

CASE REPORT

Impaction of multiple dilacerated primary incisors: a case report

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Abstract

Dental complications such as defective alveolar bone development, delayed eruption, and tooth impaction are related to neonatal oral intubation. This case report presents an example of potential complications that occur in children who have undergone oral intubation as neonates. A 20-month-old girl visited our pediatric clinic. We observed delayed, non-erupted teeth #51, #71, and #81 and determined a history of intubation during the neonatal period as a related factor. After 22 months of observation, tooth #71 erupted spontaneously. After 40 months of monitoring, teeth #51 and #81 were extracted surgically, and normal permanent teeth erupted six months later. This study is helpful for pediatric anesthesiologists, pediatricians, and dentists who diagnose and treat eruption disorders of the primary dentition.

Keywords

Oral intubation; Preterm infants; Dilaceration; Primary incisor

1. Introduction

Reports of impacted and dilacerated primary incisors are rare, and it is challenging to estimate related causes because reports on the prevalence of primary incisor dilaceration are lacking. Trauma is assumed to be the most common cause of impacted and dilacerated primary incisors because the few studies that address the etiology and treatment of impacted and dilacerated primary incisors focus on permanent teeth affected by trauma of primary teeth [1–3]. A number of studies has shown that trauma caused by oral intubation performed in preterm infants can be considered a risk factor of impacted primary incisors [4, 5].

Preterm infants born alive before 37 weeks of pregnancy represent one of every 10 babies born in the United States in 2020 [6]. In the Republic of Korea, the preterm birth rate was 8.1% in 2019 and is showing an increasing annual trend [7]. Globally, complications of preterm birth are responsible for more than 15% of deaths in children younger than five years, and many preterm infants experience problems with growth and physical and mental functions [8]. Preterm infants experience several medical complications [9–11]. Short-term complications include heart problems such as patent ductus arteriosus or low blood pressure and brain problems such as intraventricular hemorrhage, temperature control problems due to lack of body fat, and high infection risk due to underdeveloped immune systems. Long-term complications include cerebral palsy and vision, hearing, and dental problems. Among these complications, respiratory distress syndrome is common due to incomplete lung maturation [11]. If the lungs lack surfactant, which allows the lungs to expand and contract without major friction, the resultant lack of normal movement can lead to

respiratory distress syndrome. In addition, respiratory complications such as prolonged pauses in breathing or apnea may occur, and mechanical ventilation may be essential for survival. The World Health Organization advises appropriate oxygen therapy such as continuous positive airway pressure for improving the survival of preterm infants who require resuscitation [12].

Orotracheal intubation is a common procedure in preterm infants because it is safe, rapid, and promotes airway patency using a non-surgical technique [13]. The procedure involves placing an oro-tracheal tube into the oral cavity and advancing it past the vocal cords and into the lower portion of the trachea. A laryngoscope is used to provide a detailed view of the airway during insertion of the oro-tracheal tube. Although laryngoscopy and oro-tracheal intubation are safe procedures, there is low risk of cervical or neck injury, laryngotracheal trauma, stenosis of the larynx, dysphagia, and/or vocal cord paralysis [13, 14]. Several studies have shown oral and dental complications associated with oro-tracheal intubation [15, 16]; however, a few reports suggest the possibility of dilaceration and impaction of primary incisors related to oral intubation.

This case report discusses a 20-month-old female patient who showed dilaceration and impaction of the primary incisors with a history of oral intubation.

2. Case report

In January 2018, a 20-month-old female patient visited the Department of Pediatric Dentistry of Jeonbuk National University Dental Hospital because her primary central incisors had not yet erupted. She was born prematurely at 34 weeks with a low

birth weight of 2000 g. The mother had been diagnosed with gestational diabetes mellitus and was at risk of preeclampsia with a blood pressure of 180/100 mmHg without medication. The patient did not show respiratory symptoms immediately after birth; however, when bottle feeding was attempted, oxygen desaturation and systemic cyanosis were observed, and ambu-bagging was performed. Although oxygen saturation was restored to normal level through ambu-bagging, bottle feeding was not possible. Feeding was performed using a G-tube that passed through the oral cavity. After two days, the G-tube was removed. Follow-up was performed in the pediatric department of our hospital, and no physical or mental developmental problems were detected.

At presentation, clinical examination revealed no eruption of teeth #51, #71, and #81 (Fig. 1A). Additional radiographic examination showed palatal displacement of #51 and lingual displacement of #71 and #81 (Fig. 1B). The parent reported no history of dental trauma in the patient. The treatment plan was assessed periodically to identify spontaneous eruption. Typically, if spontaneous eruption is not observed and continuous eruption of the succedaneous teeth is interrupted, extraction is planned.

Five months after the initial visit, bulging was observed at the mandibular primary central incisor (Fig. 2A). After 22 months, tooth #71 spontaneously erupted lingually (Fig. 2B). At 28 months, #71 was labioversed and returned to its normal position in the dental arch (Fig. 2C). Spontaneous eruption of tooth #71 was observed on the periapical radiography, and no other tooth movement was observed (Fig. 3A,B). At 40 months after the start of regular monitoring, movement of #41 and #11 successive permanent teeth in the alveolar bone was observed on periapical radiography (Fig. 3C). We posited that teeth #51 and #81 would interfere with eruption of successive permanent teeth and planned to surgically remove these teeth. The parents requested surgical extraction under general anesthesia. Before extraction, cone-beam computed tomography (CBCT) was performed to confirm the shape and location of teeth #51 and #81. Tooth #51 was displaced on the palatal side and showed no root growth. Tooth #81 was displaced to the lingual side, and the root was curved at 90° (Fig. 4).

In May 2021, surgical extraction of teeth #51 and #81 was performed under general anesthesia, and part of the surrounding alveolar bone of #81 was removed due to severe root curvature on CBCT (Fig. 5A). As expected, tooth #51 had little root growth, while the root of #81 was curved at 90° (Fig. 5B). The teeth were extracted with no complications (Fig. 5C).

Six months post extraction, eruption of the successive permanent teeth occurred earlier than that of their opposing counterparts, and normal uneventful eruption was observed (Fig. 6A,B). In addition, #41 erupted into the oral cavity. Fourteen months post extraction, the successive permanent teeth erupted with no other observed problems except slight crowding of the anterior teeth (Fig. 7A,B).

3. Discussion

Mechanical ventilation using techniques such as orotracheal intubation is essential for survival in preterm infants with respiratory problems [11, 12]. The common use of orotra-

cheal intubation can be attributed to its procedural simplicity and safety; however, studies have reported oral complications related to its use [15, 16]. Moreover, because intubation in preterm infants is very difficult due to poor view of the vocal cords, dental damage and trauma during laryngoscopy are not uncommon [13, 17].

Many studies have reported that enamel defects and mineral deficiencies such as enamel hypoplasia in the primary dentition are likely to be increased in preterm infants [17–20]. Seow *et al.* [17] reported that 85% of intubated infants had defects of the maxillary anterior teeth, while only 21.7% of non-intubated infants had defective maxillary anterior teeth. Also, Melo *et al.* [19] reported that 86.3% of intubated infants had enamel defects, while only 13.7% of non-intubated infants had enamel defects. Seow *et al.* [17] suggested the cause of enamel defects in preterm infants to be forces exerted by the laryngoscope in traumatic swing maneuvers during laryngoscopy rather than by orotracheal intubation. This suggestion is supported by Melo *et al.* [19], who showed that laryngoscope blade area of action corresponded to the region most affected by tooth enamel defects in intubated infants. Severely preterm infants have a high possibility of hypoplasia of the mandible, and insertion of the laryngoscope can cause trauma to the anterior alveolar bone [17, 18]. In addition, the developing teeth of preterm infants may be more susceptible to local trauma due to thinner cortical bone structure due to osteopenia. Here, we suspected that root dilaceration underdevelopment and arrest of root development were due to oral laryngoscope intubation based on the medical history of premature birth but no dental trauma. Elevation of the laryngoscope blade is required to displace the tongue and expose the pharyngeal region. However, inadvertent leverage force can be exerted on the maxillary anterior alveolar ridge because the tongue is larger than normal in relation to the mandible and the anatomic oral cavity structure of preterm infants does not provide a sufficient fulcrum for lifting the anterior oropharynx. Although the details of intubation in this patient were unknown, it is possible that upper and lower arches were affected during laryngoscopy, considering the effects on the maxillary and mandibular primary incisors. Generally, laryngoscopy tends to affect the maxilla more than the mandible, although the affected side varies among studies, probably explained by different techniques and direction of leverage during laryngoscopy.

Alveolar and palatal grooves have been reported as complications related to orotracheal intubation [21, 22]. Erenberg and Nowak reported that 23.1% of preterm infants who underwent orotracheal intubation showed alveolar contour change and showed a strong association between orotracheal intubation and palatal grooving in preterm infants [21]. In another study, neonate children requiring orotracheal intubation were observed to have grooves on their upper alveolar ridge [23]. Careful observation in the Neonatal Intensive Care Unit revealed that the continuous force exerted by the orotracheal tube produced a pale ischemic area on the alveolar ridge, eventually causing indentation. As a result, long-term use of the orotracheal tube and the continuous pressure exerted by the tube affect the formation of the anterior alveolar ridge more so than does laryngoscopy [22, 24]. The prevalence of right-sided alveolar grooves can also be explained by the practical habit of

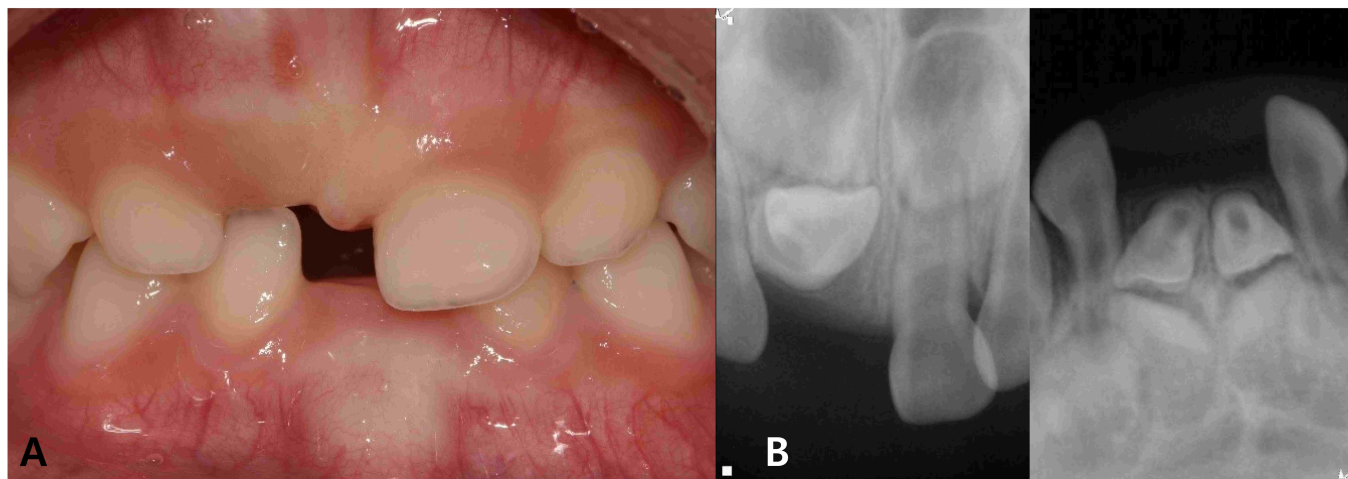


FIGURE 1. Condition of the primary incisors at the initial visit. (A) Intraoral photo at the initial visit, with no eruption of teeth #51, #71, and #81. (B) Periapical radiograph of teeth #51, #71, and #81.



FIGURE 2. Intraoral photo of the mandibular incisor area. (A) Bulging on teeth #71 and #81 (2Y 1M). (B) Lingual eruption of tooth #71 (3Y 7M). (C) Labioversion of tooth #71 (4Y 1M).

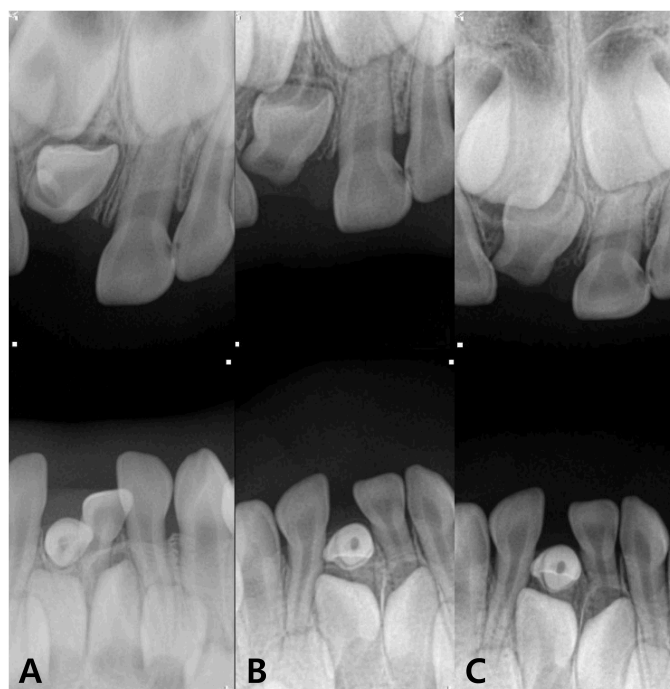


FIGURE 3. Periapical radiograph of the primary incisors. (A) Lingual eruption of tooth #71 (3Y 3M). (B) Tooth #71 spontaneously fully erupted (4Y 7M). (C) Movement of the successive permanent teeth #11 and #41 in the alveolar bone (5Y 1M).



FIGURE 4. Sagittal view of cone-beam computed tomography of teeth #51 and #81.

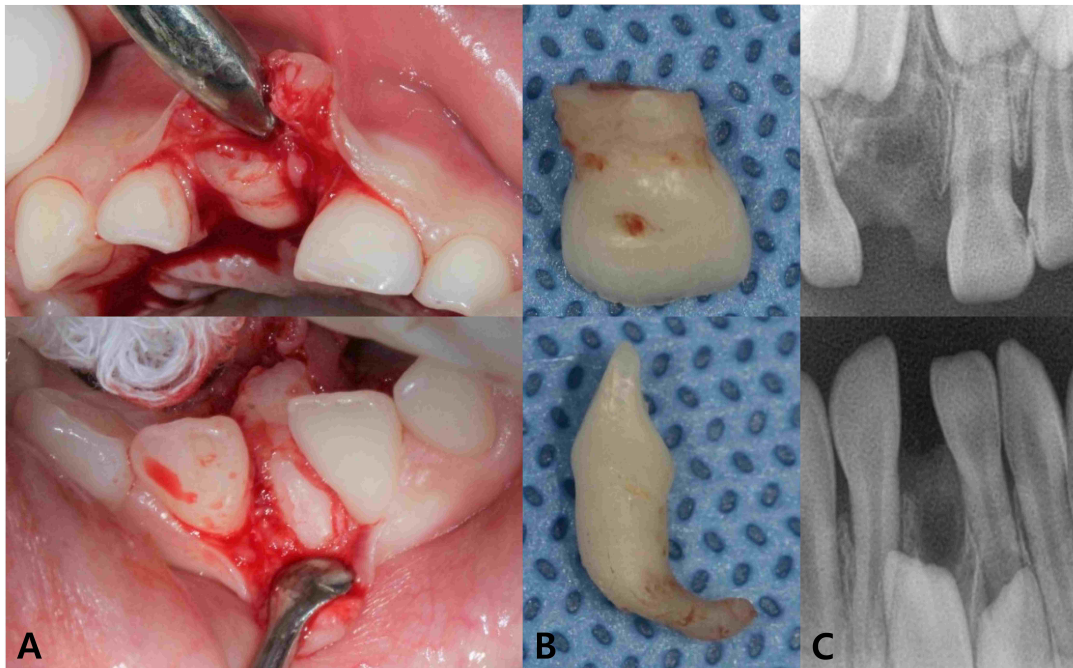


FIGURE 5. Extraction of impacted teeth #51 and #81 under general anesthesia. (A) Teeth #51 and #81 were displaced to the palatal and lingual sides, respectively. (B) Crown and root shapes of teeth #51 and #81. (C) Periapical radiograph after extraction.

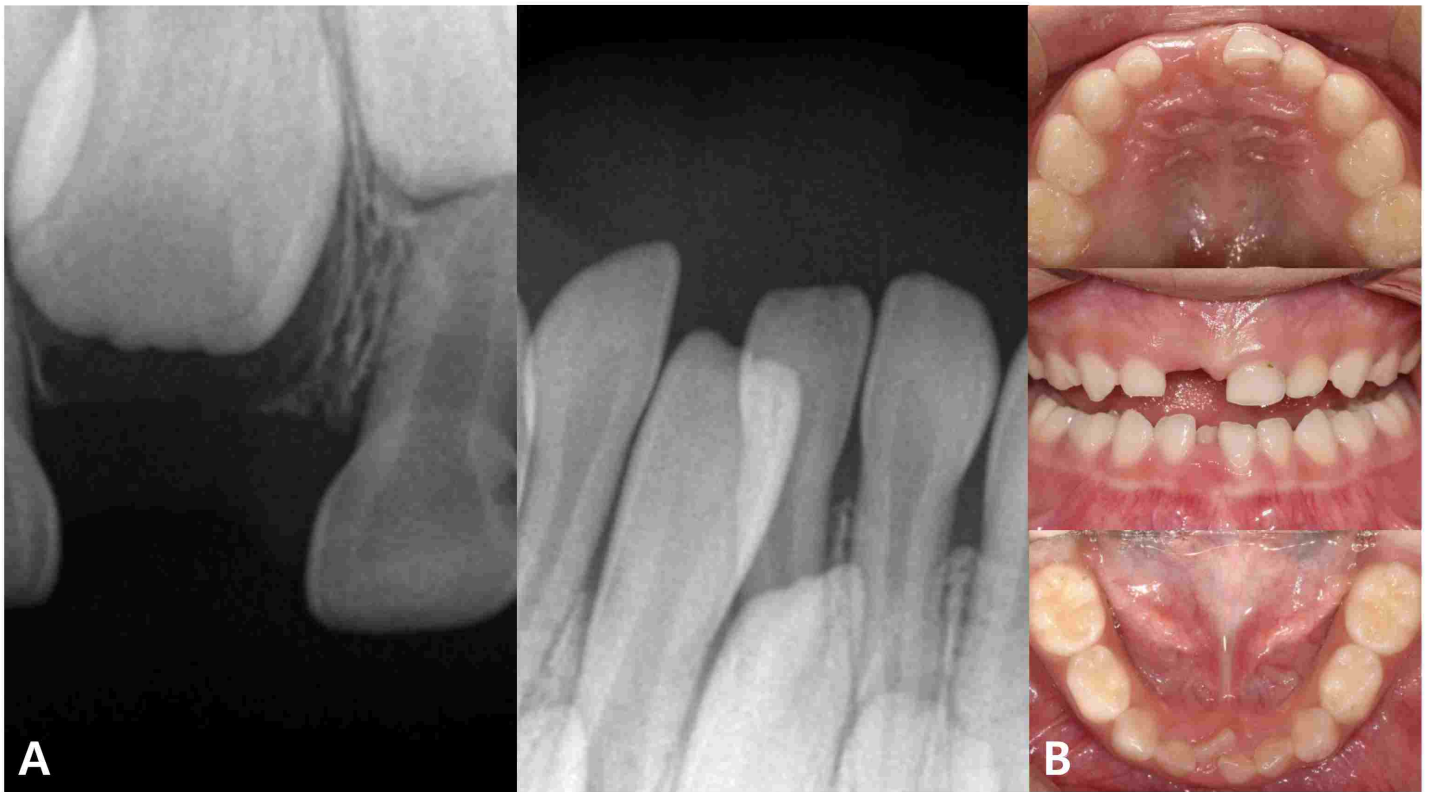


FIGURE 6. Six months after tooth extractions. (A) Periapical radiograph of teeth #11 and #41. (B) Intraoral photo.

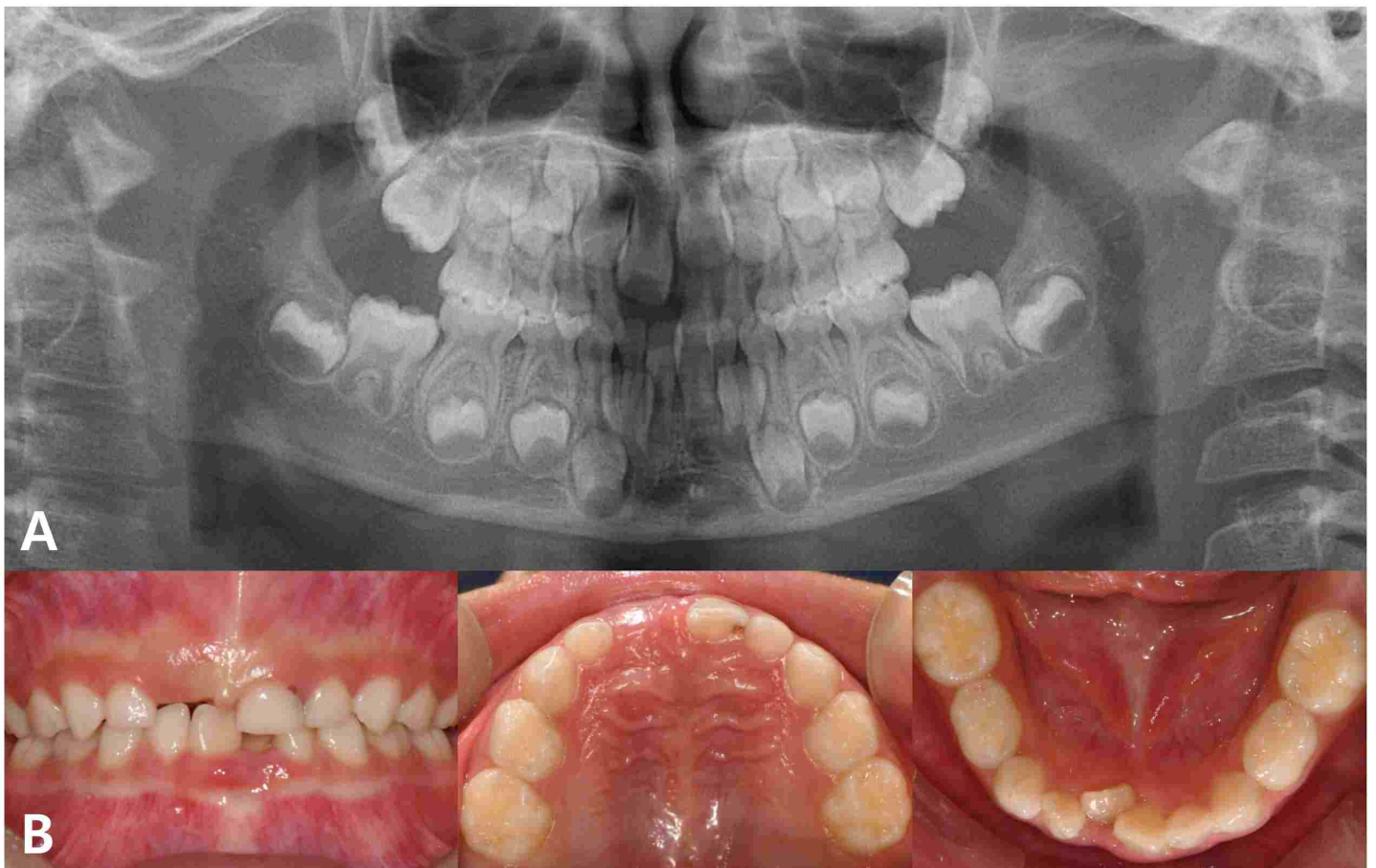


FIGURE 7. Image captured 14 months after teeth extraction. (A) Panoramic radiograph. (B) Intraoral photo. Tooth #41 was labioversed and tooth #31 erupted well.

securing the orotracheal tube on the right side of the mouth. Fortunately, in the presented case, the child had a normal alveolar arch, which was likely due to use of oral intubation for only two days.

Dental defects such as enamel hypoplasia, as well as the rate of dental development and eruption, may be affected by premature birth [25, 26]. Children with low birth weight and preterm birth have low rates of dental development, especially before six years of age. The primary incisors start to form hard tissue at four months in utero, and the enamel is mature between one and three months after birth. Eruption of the primary incisors begins six months after birth. The timing of oral intubation in preterm infants is closely related to the stage of enamel completion and the eruption of primary incisors. If the intubation tube or laryngoscope causes trauma to the alveolar ridge at this stage, the possibility of delayed eruption or late dental development increases [19]. Mason *et al.* [26] reported a 22-month-old boy who required orotracheal intubation at nine days old who presented with non-eruption of his maxillary primary central incisors. The authors suggested that delayed eruption, abnormal morphology of primary incisors, and follicular displacement were due to continuous pressure on the mucosa by the tube.

Delayed eruption and tooth impaction in the primary dentition are rare compared with those of the permanent dentition. Causes of delayed eruption and impaction in primary dentition include systemic diseases such as ectodermal dysplasia, cleft palate, and oral trauma that result in thick scar tissue formation. Such tissue hinders eruption of primary teeth and can damage the alveolar ridge [27]. In the presented case, delayed eruption and tooth impaction were observed in some of the primary incisors. Considering the absence of systemic disease or daily activity trauma history and impaction of only some primary incisors and not all teeth, it was presumed that trauma during oral intubation after premature birth delayed the eruption of incisors and impaction.

To prevent such oral complications, intraoral protective devices have been created for orotracheal intubation [28–30]. To prevent continuous force from being transmitted to the palate or alveolar ridge during orotracheal intubation, the tube is separated from the oral tissue using acrylic resin. Several studies have reported that the incidence of oral complications was significantly lower in groups with intraoral protective devices than in those without [28]. However, intraoral protective devices are not often used in practice. According to Pamukcu, 83.2% of anesthesiologists and 95.7% of neonatologists are aware of oral complications related to orotracheal intubation in preterm infants [16]; however, 98.3% of them preferred attaching adhesive tape to the perioral region to stabilize the orotracheal intubation tube rather than using devices. Only 1.3% of such medical providers were found to use intraoral protective devices in clinical practice [16].

If oral intubation is essential in premature birth, doctors must be aware of potential dental complications during the intubation process and must be equipped to obtain informed consent from parents. In addition, parents should fully understand the oral complications that may be caused by oral intubation and should consult pediatric dentists if complications such as delayed eruption are observed. Pediatric dentists should be

aware of the possibility of oral complications that may occur in preterm infants who undergo oral intubation and should be able to perform differential diagnosis between trauma due to oral intubation and that due to general daily life through appropriate collection of medical history. In addition, as oral intubation and localized disorders of dental development are interrelated, pediatric dentists should recall patients periodically to monitor their dental development for delayed eruption or impaction in the primary dentition due to the possibility of spontaneous eruption [26]. If impacted or dilacerated primary teeth are expected to interfere with eruption of successive permanent teeth, the pediatric dentist should surgically remove them at an appropriate time. Alternatively, orthodontic treatment may be considered if tooth dilaceration is not severe and patient cooperation is good.

4. Conclusions

We present a rare case of dilaceration and impaction of the primary incisors. Although the exact cause is not known, oral intubation performed during neonatal treatment is a risk factor for this case. Owing to the possibility of spontaneous eruption, we performed regular monitoring and surgical extraction at an appropriate time for normal succedaneous tooth eruption. If a patient's primary teeth are impacted or dilacerated, pediatric dentists must be able to distinguish, through history taking, whether the symptoms are caused by systemic disease, local factors interfering with eruption, or by trauma during the neonatal period. Also, pediatric dentists must be aware of the possibility of oral complications in preterm infants who have previously undergone oral intubation to make an appropriate treatment plan based on accurate diagnosis.

AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

AUTHOR CONTRIBUTIONS

DWL and SEM—designed the research study. DWL, SEM, JGK and YMY—performed the research. DWL, SEM, JGK and YMY —analyzed the data. DWL and SEM —wrote the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was approved by the Institutional Review Board of Jeonbuk National University Hospital (IRB No: 2022-11 048), and written informed consent was obtained from the patient's guardian.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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