# Clinical observations of odontomas in Japanese children: 39 cases including one recurrent case

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**Summary.** Retrospective investigations of odontomas in Japanese children and one recurrent case were carried out. Thirty-nine cases of odontoma in 38 children were treated in the Paediatric Dentistry Clinic of Niigata University Dental Hospital between September 1979 and December 2002. The patients consisted of 23 males and 15 females and their ages ranged from 1 year 2 months to 14 years old. The chief complaints were delayed tooth eruption in 19 cases (five: primary teeth, 14: permanent teeth), retention of primary teeth in 11, incidentally found on the radiographic examination in eight cases, and swelling of the jaw in one case. Thirty-four cases (87%) were associated with tooth eruption disturbances. The most frequently affected region was the maxillary anterior region. Treatment consisted of surgical removal of odontoma in 30 cases, complex odontoma (n = 7), and compound and complex odontoma (n = 2). A retrospective study of the radiographs revealed the developing process of odontomas in four cases and odontoma disturbed tooth eruption since the early uncalcified developing stage.

A recurrent case was a boy aged 6 years 5 months in whom the first surgical removal of odontoma was performed at the age of 1 year 8 months. Recurrence of an odontoma is very rare, but in very young children odontomas are in the early developing stages, containing uncalcified portions, so it is important to perform periodical observations until the succedaneous teeth erupt.

# Introduction

Odontomas are classified as odontogenic tumours; however, they are thought to be developmental anomalies in which all dental tissues are represented, rather than benign neoplasms [1-4]. There are two types: complex and compound odontoma. In the former, individual tissues are mainly well formed but occur in a disorderly pattern, and in the latter all dental tissues are represented in a more orderly pattern than in a complex odontoma [1,2,5]. They are usually diagnosed during the first two decades of life [1-3,5-10], and in most children these tumours are associated with tooth eruption disturbances such as delayed eruption of the primary and permanent teeth or overly retained primary teeth [1,3,8,9,11]. Thus it is very important for paediatric dentists to understand the clinical features of odontoma in children.

There have been many studies of odontoma, clinical investigations [1,3–10,12,13] and case reports [14–18]; however, those investigations included odontomas in all age groups. We had reported 16 cases of odontoma in children [19] before experiencing a rare recurrent case. There have been few clinical investigations focused on children's

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odontoma or recurrent cases. This paper reports the clinical findings of 39 cases of odontoma in children, including a retrospective study of radiographs showing the developmental process in four cases and one recurrent case.

## Methods

We investigated 39 cases of odontoma in 38 Japanese children treated at the Paediatric Dentistry Clinic of Niigata University Dental Hospital between September 1979 and December 2002. The recurrent patient was counted as two cases of odontoma. The retrospective study used clinical records, radiographs, oral photographs and pathological reports. Data included ethnicity, age, gender, location, main complaints at presentation, radiographic findings, treatment, influences on dentition and pathological diagnoses.

## Results

## Age and gender

The patients included 23 males and 15 females (1.5:1) and their ages ranged from 1 year 2 months to 14 years (Fig. 1).

# Location

Twenty-six cases were located in the maxilla and 13 in the mandible.

Five cases were found in the primary dentition and 34 cases in the mixed or permanent dentition. In the primary dentition, two cases were in the maxillary primary central incisor, two in the upper primary canine and one in the lower second primary molar region.

As for permanent teeth, the maxillary central incisor region (eight cases) was the most frequently involved region, followed by the maxillary lateral incisor (seven cases). Figure 2 shows the relationship between the location and the pathological diagnosis.

## Main complaints at presentation

The chief complaints were delayed tooth eruption in 19 cases: five cases of the primary teeth and 14 cases of permanent teeth, retention of primary teeth in 11, incidentally found on radiographic examination in eight cases, and swelling of the jaw in one case.



Fig. 1. Distribution of age and gender.



Fig. 2. Location and pathological diagnoses of odontomas.

#### Radiographic findings

Periapical or panoramic radiographs revealed a radiopaque mass surrounded by radiolucent areas; however, in some cases, radiopacity was not quite clear and images of the teeth shadowed very tiny odontomas. The size of the odontoma ranged from the size of a tiny rice granule to 25 mm in diameter.

From the retrospective study of radiographs taken at different times in the same child, the developmental process of odontoma was observed from the early soft tissue stage to calcified radiopaque status in four cases (Figs 3–6). The interval between the two radiographs was 3 years 2 months, 2 years 2 months, 1 year 1 month, and 8 months, respectively.

## Treatment

All odontomas were surgically removed with or without extraction of the retained primary teeth.



Fig. 3. Development of an odontoma in the lower right canine region.



Fig. 4. Development of an odontoma in the upper left lateral incisor region.

Upon surgical removal of the odontomas, there were more extirpated substances than revealed in the radiographs.

In the cases associated with impacted teeth, the observation period was approximately 3 months after removal of the odontomas. Then, if the impacted teeth did not erupt, exposure of the tooth crown and/or orthodontic traction was applied.

## Influence on dentition and prognosis

In five cases there were no disturbances to the dentition because in one, the odontoma, existed distant from the dental arch, and four were found earlier than the eruption period. Also, all the odontomas were small.



Fig. 5. Development of an odontoma in the upper left central incisor region.



**Fig. 6.** Development of an odontoma and delayed eruption of the lower right lateral incisor (a: 6 years 10 months, b: 7 years 1 months, c: 7 years 6 months, d: 7 years 10 months).

Tooth eruption disturbances such as delayed eruption with or without displacement were observed in 34 cases (87%); five of which were in primary teeth. In one case, an impacted upper primary canine was extracted with the removal of the odontoma. As for the prognosis of the 33 cases after the removal of the odontomas, the impacted teeth erupted in 25 cases: eight spontaneously, six with surgical exposure of the crowns, two with orthodontic treatment, and nine with surgical exposure and orthodontic treatment. Six cases were referred to orthodontists or their routine care dentists and two remained under observation or treatment (Table 1).

# Pathological diagnoses

The pathological diagnoses were compound odontoma in 30 cases, complex odontoma in seven cases, and compound and complex odontomas in two cases.

	Erupted teeth $(n = 25)$					
	Spontaneous eruption	With surgical exposure (SE)	With orthodontic treatment (OT)	With SE + OT	Others $(n = 2)$	Unknown* $(n = 6)$
Primary teeth	2	1	0	0	0	1
Permanent teeth	6	5	2	9	2†	5
Total	8	6	2	9	2	6

Table 1. Prognosis of the 33 impacted teeth after surgical removal of odontomas.

\*Referred to orthodontists or home dentists.

<sup>†</sup>One under treatment and one under observation.

#### **Recurrent case report**

A boy aged 1 year 8 months was referred to our dental clinic with complaint of delayed eruption of the maxillary left primary central incisor. Radiographic examination revealed an irregularly shaped radio-opaque mass superimposed onto the crown of the unerupted incisor. Under local anaesthesia, the odontomas were removed surgically. The specimen was composed of a few small, calcified structures encapsulated with fibrous connective tissue (Fig. 7). Microscopically, the odontomas were constructed of immature enamel, dentin and pulp but there was no root formation. In the connective tissue capsule around the odontomas, a significant number of odontogenic epithelial islands were observed. There were several foci of dental papilla lined by ameloblasts but without dental hard tissue formation. It was in the early stages of odontoma formation. The pathological diagnosis was compound odontoma. Nine months after removal of the odontoma, the primary central incisor had not started to erupt, so the incisal edge was exposed surgically and it subsequently erupted. After that the patient was kept under observation at our clinic.

At the age of 6 years 5 months, periapical and panoramic radiographs were taken and recurrence of the odontoma in the left maxillary central incisor region was found. The recurrent odontomas were removed surgically (Fig. 8). Microscopically, the specimen was composed of several tooth-like calcified substances



**Fig. 7.** Recurrent case at the age of 1 year 8 months (a: impaction of the upper left central incisor, b: periapical radiograph, c: surgically removed odontoma).



**Fig. 8.** Recurrent case at the age of 6 years 5 months (a: periapical radiograph, b: surgical removal of odontoma, c: extracted upper left primary central incisor and odontoma).

surrounded by soft tissue. The calcified substances were composed of enamel, dentin and pulp. Odontogenic epithelium was seen in the surrounding connective tissue. The pathological diagnosis was compound odontoma. Seven months later the left maxillary central incisor had not erupted but the right side had erupted. Therefore, exposure of the incisal edge was performed and after 1 month the left maxillary central incisor began to erupt, erupting fully 6 months later. No further recurrence has been observed.

# Discussion

There have been many reports of odontomas; however, there are relatively few studies reporting more than 100 cases of odontoma [3,5–8]. From the analysis of previous data, the female incidence was  $47 \cdot 1-52\%$  suggesting no predilection by gender. In the present study, the female incidence was 39% (15/38).

Many studies have reported that odontoma occurs most frequently in the second decade of life [1-3,5-10]. Katz used 5-year intervals to describe a frequency distribution and reported that odontomas were most commonly removed from the 11–15-year-old age group. In the present study, 50% were in the first decade of life and the youngest patient was 1 year 2 months old.

As for location, the incidence of odontomas in the maxilla was 50.9-59.3% [3.6-8]. In studies by Regezi [6] and Kaugars [8] the most common location was the anterior portion (incisor and canine region) of the maxilla followed by the anterior portion of the mandible. Kaugars [8] also reported that the percentage of odontomas in the molar region gradually increased with each successive decade of life. Katz [5] reported that odontomas were apparently age and location related. Those from incisor locations were diagnosed and treated at an earlier age than those from the canine or third molar regions. In the present study, the maxillary central and lateral incisor region was the most common location. Katz [8] reported that odontomas rarely involved the primary dentition and found only five of 396 odontomas (2%), with the youngest being a 2-year-old patient. In the present study, we reported five cases of odontoma in the primary dentition, with the youngest case being a 1-year 2-month-old girl [16].

There have been many studies [1,5,11,15–18] in which odontomas caused various disturbances to tooth eruption. We previously reported tooth eruption disturbances of the primary and permanent teeth [20-27]. In those cases, odontoma was always included in the local aetiologic factors of impaction or delayed eruption of the teeth. Katz [5] studied 396 cases of compound, complex and mixed odontomas in all age groups and found that 41% were associated with unerupted teeth. Kaugars [8] reported that unerupted teeth were found in 47.6% (167/351). In the present research, 87% (34/39) of odontomas were related to tooth eruption disturbances. This percentage is higher than previous studies because the odontomas were found in children at a paediatric dentistry clinic. The most frequent cause of discovery of an odontoma is impaction of the permanent teeth with or without persistence of the primary teeth or, less frequently, symptomless swelling or accidental radiographic finding [1]. In the present cases, 49% of the chief complaints were delayed tooth eruption, 28% were retention of the primary teeth, 20% were incidentally found on radiographs, and 3% were swellings of the jaw.

The radiographic findings of odontoma were previously described as radiopaque areas surrounded by radiolucent areas. In the reported cases, most odontomas were shown as well-calcified masses on radiographs [14,17]; however, a few cases associated with delayed tooth eruption were not diagnosed on the first radiographic examination [15,28]. The present retrospective study of radiographs taken at different times in the same child revealed that at the early developmental stages of odontoma, calcification remains immature and is difficult to diagnose on radiographs. These findings could be obtained only in odontoma in children. Moreover, it becomes clear that from the early stages such a developing force could disturb tooth eruption. This suggests that when delayed eruption of the teeth is found, periapical radiographs should be taken to clarify whether any small area of radiopaque material exists.

As for treatment of odontomas, surgical removal with or without denudation of the impacted teeth is usually performed. Morning [11] examined 42 cases of impacted teeth in relation to odontomas in a 3–22-year-old group and reported that 17 of 35 impacted teeth erupted after removal of odontomas with or without exposure of the impacted teeth. In the present study, eight of 25 impacted teeth erupted spontaneously after surgical removal of odontomas. An observation time of three or more months after the first removal was used in the present study. If the impacted teeth had not erupted, then surgical exposure of the teeth crown with or without ortho-dontic traction was performed.

As for the recurrent case, we previously reported this patient as one of two 1-year-old cases of compound odontomas associated with impacted maxillary primary central incisors [16]. The reason for the recurrence must have been the existence of a residual mass at the first operation; however, it may have been because the odontoma was at the early developmental stage and the residual mass was not calcified [2]. At the age of 6 years 5 months, the odontoma was incidentally found on radiographs, and had developed to a size larger than the first tumour. The duration of development was 4 years 9 months. This case suggests that odontomas are apparently a developmental anomaly, even if they are not benign neoplasms.

If an odontoma is found in a very young child, it should be considered that it contains an uncalcified part, although it is usually a calcified mass, and periodic observations are important until the succedaneous tooth erupts.

**Résumé.** Une analyse réetrospective a été menée sur les odontomes chez les enfants japonais et chez un cas récurrent. Trente neuf cas d'odontomes chez 38 enfants ont été traités au sein dela clinique de dentisterie pédiatrique du Niigata University Dental Hospital entre septembre 1979 et décembre 2002. Les patients comprenaient 23 garçons et 15 filles, âgés de 1 an et 2 mois à 14 ans. La principale conséquence était le retard d'éruption dans 19 cas (5 dents temporaires, 14 dents permanentes), la rétention de dents temporaires dans 11 cas, la découverte accidentelle à l'examen radiographique dans 8 cas, et un œdème de la mâchoire dans un cas. Trente huit cas (87%) ont été associés à des troubles d'éruption. La région maxillaire antérieure était la plus fréquemment atteinte. Le traitement a consisté en l'avulsion des odontomes, suivie, si la dent impactée ne sortait pas, d'un dégagement de la couronne avec ou sans traction orthodontique. IL s'agissait d'odontomes composés dans 30 cas, d'odontomes complexes (7) et d'odontomes complexes et composés (2). Une étude rétrospective des radiographies a révélé le processus de développement des odontomes dans 4 cas, les odontomes perturbant l'éruption dentaire depuis le stade précoce non calcifié. Un cas d'odontome récurrent a été détecté chez un garçon de 6ans 5 mois chez lequel l'ablation chirurgicale de l'odontome a été réalisée à l'âge de 1 an 8 mois. La récurrence de l'odontome est très rare, mais chez les très jeunes enfants les odontomes sont à des stades de développement précoces, comportant des zones non calcifiées. Il est donc important d'effectuer des contrôles périodiques jusqu'à éruption des dents successionnelles.

Zusammenfassung. Eine retrospektive Untersuchung von Odontomen bei 38 japanische Kindern (davon ein Fall mit Rezidiv) wurde durchgeführt. Die 39 Odontome bei 38 Kindern wurden in der Kinderzahnheilkunde der Niigata Universitätszahnklinik behandelt zwischen September 1979 und Dezember 2002. Bei den patienten handelte es sich um 23 Jungen und 15 Mädchen im Alter von 1 Jahr und 2 Monate bis 14 jahren. Hauptsymptom war verzögerter zahndurchbruch (5 Milchzähne, 14 bleibende Zähne) Retention von Milchzähnen (11 Fälle, davon 8 zufällig röntgenologisch festgestellt) und Kieferschwellung in einem Fall. Vierunddreißig Fälle (87%) waren mit Zahneruptinsstörungen vergesellschaftet. Am häufigsten betroffen war die Oberkiefer-Frontzahnregion. Die Behandlung bestand in allen Fällen aus der chirurgischen Entfernung der Odontome, falls impaktierte Zähne danach nicht spontan durchbrachen wurde eine Freigegung und/ oder kieferorthopädische Mobilisierung vorgenommen. Pathohistologisch handelte es sich in 30 Fällen um ein zusammengesetztes Odontom, in 7 Fällen um ein

komplezes Odontom und in 2 Fällen um zusammengesetztes und komplexes Odontom. Eine retrospektive Analyse von Röntgenbildern zeigte in vier Fällen die Entwicklung der odontome, die Zahneruptionsbehinderung war bereits in frühen, nichtcalcifizierten Stadien gegeben. Ein Rezidiv wurde bei einem Jungen im Alter von 6 Jahren und 5 Monaten festgestellt, er war im Alter von! Jahr und 8 Monaten erstmals operiert worden. Ein Rezidiv ist selten, aber bei einem sehr jungen patienten könnte uncalcifiziertes Odontomgewebe belassen werden, daher ist die Nachkontrolle bis zur Eruption der bleibenden Zähne erforderlich.

Resumen. Se exponen investigaciones sobre odontomas en niños japoneses y un caso recurrente. Entre septiembre de 1979 y diciembre de 2002 en la clínica de Odontopediatría del hospital dental de la Universidad de Niigata, se trataron 39 casos de odontoma en 38 niños. Los pacientes fueron 23 varones y 15 niñas y sus edades variaron desde 1 año y 2 meses hasta 14 años. Los motivos de consulta principales fueron retraso de la erupción en 19 casos (5 dientes primarios, 14 dientes permanentes), retención de 11 dientes primarios encontrados de forma casual en el examen radiográfico de 8 casos e inflamación de la mandíbula en 1. Treinta y cuatro casos (87%) se asociaron con alteraciones de la erupción dentaria. La región más frecuentemente afectada fue la región anterior maxilar.

El tratamiento consistió en la extracción quirúrgica de los odontomas en todos los casos, después de lo cual si los dientes impactados no erupcionaban, se realizaba exposición de la corona y/o tracción ortodóncica. El diagnóstico anatomopatológico fue odontoma compuesto en 30 casos, odontoma complejo (7) y odontoma compuesto y complejo (2). Un estudio retrospectivo de las radiografías revela el proceso de desarrollo de los odontomas en cuatro casos y alteración de la erupción dentaria por el odontoma desde el estadío de desarrollo temprano no calcificado. Un caso recurrente fue un niño de 6 a y 5 m de edad en el que la primera extracción quirúrgica del odontoma se realizó a la edad de 1 a y 8 m. La recurrencia del odontoma es muy rara, pero en niños muy pequeños los odontomas están en estadíos de desarrollo temprano y contienen porciones no calcificadas, así que es muy importante realizar observaciones periódicas hasta la erupción de los dientes sucesores.

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