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Case Report

Oral findings and microbiological evaluation in a case of triple-X syndrome

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Abstract We present a case of a triple-X syndrome in a Japanese girl who was treated from 5Y9M to 14Y6M of age at the Pedodontic Clinic of Osaka University Dental Hospital. At the first examination, multiple dental caries were identified, and a pulpectomy and extraction of the affected teeth were carried out. There were no congenitally absent teeth, including third molars, though the development of permanent teeth was approximately 2 years behind chronological age. At 14Y6M, a PCR analysis of 10 putative periodontopathic bacteria was performed, and all 10 species were found in saliva samples. At the same time, an oral examination revealed that the gingival tissues were highly inflamed with an extremely large amount of calculus, suggesting a risk for the early onset of periodontitis.

Key words
PCR,
Periodontitis,
Periodontopathic bacteria,
Retarded eruption,
Triple-X syndrome

Introduction

Triple-X syndrome with premature ovarian failure was first described by Jacobs *et al.*[1] In 1991, a cytogenetic survey of 14,835 consecutive live births (7,608 males and 7,227 females) in Japan showed that 6.27 per 1,000 had a major chromosomal abnormality, and 0.98 per 1,000 females were diagnosed as having triple-X syndrome[2]. There were 12 newborns with triple-X syndrome born between 1964 and 1974 and then longitudinally monitored until 15 to 22 years of age for whom medical manifestations until adolescence were recorded in detail[3]. It is thought that problems with triple-X syndrome are more likely to have verbal processing deficits, lower IQ scores, and a global delay[4]. However, there have been few reports on oral manifestations associated with this syndrome; the only reported findings are that tooth enamel in subjects with this syndrome was thicker than that in normal subjects without a systemic disorder[5] and that bilateral maxillary second molars were congenitally missing in one case[6].

Periodontitis is rarely seen in children and adolescents except in association with systemic diseases such as neutropenia, Chediak-Higashi syndrome, Papillon-Lefevre syndrome, Down's syndrome, diabetes mellitus, hypophosphatasia, Histiocytosis syndrome, Ehlers-Danlos syndrome, and acquired immunodeficiency syndrome[7]. We previously reported the detection of 10 putative periodontopathic bacteria found in subgingival plaque samples from 144 systemically healthy children without periodontitis using a PCR method[8]. *Capnocytophaga ochracea*, *Capnocytophaga sputigena*, *Campylobacter rectus*, *Eikenella corrodens*, and *Actinobacillus actinomycetemcomitans* were found in approximately half of the samples from children of all ages. *Prevotella nigrescens* was also frequently detected in samples from older children, whereas *Tannerella forsythensis* and *Prevotella intermedia* were detected less frequently, and *Porphyromonas gingivalis* and *Treponema denticola* were not found in any of the samples. In a series of studies on Down's syndrome patients, the distribution of periodontal bacteria was evaluated using the same methods, and

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it was revealed that the Down's syndrome patients possessed significantly larger numbers of those species than did generally healthy subjects[9]. In this report, we describe oral manifestations in a Japanese girl with triple-X syndrome and results of microbiological analysis.

Case Report

Case description and oral findings

A girl aged 5 years (Y) and 9 months (M) was referred to the Pedodontic Clinic of Osaka University Dental Hospital by her pediatrician for treatment of multiple dental caries. She was reported to be complicated with triple-X syndrome, with developmental retardation. Our initial intraoral examination revealed 11 primary teeth; 9 other primary teeth had been extracted due to dental caries prior to visiting our clinic. According to her mother, she was born after a 40-week gestation without any abnormalities or delivery complications. The first primary tooth had erupted at the age of 12 months. According to the record, her height was 48.0 cm (approximately 25th percentile) and body weight was 2,720 g (approximately 50th percentile) at the time of birth, whereas those at 6 months, and at 1, 2, and 5 years of age showed her to be lower than the 10th percentile. At 6Y1M and 6Y3M, we performed pulpectomies on the maxillary left and right second primary molars, respectively. The maxillary right primary lateral incisor and canine, mandibular right and left primary central incisors, and right primary lateral incisor were also extracted, followed by the application of partial dentures.

At 7Y6M, the mandibular central incisors, lateral incisors, and first permanent molars began to emerge into the oral cavity, and eruption of maxillary central incisors was confirmed at 9Y1M. Since retarded development of all permanent teeth was suspected, orthopantomograph images were routinely taken to evaluate tooth development at 10Y2M, 13Y1M, and 14Y5M (Fig. 1). Although there were no congenitally absent teeth, all teeth showed retarded development. Table 1 shows a comparison of chronological age and dental age, which was calculated by the developmental stage of the permanent teeth in orthopantomograph images by a method described previously[10]. After analyzing 5 images taken from 10Y2M to 14Y5M, dental age was determined to be approximately 2 years behind chronological age.

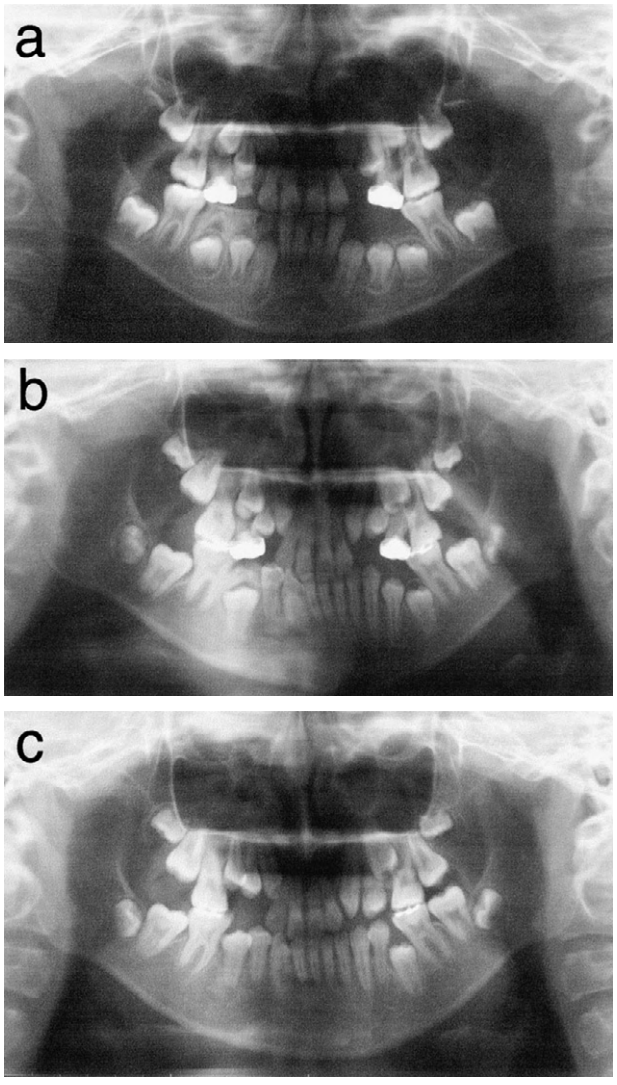

Fig. 1 Orthopantomographs images taken at 10Y2M (a), 13Y1M (b), and 14Y5M (c)

Table 1 Comparison of chronological and dental ages of the patient

Chronological age (years)	Dental age [mean ± SD (years)]
10Y 2M	7Y9M ± 1Y 1M
11Y11M	10Y3M ± 9M
13Y 1M	11Y2M ± 11M
13Y 7M	12Y0M ± 9M
14Y 5M	12Y8M ± 1Y 3M

Dental age was calculated by evaluating the developmental stages of the permanent teeth by the method described previously[10].