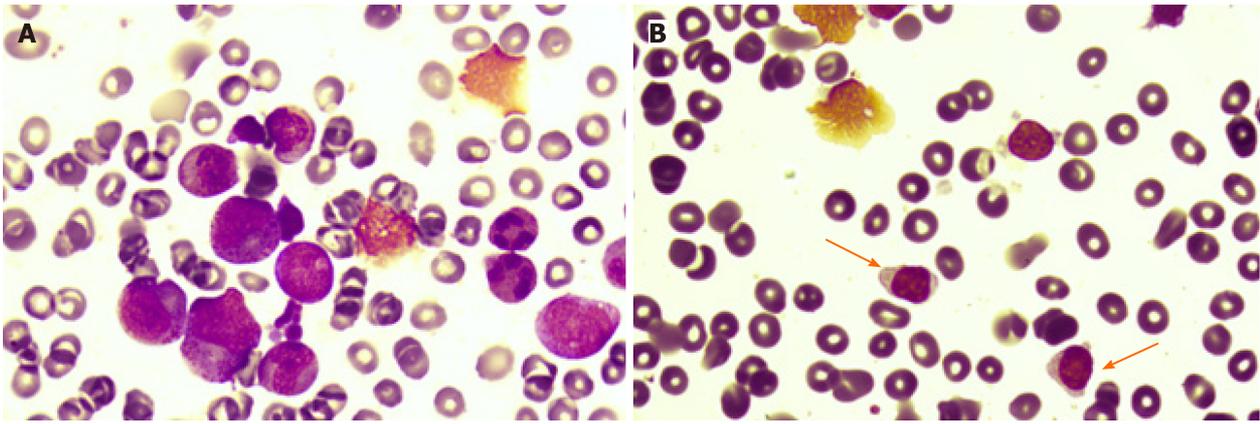


Figure 1 Bone marrow morphology. A: Myeloproliferative actively and absence of erythroid hyperplasia; B: Large granular lymphocyte (arrowheads). A and B: Wright staining, 1000 \times .

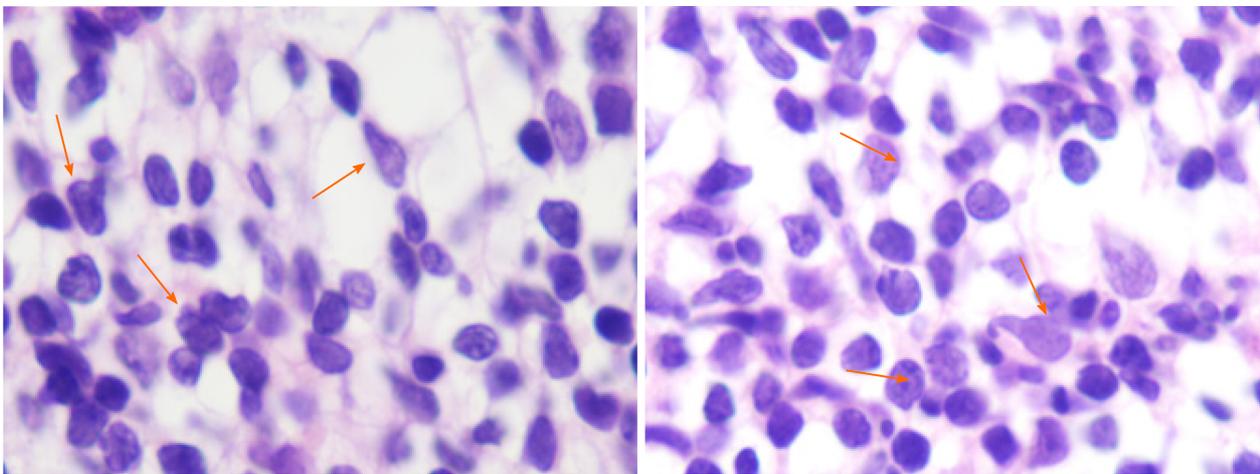


Figure 2 Histopathology of bone marrow tissue. Large granular lymphocyte (arrowheads) (hematoxylin and eosin staining, 1000 \times).

involved PRCA[4]. $\gamma\delta$ - and $\alpha\beta$ T-LGLL show very similar clinico-biological behaviors as well as antigen-driven T-cell lymphoproliferation[5]. The typical immunophenotype of $\gamma\delta$ T-LGLL is CD2+/CD3+/CD4-/CD7+/CD8-/CD16-/CD56-/CD57+/TCR $\alpha\beta$ -/TCR $\gamma\delta$ +

This report describes a case of $\gamma\delta$ T-LGLL with PRCA (immunophenotype CD57-), which is atypical for $\gamma\delta$ T-LGLL. Zhang *et al*[6] presented one case of CD57- $\gamma\delta$ T-LGLL with multiple organ failure. In their case, which involved a chromosomal abnormality, three courses of chemotherapy including CTX, fludarabine, and granulocyte-colony stimulating factor were administered for hemophagocytic syndrome, but the patient died of an infection. Sylvia *et al*[7] also presented one case of CD57- and CD56- $\gamma\delta$ T-LGLL, but no related treatments or prognoses were reported.

At present, the treatment for T-LGLL mainly focuses on immunosuppressive therapy, including MTX, CTX, and CSA, as well as purine drugs, combined chemotherapy, and splenectomy. Second- and third-line treatments such as targeted drugs are available[8,9], but there is no consensus as to whether immunosuppressive agents should be used as a single agent or in combination. Our patient had a medical history of anemia for more than 20 years. She showed no improvement in the Hb level following CSA treatment for PRCA. Frequent red blood cell transfusions caused iron overload, and her condition gradually worsened. After the diagnosis of T-LGLL was confirmed, prednisone combined with MTX and erythropoietin was administered. Combination of prednisone with first-line immunosuppressive agents (MTX, CSA, or CTX) may lead to rapid hematologic improvement[10]. MTX has anti-inflammatory and anti-proliferative effects, in part by inducing the apoptosis of activated T-cells[11]. However, there was no significant improvement in the Hb level of our patient. The Hb level increased to a value close to normal after 3 mo of treatment with CTX alone. The patient no longer required regular red blood cell transfusion, which further confirmed

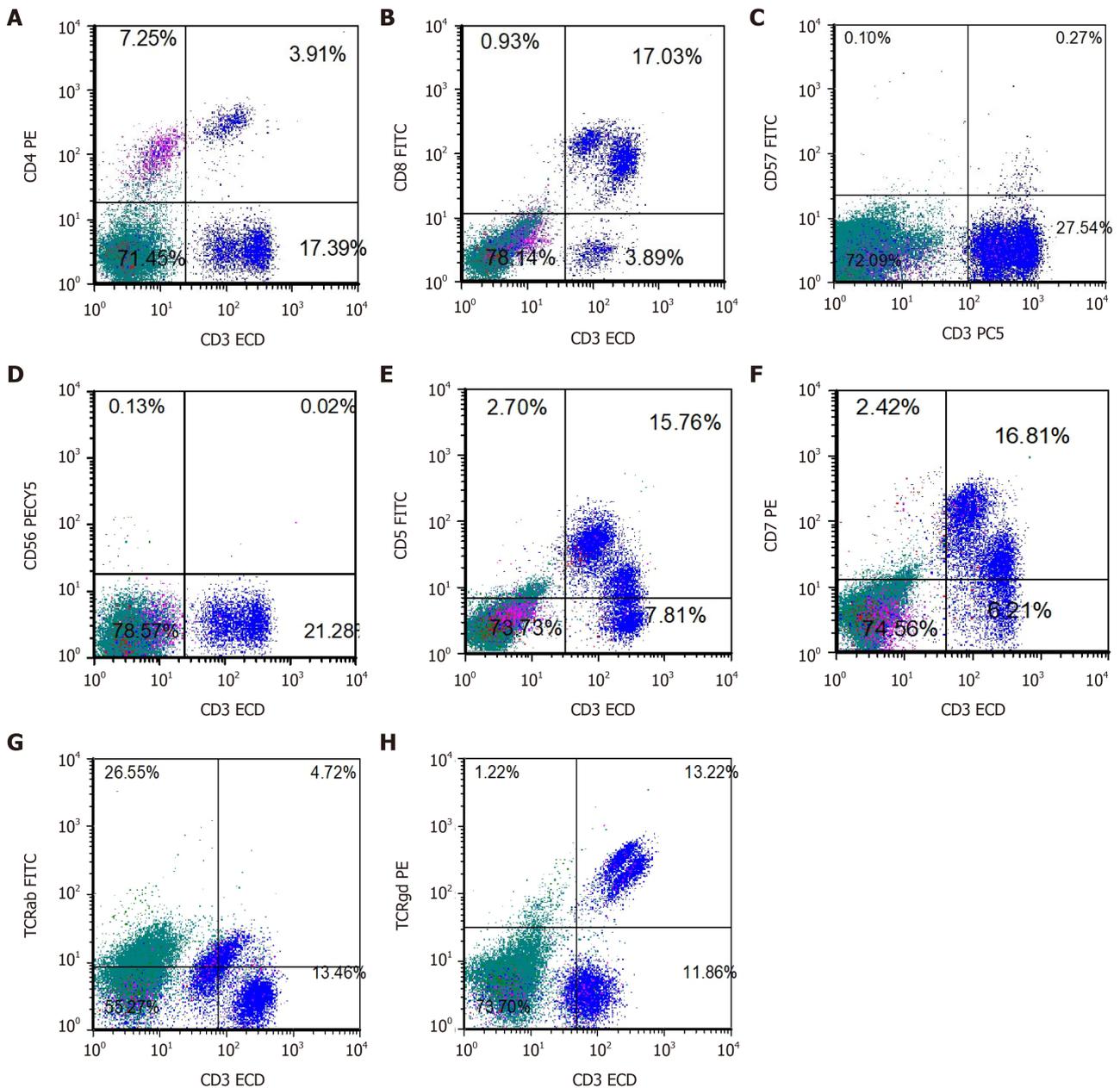


Figure 3 Immunophenotype of bone marrow by flow cytometry. A: CD4 PE; B: CD8 fluorescein isothiocyanate (FITC); C: CD57 FITC; D: CD56 PECY5; E: CD5 FITC; F: CD7 PE; G: T-cell receptors ab FITC; H: TCRgd PE. TCR: T-cell receptors.

the therapeutic potential of CTX for T-LGLL combined with PRCA. Sanikommu *et al* [12] presented the cases of 118 LGLL patients who received CSA, MTX, or CTX. CTX had the highest initial response rate and the shortest response duration. In atypical CD57-negative T-LGLL cases, the effect of CTX is particularly significant. The mechanism of CTX in T-LGLL combined with PRCA is still unknown. One possible mechanism is that CTX reduces the count of cytotoxic T lymphocytes (CTLs) that damage antibody-bound erythroblasts directly. Another possible mechanism is that CTX reduces the cytotoxic T lymphocytes-mediated damage to hematopoietic progenitors, which leads to anemia[13]. In our case, combined chemotherapy, splenectomy, and other treatments that pose a greater risk of side effects were avoided.

CONCLUSION

We describe our experience regarding the treatment of CD57-negative $\gamma\delta$ T-LGLL with PRCA and provide a reference for evaluating prognosis in this patient group. Our

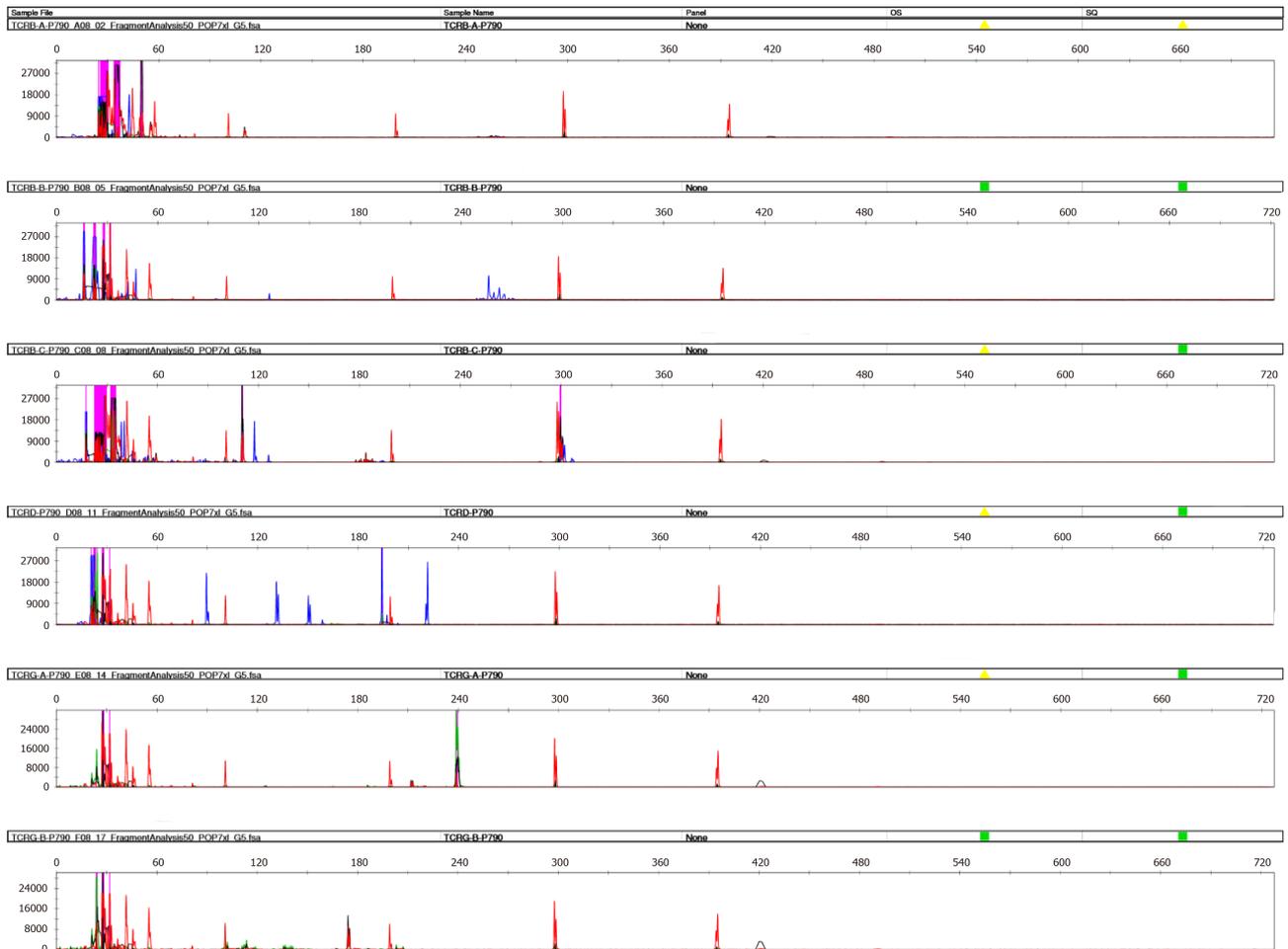


Figure 4 Monoclonal T-cell receptors rearrangement of bone marrow.

patient responded well to CTX and resumed routine activities. The mechanism of CTX in CD57-negative T-LGLL with PRCA still needs to be explored.

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