Congenital defect of maxillary primary central incisor associated with exposed pulp and gingiva: case report

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This report describes a rare case of hypoplastic primary incisor in which the pulp was exposed at the crown portion and covered by the gingiva in a 1-year-11-month-old boy. The patient was referred to us due to swelling of his labial cervical gingiva of the maxillary right primary central incisor, and on examination, extended to the hypoplastic labial surface. Radiographically, there was a round radiolucent area on the crown including the edge. Surgical removal of the swollen gingiva revealed a large defect of the labial aspect of the incisor, showing pulpal tissue inside. The tooth was treated by vital pulpotomy. Histopathologically, the removed gingival tissue contained many pieces of dysplastic tooth elements in the lamina propria portion which should have been connected to the exposed pulp. The findings suggested that pulp exposure resulted from focal dental hypoplasia not from resorption of the tooth.
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INTRODUCTION

Tooth defects result from various disturbances in the developing process of the tooth germ and are caused by general factors such as a dystrophy, hormonal imbalance, medicament, and heredity, or local factors such as a trauma, inflammation and radiation.[1] Tooth defects of primary teeth are rare compared with that of permanent teeth, because most processes of primary tooth germs begin in the stabilized womb.[2-4]

The degree of hypoplasia differs depending on the developmental stage of the tooth germ, or the kind and intensity of disturbances. Clinically, defects are visually and morphologically identified by various irregularities, such as pits, grooves, or absence of enamel structure over the tooth surfaces.[1] However, hypoplastic lesions are usually localized within the enamel and dentin, and we could not find any reports, which showed a hypoplastic lesion had penetrated the pulp.

This report describes a case of severely affected hypoplastic maxillary right primary central incisor with exposed pulp, and swollen gingiva connected to the pulp. The swollen gingiva was surgically removed, the tooth was treated by vital pulpotomy, and it has remained asymptomatic.

CASE REPORT

A Japanese boy aged 1 year and 11 months was referred to the Pedodontic Clinic of Niigata University Dental Hospital due to swelling of the cervical gingiva of the maxillary right primary central incisor. The tooth had erupted at the age of 8 months, and the labial surface had been covered with the gingiva since then. There was no relevant medical history, or family history of dental abnormalities.

On intraoral examination, the following teeth were erupted:

DCBA | ABCD (54, 53, 52, 51; 61, 62, 63, 64
DCBA | ABCD 84, 83, 82, 81; 71, 72, 73, 74)

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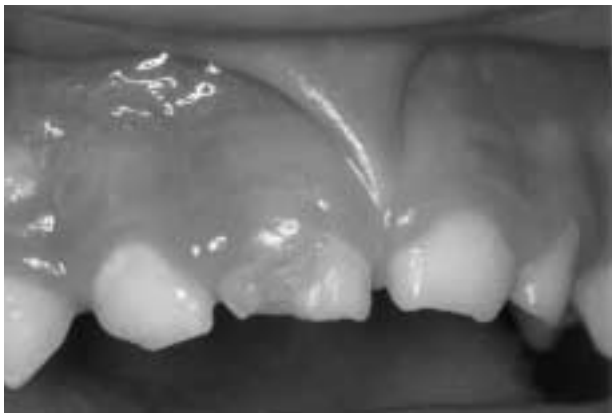

Figure 1. Photograph showing the swelling of cervical gingiva of maxillary right primary central incisor, which extended to the hypoplastic labial surface.

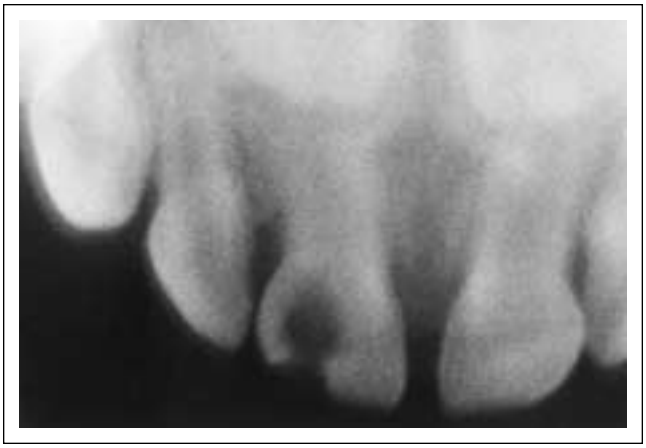

Figure 2. Radiograph showing the round radiolucent area connected to the pulp chamber.

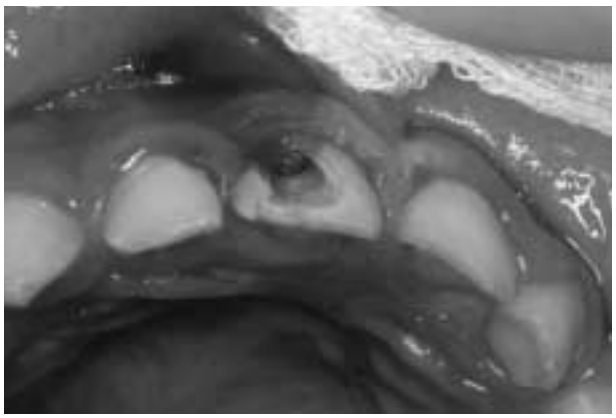

Figure 3. Photograph after the swollen gingiva was surgically removed, and the central incisor was treated by vital pulpotomy.

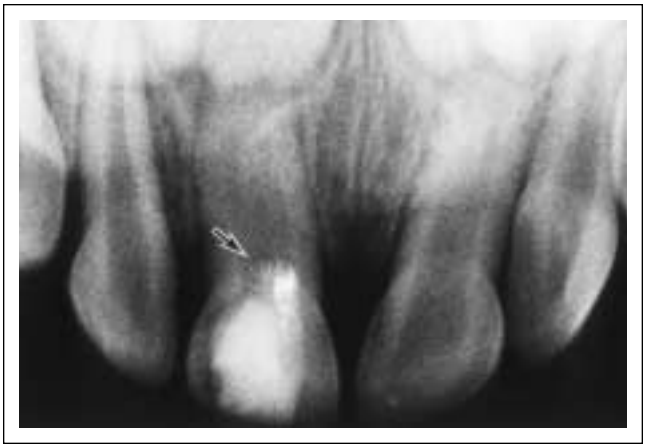

Figure 4. Radiograph showing the formation of the dentin bridge 1 year and 3 months after the operation (arrow).

The labial cervical gingiva of the maxillary right primary central incisor was swollen, and extended to the hypoplastic labial surface. The incisor edge was slightly concave, however, clinical examination revealed there was neither caries, pain nor mobility. The palatal gingiva was normal (Figure 1).

Radiographic examination revealed a round radiolucent area connected to the pulp chamber. Root formation of the tooth was about two thirds and appeared to be normal in development (Figure 2). The lesion was clinically diagnosed as epulis.

Under local anesthesia with 2% lidocaine, the swollen gingiva was surgically removed from the labial surface of the incisor, exposing vivid pulp tissue in a defected labial surface of the incisor. The central incisor was treated by vital pulpotomy with a calcium hydroxide paste, and restored with composite resin (Figure 3). The tooth has remained asymptomatic and formed a dentin bridge 1 year and 3 months after the operation (Figure 4).

The specimen was histopathologically examined (Figure 5). Microscopically, the mucosal epithelium of the gingival tissue showed parakeratosis and acanthosis. There was fibrosis in the lamina propria. At the surgical edge of the specimen, where the gingival tissue was connected to the pulp, there was inflamed dental sac like myxoid connective tissue containing many pieces of dysplastic dental elements which consisted of dentin and enamel matrixes (Figure 6). Histopathological diagnosis was granulation tissue with pieces of dysplastic dental substance.

DISCUSSION

Hypoplastic teeth show many clinical features such as white spots, pits, grooves, or absence of enamel structure. The incidence of enamel defects of the primary teeth are reported to be from 5.9 to 33.0% in healthy children.[5-11] However, hypoplasia of primary teeth is rare compared with that of permanent teeth. Teeth most commonly involved are maxillary primary central and lateral incisors (40.8 % and 39.2 % in each), followed by maxillary primary canines (25.7 %).[11] Clinically, defects are visually and morphologically

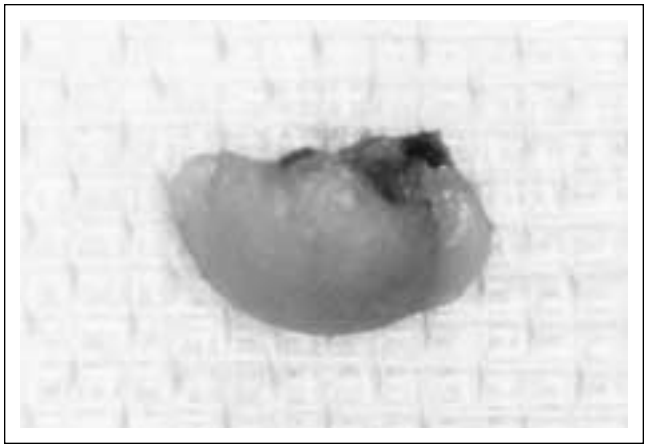

Figure 5. Surgical specimen of gingival tissue.

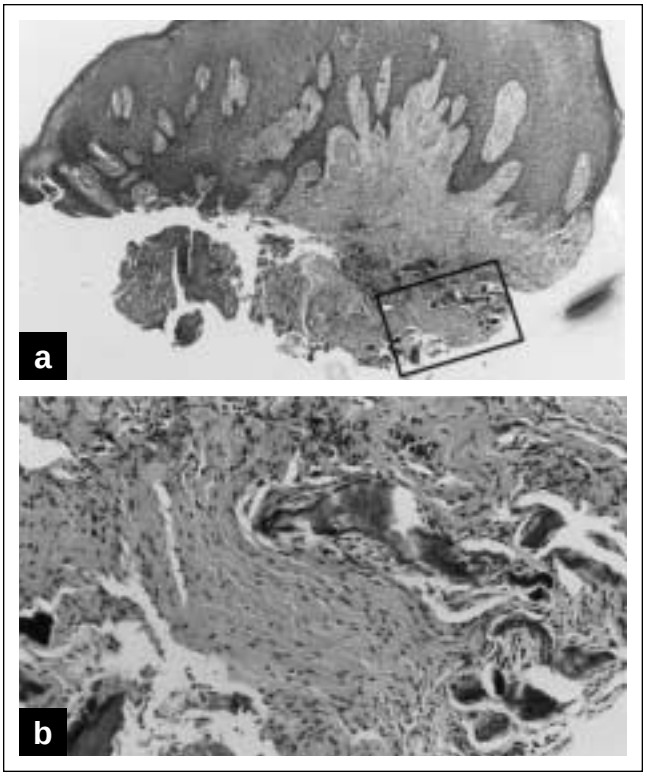

Figure 6. a: Histology of the gingival tissue. Many pieces of dysplastic dental_substance were scattered in the fibrous granulation tissue at the lower edge of the gingival tissue (Haematoxylin and eosin, x220). **b:** Higher-power view of dysplastic dentin fragments (x 1,050).

identified by various irregularities over the tooth surfaces,[1] but they are usually localized within the enamel and dentin. We were unable to find a report of hypoplastic lesion penetrating the pulp.

There are two causes of unerupted tooth defects with exposed pulp. One is tooth hypoplasia in which tooth formation is absent from the beginning. The other is tooth resorption which occurs after the completion of tooth formation. To date, several cases of unerupted permanent tooth defects with exposed pulp were reported, and most were thought to be caused by tooth resorption.[12-15] In these reports, resorption in unerupted teeth was considered to occur due to destruction of the reduced enamel epithelium as a result of factors such as apical inflammation of the primary tooth, trauma, orthodontic therapy and pressure from local disease (tumors and cysts).[13,15-18] In normal development, the crown of an unerupted tooth is covered by reduced enamel epithelium. Blackwood[12] and Skaff *et al.*[19] reported that a breakdown in this protective layer exposed enamel to surrounding vascular connective tissue, allowing resorption to occur.[12,14,19,20]

In our case, the gingival tissue remained covered on the defect part of the crown of the incisor during the eruption process. Usually, the crown of primary central incisors is completed between 1.5 months and 2.5 months after the birth. Since the patient had no obvious medical history such as local inflammation or trauma, which may cause tooth defects from birth to the beginning of eruption at 8 months of age, it is likely that the tooth defect did not occur after the birth.

In the histopathological findings, there was inflammatory evidence in the gingival tissue covering the crown. However, judging from the mild degree and the small extension of the organization, the lesion could not have caused resorption of a completely formed tooth crown. Therefore, the granulation tissue of the gingiva should be regarded as a result of traumatic stimuli. In addition, there was no multinucleated odontoclasts, which were observed in other cases of abnormal tooth resorption.[12,13,20,21] The presence of fibrosis in the gingival tissue may indicate that the resorptive activity stopped on the tooth surface in this case. In the pieces of dysplastic dental substance, there was no Howship's lacunae harboring clast cells or addition of osteodentin after resorption. This suggests that the dysplastic dental tissue was not a remnant of resorption of the incisor, and that pulp exposure resulted from localized dental malformation not from secondary resorption of the tooth.

For therapy of the severely affected hypoplastic teeth with exposed pulp, either pulp therapy (direct pulp capping, vital pulpotomy and pulpectomy), or extraction can be performed. In this case, the tooth was treated by vital pulpotomy, it has remained asymptomatic, and formation of the dentin bridge was confirmed 1 year and 3 months after the operation. We intend to make clinical observations of the succedaneous tooth.

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