Original Article

An Extensive Case of Cherubism: a long term follow-up case

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Abstract

The present paper describes the radiological features, including CT, of a case of cherubism followed for a relatively long period. The case demonstrated the active phase of the lesion, which progressed from grade 1 to grade 3 during a 6 year period. The radiographic signs, such as increased radiolucency, inflammatory signs and root resorption as well as laboratory data seemed to be important to diagnose the stage of cherubism and to predict the clinical course of the disease.

Introduction

Cherubism is a hereditary abnormality which affects the jaw bones. It is characterized by a painless swelling which occurs bilaterally at the angle of the mandible. Sometimes the lesion is also present at the maxillary tuberosities. This disorder was first described by Jones in 1933, under the title of "Familial multilocular cystic disease of the jaws". Since then, a relatively large number of reports were published and the etiology, pathology and genetic aspects have been discussed.

However, no patient with early typical
cherubism has been carefully followed long term without some form of treatment\(^2\). The authors have encountered a typical case followed for a relatively long period under observation. We report herein the course of the lesion radiologically and discuss the activity of this lesion with the clinical information available.

**Case Report**

The patient, a 6-year old female was first referred to NAGASAKI UNIVERSITY DENTAL HOSPITAL with the chief complaint of painless swelling of the bilateral mandibular angles in January, 1982. She had had a painless swelling of the right mandibular angle since 3 years of age, swelling of the contralateral side followed at 4, since then, it has grown slowly and diffusely.

Physical examination revealed massive swelling extending bilaterally from the mandibular angles to the cheek. The submandibular lymph nodes were bilaterally enlarged. Teeth present in the mouth were 6EDC2112CDE6. Blood chemistry showed slightly elevated serum alkaline phosphatase (24.4 KAU). Hematological examination

![Radiograms at 6 years of age.](image)

Fig. 1 Radiograms at 6 years of age. 
(a) Panoramic view. (b) Waters' view. Multilocular cystic radiolucency is observed bilaterally at the mandibular angles. There is no evidence of maxillary lesion.