

The patient was a 34-yr-old man who presented with complaints of fever and a chronic cough. He was a smoker and had a history of pulmonary tuberculosis that had been treated and cured. A computed tomographic (CT) scan revealed multiple tiny nodules in both lungs. A thoracoscopic lung biopsy was taken from the right upper lobe. The microscopic examination revealed a typical LCH. The tumor cells had vesicular and grooved nuclei, and they formed small aggregations around the bronchioles (Fig.1). The tumor cells were strongly positive for S-100 protein, vimentin, CD68 and CD1a. There were infiltrations of lymphocytes and eosinophils around the tumor cells. With performing additional radiologic examinations, no other organs were thought to be involved. He quit smoking, but he received no other specific treatment. He was well for the following one year. After this, a follow-up CT scan was performed and it showed a 4 cm-sized mass in the left lower lobe, in addition to the multiple tiny nodules in both lungs (Fig.2). A needle biopsy specimen revealed the possibility of a sarcoma; therefore, a lobectomy was performed. Grossly, a 4 cm-sized poorly-circumscribed lobulated gray-white mass was found (Fig.3), and there were a few small satellite nodules around the main mass. Microscopically, the tumor cells were aggregated in large sheets and they showed an infiltrative growth. The cytologic features of some of the tumor cells were similar to those seen in a typical LCH. However, many tumor cells showed overtly malignant cytologic features such as pleomorphic/hyperchromatic nuclei and prominent nucleoli (Fig.4), and multinucleated tumor giant cells were also found. There were numerous mitotic figures ranging from 30 to 60 per 10 high power fields, and some of them were abnormal. A few foci of typical LCH remained around the main tumor mass. Immunohistochemically, the tumor cells were strongly positive for S-100 protein (Fig.5) and vimentin; they were also positive for CD68 (Dako N1577, Clone KPI), and focally positive for CD1a (Fig.6), and they were negative for cytokeratin, epithelial membrane antigen, CD3, CD20 and HMB45. The ultrastructural analysis failed to demonstrate any Birbeck granules in the cytoplasm of the tumor cells. Now, at five months after lobectomy, the patient is doing well with no significant change in the radiologic findings.