Separate apical root formation of injured immature teeth: a case series and literature review
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Abstract
This report presents three cases of separated root tip formation following traumatic injury with or without inflammation. The first case showed continued separate apical root formation; however, development of the main root with bony ingrowth was absent. The second case showed that separated root formation continued under the 2-year inflammatory conditions, even after apexification. The third patient showed partially connected root tip formation after apexification. By summarizing previously reported similar cases, we assessed the injury type, prevalence age, treatment performed and pattern of root development. This phenomenon may be elucidated by the special features of the apical papilla (AP). Understanding the capability of AP could help with treatments, such as regenerative endodontic procedures and trauma management for immature permanent teeth.

Keywords
Apical papilla; Hertwig’s epithelial root sheath (HERS); Separated root tip; Trauma; Inflammation

1. Introduction
Dental trauma occurs frequently and involves 5% of all injuries. The trauma takes more place with preschool children, where 18% of all injuries are involved. The traumatic injury could cause loss of pulp vitality, which could halt root development. Then, the root may remain in an immature status having an open apex [1].

The essential contribution to the root formation was the function of Hertwig’s Epithelial Root Sheath (HERS) and apical papilla (AP). The apical part of an immature root consists of the apical papilla (AP), a smooth-surfaced soft tissue that is exposed apically from the pulp tissue and can easily be detached from it [2]. The apical part of the papilla is known to compose of HERS. Mesenchymal tissue of the AP, which comes in contact with the inner layer of Hertwig’s epithelial root sheath (HERS), is differentiated into odontoblasts, and then radicular dentin is formed [3]. HERS, a bilayer epithelial structure formed from ectodermally derived outer and inner enamel epithelia, migrates apically and guides root formation, including the number, size and shape of the roots [4, 5]. It is a morphological boundary between two dental ectomesenchymal tissues: the dental papilla and follicle. It also prevents the ingrowth of periapical tissue into the root, providing space for root dentin formation derived from the AP [5–7].

If AP and HERS are damaged owing to trauma, infection or iatrogenic factors, normal root formation may be disturbed [8]. However, some case reports have demonstrated their capability to maintain continuous root formation in the aforementioned environments [6, 9, 10]. Most of them revealed separated root tip formation, which has been named “phantom root”, “separated root” or “detached root” [10, 11].

This report presents three cases of separated root tip formation following traumatic and inflammatory injuries. Additionally, injury type, age of prevalence, treatment type and patterns of root development are also discussed, along with a summary of similar previously reported cases.

2. Case report
2.1 Case 1
A 10-year-old boy with a non-contributory medical history was referred from the emergency room for further treatment of traumatized teeth. The patient fell while riding a bicycle and the left upper lateral incisor was avulsed. The tooth was repositioned and splinted 3 h after the traumatic incident. At the first visit to the Department of Conservative Dentistry at Dankook University Dental Hospital, a clinical examination revealed sensitivity to percussion and palpation, and the left upper lateral incisor had a positive response to the cold test. Radiographic examination revealed a half-grown root with an open apex (Fig. 1A).

After 2 weeks, the splint was removed according to International Association of Dental Traumatology (IADT) guideline [1]. At 4 weeks of recall, the tooth still showed sensitivity to percussion with mild mobility. The radiolucency of the apical area appeared enlarged. However, the tooth responded to the cold test, although at low intensity (Fig. 1B). At 6 weeks of recall, the tooth still responded to the cold test with a low
intensity, and the sensitivity to percussion and palpation decreased. The radiolucency of the apical area also decreased and interestingly, an unknown radiopaque tissue was observed in the apical area (Fig. 1C, yellow arrow). Radiography at the 3-month follow-up visit showed increased radiopaque tissue with no apical radiolucency (Fig. 1D). The tooth had no specific signs and symptoms, with a positive and stable response to the cold test and no sensitivity to percussion and palpation. At the 6-month recall, no specific signs or symptoms were present on clinical examination. The radiopaque tissue in the apical area, which looked like a supernumerary tooth, became more distinct and the lower part of the main root canal appeared to be occupied with a bony structure (Fig. 1E). At the 1-year follow-up, the tooth showed a positive response to the cold test but a negative response to the electric pulp test. On radiography, no further lengthening of the main root was observed, and invagination of alveolar bone growth was detected of the periodontal ligament lining into the root canal (Fig. 1F). Clinically, the tooth showed normal mobility and sound on percussion, and signs of anklylosis were absent. The separated apical root tip showed a developmental stage similar to that of the right upper lateral incisor on Cone-beam computed tomography (CBCT) images (Fig. 1G). At the 17-month follow-up visit, root development seemed to be complete, showing apical closure in the apical root tip, accompanying positive response to cold test and negative to percussion (Fig. 1H). There was no discoloration when comparing the first and 17 months of recall visits (Fig. 1IJ).

2.2 Case 2

A 9-year-old girl with a noncontributory medical history was referred from a local clinic for evaluation and treatment of the right upper central incisor. According to the previous records from the local clinic, the patient had a laterally luxated injury on the right upper central incisor when she was 6 years old. The patient visited the local clinic a day after the traumatic injury, and the tooth was repositioned and splinted (Fig. 2A). One month later, the patient had spontaneous pain with gingival swelling and pus discharge from the sulcus of the tooth and underwent a one-visit apexification with a mineral trioxide aggregate (MTA) apical plug (Fig. 2B). However, even after the treatment, a sinus tract formed on the facial gingiva of the treated tooth (Fig. 2C, yellow arrow). Although the intervention was necessary to manage the inflammation, any subsequent treatment could not be performed owing to difficulty with patient compliance; thus, periodic follow-up was planned. Antibiotics were prescribed to subside the sinus and the splint was removed 2 months after the trauma. Three months after apexification, periapical radiolucency was observed around the apical area, with a more apically located radiopaque tissue (Fig. 2D, yellow arrow). At 3 years, the patient was old enough to undergo dental treatment and was referred to our clinic for further treatment. The patient did not report any subjective signs and symptoms for 3 years. On preoperative clinical examination, the right upper central incisor showed tenderness upon percussion and seemed slightly darker than adjacent teeth, however, it was not clear if it was MTA discoloration. Radiographic examination revealed periapical radiolucency with a separated root fragment partly away from the main root in the periapical radiographs and CBCT images (Fig. 2E–H). The tooth was diagnosed with previously treated tooth with symptomatic apical periodontitis. Considering the difficulty of accessing the apical area by orthograde approach, endodontic microsurgery was planned.

After removing the granulation tissue, hard tissue was detected in the bone cavity and taken out (Fig. 3A, white arrow). According to the protocol, root-end preparation and filling were performed using RetroMTA (BioMTA, Seoul, Korea) (Fig. 3B–D). The periapical radiograph at the 1-year recall showed complete healing with clear lamina dura around the root surface (Fig. 3E). With the hard tissue obtained during surgery, histological analysis with hematoxylin and eosin (H&E) staining was performed. It showed a typical root tip with pulp tissue in the canal with the foramen (Fig. 3F). In the magnified image, a normal root structure was observed with the pulp tissue, odontoblast layer, predentin and dentin with dentinal tubules (Fig. 3G).

2.3 Case 3

A 7-year-old girl was referred to the Department of Conservative Dentistry at Dankook University Dental Hospital by a local clinic. According to a letter from the clinic, the right upper central incisor was avulsed by hitting a wall 2 weeks ago and was splinted with resin wire after repositioning. Clinical examination revealed tenderness to percussion and no response to the cold test. Radiographic examination showed a two-third grown root with an open apex (Fig. 4A). The splint wire was removed according to IADT guideline [1], and periodic follow-up was planned.

After 6 weeks, external root resorption was observed at the mesial and distal surfaces (Fig. 4B, yellow arrowheads), and root canal treatment was initiated (Fig. 4C). When the access cavity was opened, active bleeding from the canal occurred. After copious irrigation with 2.5% sodium hypochlorite (NaOCl), calcium hydroxide (Ca(OH)2) (Ultradent® XSTM; Ultradent Products Inc., South Jordan, Utah, USA) was used as an intracanal medication. Two weeks after treatment, no further progression of the external root resorption was observed (Fig. 4D). After 3 months of intracanal dressing, no specific signs or symptoms were present; however, intracanal bleeding could not be obtained enough. Therefore, the root canal was obturated using MTA (ProRoot MTA; Dentsply Sirona, Charlotte, NC, USA). On postoperative radiography, an unknown radiopaque tissue was observed apically, partly away from the MTA filling (Fig. 4E, yellow arrow). At the 6-month follow-up after MTA apexification, the tooth remained asymptomatic, and periapical radiolucency was resolved (Fig. 4F). At the 1-year follow-up, an apical root tip that was partially connected to the main root and surrounded by the periodontal ligament was detected on radiography (Fig. 4G).

3. Literature review and discussion

Our three cases showed a fully developed apical root tip after traumatic injury with or without subsequent inflammation.
FIGURE 1. Serial radiographs and clinical photos of case 1. (A) The first radiograph of the splinted left upper lateral incisor. (B–E) Radiographs at the 4-week, 6-week, 3-month and 6-month follow-ups. Yellow arrows indicate the unknown radiopaque tissue around the apical area. (F,G) Radiograph and cone-beam computed tomography (CBCT) images at the 1-year follow-up. The arrowhead indicates the apical area of the separated root tip of the left upper lateral incisor. (H) A radiograph at the 17-month follow-up. (I,J) Clinical photos at the first visit and the 17-month follow-up.
In addition to our cases, we will review cases showing the completion of separate root tip development after traumatic injury or during inflammation. We searched Google Scholar and PubMed with the words “phantom root”, “separated root” or “detached root” for articles in English published from 1975 to 2022. Including our 3 cases, we found 27 cases from 18 reports and series and sorted them according to injury type (Tables 1 and 2). Eighteen anterior teeth suffered a traumatic injury, and eleven premolars had inflammatory injuries. Since the terminology for this phenomenon is not standardized by academics, we may not have retrieved all reported cases.

In the 18 cases of traumatic injury, the average age of the patients was 7.4 ± 1.7 years, which is related to the age at which traumatic injuries are prevalent [12]. Luxation injuries (13 cases), including avulsion (9 cases), were the most common trauma type. Among the 11 patients who underwent repositioning and splinting, pulp vitality was maintained without any further treatment in 4 patients. Three avulsed teeth were lost and healed in an edentulous state, and an apical root tip was newly generated in the edentulous alveolar bone area [13–15].

HERS and AP can function normally to form a root tip in traumatically injured teeth without pulpal inflammation. However, AP can be easily detached from the main root, which causes displacement of the newly generated root tip and influences integral root development. Including Case 1, several cases have reported separated growing root tips under the repositioned main root after avulsion [9, 13, 16].

In contrast, detachment of the AP may cause abnormalities in the development of the main root. In Case 1, the coronal part of the root arrested development with internal bone growth in the canal space. Interestingly, Im et al. [6] reported integral root formation with bony ingrowth after extrusive luxation. The lateral discontinuity of the root, suspected to be a portal of bony invasion, might be the result of partially detached HERS. Considering that HERS plays a role in preventing the ingrowth of the periapical tissue into the root and giving space for root dentin formation [5, 7], damaged HERS acts as an obstacle to typical root formation, in which the canal space is filled with pulp tissue and root dentin is integrally connected [8, 17].

Severe traumatic damage may be followed by complications such as pulp necrosis or root resorption, requiring intervention. In immature teeth, continuous root formation can occur even with apical periodontitis or abscesses with extensive bone resorption [10, 18–20]. Case 2 showed inflammatory signs and symptoms after 1 month, and Case 3 showed external root resorption after 6 weeks. After apexification to manage inflammation, apical root tips formed downward from the
FIGURE 3. Clinical photographs and radiographs and histologic images of case 2. (A) Hard tissue taken during surgery (arrow). Clinical photographs of (B) root preparation and (C) root-end filling using RetroMTA. (D,E) A postoperative radiograph (D) immediately and (E) 1 year after the surgery. (F) Haematoxylin and eosin (H&E) staining of the hard tissue of (A). (G) High magnification of dotted square of (F).
existing roots in both cases. In Case 2, the inflammatory condition persisted for more than 2 years, even after treatment. Nevertheless, the newly formed root tip showed a normal histological structure with pulp, the odontoblast layer, predentin and dentin, as determined by H&E staining (Fig. 3C). This is consistent with the findings of Palma et al. [19] who reported a histopathological assessment of the apical fragment [21].

Continuous root development under inflammatory conditions may be explained by the special features of AP. It is less susceptible to pathological events such as pulp necrosis and apical periodontitis [10, 18]. According to Driesen et al. [7], this is possible because it has a tissue structure with its own vascular network and innervation, which is encapsulated by a single-layered cuboidal epithelium. Huang et al. [22] also reported that stem cells in the AP may have survived despite the development of a periapical abscess because of the rich blood supply through the wide-open apex or their proximity to the periapical tissues. Regenerative endodontic procedures (REPs) for immature teeth with necrotic pulp are based on this.

We found nine cases that had an inflammatory condition without traumatic injury. As shown in Table 2, all cases involved premolars that frequently had dens evaginatus. Owing to the high possibility of cusp fractures in dens evaginatus, premolars tend to have pulp necrosis and apical periodontitis [23]. The average age of the patients was 11.2 ± 2.3, when premolars erupt and are still in immaturely developed state. The teeth showed various degrees of mobility, with apicification in two cases and REPs in seven cases. All cases except one showed completely detached root segments.

Interestingly, a separated root fragment existed away from the upper body of the root without traumatic movement. Palma et al. [24] suggested that infection or iatrogenic factors could also be responsible for AP dislodgement from the main root walls. Some studies have reported increased tooth mobility under inflammatory conditions [10, 18, 20], suggesting the possibility of AP detachment. However, a separated root tip at a distance was observed even when tooth mobility was normal [19, 21, 25]. As the inflammatory tissue around the root end grows, it pushes the surrounding periapical tissues such as AP and HERS away from the root body [26]. The first case reported by Jung et al. [10] showed the distance between the separated root tip and main root end increased immediately after treatment; however, the root tip moved toward the main root at the 3-year recall visit. Thus, resolution of the inflammation provided space for the displaced apical root tip to return to its original position.

Meanwhile, Yang et al. [21] suggested that the detachment or separation of stem cells from the AP (SCAP) and HERS from the main root might be due to apicification treatment. Jung et al. [10] recommended a conservative approach such as REP as the first choice of treatment. However, even in cases treated with REP, separated root tips have been observed [18, 25]. Additionally, three cases (Table 2) showed root tip development before endodontic treatment, which indicates no relationship between treatment type and the formation of a separated root [10, 19, 20].
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<th>Sex</th>
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*Root tip development*: separation state and detection time; Mn Bone Fx: mandibular bone fracture; PN: pulp necrosis; ERR: external root resorption; Repos & Fix: Repositioning and fixation; Apexi: Apexification; REP: Regenerative endodontic procedure; post tr: post-trauma; post op: post-operation; IC: intracanal calcification; m: months; yr: years; **: Extraction time; ***: Retreatment & Outcome of retreatment (Final recall); EMS: Endodontic microsurgery; M: Male; F: Female.
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<td>No</td>
<td>Survival (12 m)</td>
</tr>
<tr>
<td>2022</td>
<td>Tamanna et al.</td>
<td>20</td>
<td>15</td>
<td>M</td>
<td>DE</td>
<td>PN/SAP</td>
<td>0</td>
<td>REP</td>
<td>Separate, post op</td>
<td>Thickening</td>
<td>Survival (36 m)</td>
</tr>
</tbody>
</table>

Root tip development*: separation state and detection time; DE: Dens Evaginatus; PN: pulp necrosis; SAP: symptomatic apical periodontitis; CAA: chronic apical abscess; AAA: acute apical abscess; AAP: acute apical periodontitis; CAP: chronic apical periodontitis; Apexi: Apexification; REP: Regenerative endodontic procedures; post op: post-operation; pre op: pre-operation; IC: Intracanal calcification; PC: Partial calcification; m: months; **: Extraction time; M: Male; F: Female.
Seven cases of REPgs (Table 2) addressed successful outcomes, even with separated root tips. However, none of them showed ideal root development, including thickening and lengthening of root dentin [10, 18–21, 25]. Some cases showed root thickness, but no root lengthening. Considering the purpose of the bleeding induction, deeper movement of the file might be required to recruit remote mesenchymal stem cells into the canal.

4. Conclusions

Several clinical cases have demonstrated the high capability of AP, even under traumatic or inflammatory conditions. All patients showed continued root tip formation, even in a separate state. Understanding root development patterns and outcomes according to injury type, age of prevalence or treatment type could help with treatments such as regenerative endodontic procedures and trauma management for immature permanent teeth.

ABBREVIATIONS

AP, apical papilla; HERS, Hertwig’s epithelial root sheath; CBCT, Cone-beam computed tomography; MTA, mineral trioxide aggregate; REPgs, Regenerative endodontic procedures.

AVAILABILITY OF DATA AND MATERIALS

Not applicable.

AUTHOR CONTRIBUTIONS

YK and MS—designed the study. YK and MK—summarized and analyzed the case series. MK and MS—searched and analyzed the literature. MS—provided help and advice on the study. MK, YK and MS—wrote the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This case report has been written according to Preferred Reporting Items for Case reports in Endodontics (PRICE) 2020 guidelines. All cases were evaluated and treated after taking informed consent from the patients’ parents or guardians. Oral informed consent was obtained from the patients’ parents or guardians for publication of this case report. The clinical data of the patients were searched by the Department of Conservative Dentistry at the Dental College, Dankook University (DKUDH IRB 2022-04-003).

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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