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CORRESPONDENCE

Systemic chemotherapy alone for Langerhans cell histiocytosis in the mandibular condyle with preservation of the alveolar bone

Langerhans cell histiocytosis (LCH) is a rare disease with unknown pathogenesis. According to the characteristics of the illness, LCH can be divided into three clinical forms: eosinophilic granuloma, Hand-Schüller-Christian disease, and Letterer-Siwe disease. In 1953, Lichtenstein found the common lesional histiocyte-like cell X in the histological section of LCH, and he collectively named these diseases "histiocytosis X" (X represents its unknown etiological factor).[1] Then, the lesional histiocyte-like cell X was found to be the Langerhans cell; therefore, the disease was renamed Langerhans cell histiocytosis.[2]

Here, we reported a case of stage I LCH in a 2-year-old girl who underwent chemotherapy in 2010. However, she was found to have recurrence of LCH in the left mandibular ascending ramus and condylar region in 2011. Head and neck computed tomography showed a radiolucent bone lesion near her left mandibular condylar process ~20.7 mm in diameter. A dental panoramic radiograph also revealed a solitary punched-out osteolytic bone lesion without a sclerotic border in the left mandibular ascending ramus and condylar region (Figures 1A and 1B). Aside from these findings, there were no specific abnormal manifestations within the oral cavity of the patient, including the teeth and gingival regions. A biopsy was performed, and the histopathological examination showed a sheet of histiocyte-like cells with coffee-bean nuclei admixed with numerous eosinophils and few multinucleated giant cells (Figure 2A). Immunostain revealed that the histiocyte-like cells were positive for CD1a (Figure 2B). The patient was subsequently treated with systemic chemotherapy that finally resulted in a successful lesional regression (Figures 1C and 1D). According to the most recent bone scan in 2015, the bone lesion was effectively controlled, and the bone was reconstructed very well. The patient still received regular follow-up examinations as an outpatient in the pediatric oncology department.

Current treatment modalities of LCH include surgical curettage, excision, radiation therapy, and chemotherapy. For focal bone lesions, surgical curettage or bone resections remain the treatment of choice.[2] For children with LCH involving the mandible, radiation cannot be used for therapy due to the potential damage to the permanent dental follicles. Chemotherapy in combination with steroids and cytotoxic agents could reduce LCH recurrence while maintaining drug efficacy.[2] Therefore, we administered systemic chemotherapy alone for our patient.

LCH is a rare disease with unknown pathogenesis. The major key point to diagnose this disease is the identification of Langerhans cells in the biopsy tissue section. To date, anti-CD1a immunostaining is the most commonly used method to identify Langerhans cells in LCH and several related oral or skin lesions.[3–5] Here, we described a LCH patient treated with systemic chemotherapy alone to achieve a complete regression of the LCH lesion. Based on long-term follow-up examinations, the LCH lesion of this patient has been effectively controlled, and bone regeneration has been observed. Dental practitioners should note that clinical symptoms/signs of LCH include the presentation of single or multiple bone lesions in the oral and maxillofacial regions, especially in the jawbone. The manifestations of LCH in the oral cavity might mimic the symptoms/signs of common dental problems, such as moderate or severe periodontitis. Dental practitioners play important roles in early diagnosis of pediatric patients with LCH.

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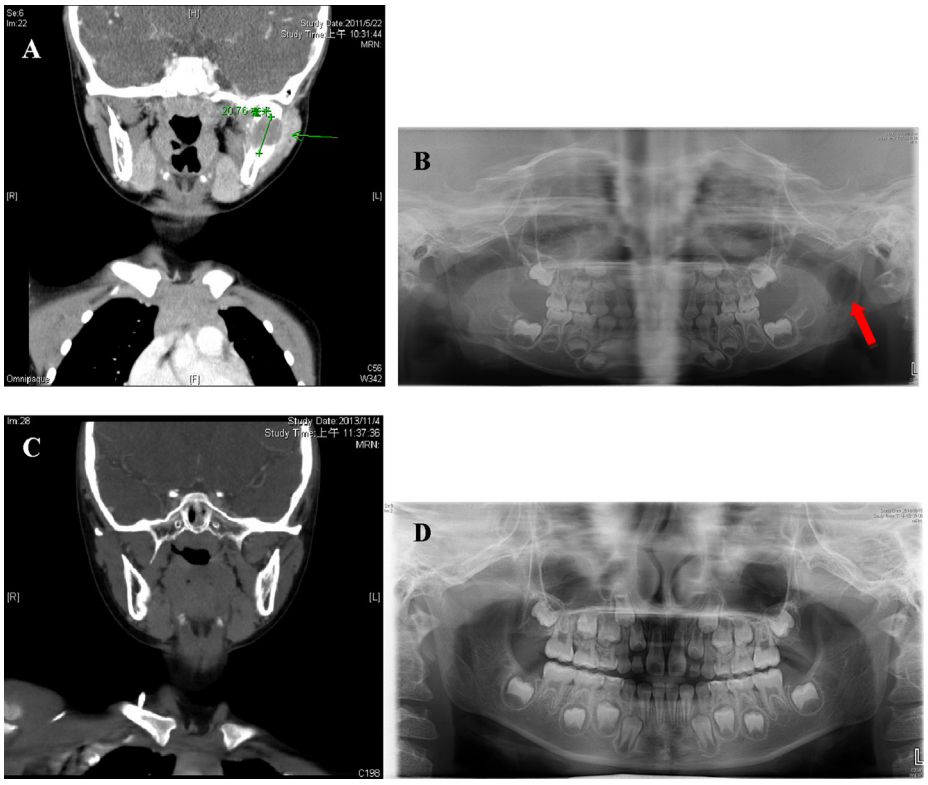

Figure 1 Head and neck computed tomography and dental panoramic radiography showing a radiolucent lesion in the left ascending ramus and condylar region of a pediatric patient (A and B) before and (C and D) after treatment.

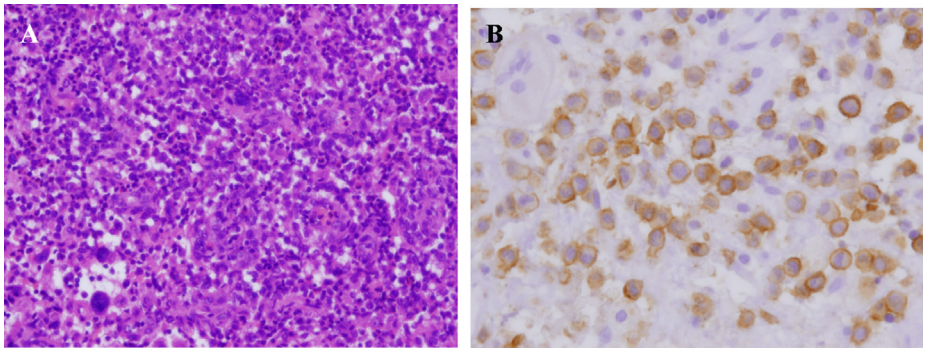

Figure 2 The microphotograph of a (A, original magnification: 200×) hematoxylin-and-eosin-stained tissue section showing a sheet of histiocyte-like cells with coffee-bean nuclei admixed with numerous eosinophils and few multinucleated giant cells and (B, original magnification: 200×) an immunostained tissue section showing that the histiocyte-like cells were positive for CD1a.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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