DOES PULMONARY LANGERHANS’ CELL HISTIOCYTOSIS HAVE A RECURRENCE PATTERN AFTER LUNG TRANSPLANTATION?

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Objectives: Pulmonary Langerhans’ cell histiocytosis (PLCH) is an uncommon indication for lung transplantation (LuTx); recurrence of the disease after LuTx is rare.

Case description: A PLCH and secondary PAH (sPAB: 90 mmHg) patient had undergone double LuTx with alemtuzumab induction and under assistance of central veno-arterial ECMO 25 months ago. She was discharged from the operating room to the ICU with a peripheral veno-arterial ECMO. She had a grade III PGD on the 5th day. ECMO was weaned on the 7th day. On the 8th day she had fever (39°C) with infiltrations on chest X-ray. Bronchoscopic biopsy revealed an acute fibrinous and organizing pneumonia. Pulse steroid therapy was initiated. She had polyuria from the first day of ICU follow-up; diabetes insipidus was diagnosed without any sign on preoperative evaluation and received demopressin management. She reached best FEV1 at 11 months (75%). At 12th month she resumed smoking and we recognized cavitory reticulonodular infiltrates in the bilateral upper lobes. After cessation of smoking lesions regressed. On the 24th month of follow-up, thoracic CT revealed reticulonodular pattern in the bilateral upper lobes again. Bronchoscopic biopsy revealed non-specific histological pattern without any rejection (A0Bx). Smoking history was detailed and we recognized that she again resumed smoking, half packs per day during 3 months. The radiological and pathological signs were diagnosed as recurrence of PLCH; steroid therapy (1 mg/kg) was initiated and complete radiological regression was observed after one month of treatment.

Conclusion: PLCH is an uncommon, granulomatous disease which destructs more than one tissue and system. Cigarette was the major etiological factor. Progressive parenchymal infiltrates could be observed without any clinical symptoms. Recurrence of primary disease after LuTx is uncommon; recurrence commonly could be observed in patients who had extra-pulmonary disease and in patients who resumed smoking. During ICU follow-up our patient was diagnosed with central diabetes insipidus; this revealed the extra-pulmonary disease of PLCH and she resumed smoking after LuTx. Treatment was cessation of smoking and steroid therapy. At 27 months she is without any symptom.

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