INTRODUCTION

Dental abnormalities such as fusion or gemination have been described in both dentitions. Fused teeth correspond to the union of two or three normal tooth germs (synodontia), or one or two normal tooth germs and one supernumerary tooth. Depending on when it happens during tooth development, the fusion can be complete or incomplete; the pulp chamber and root canal may be joined or separated. When fusion occurs after crown completion, the teeth are united only by the cementum; this is called concrescence. Gemination is the failure of attempted tooth-germ cleavage with incomplete formation of two teeth, usually with one pulp chamber, a single root, and a common pulp canal. Twinning represents complete formation of two nearly identical teeth, one normal and one supernumerary tooth, but fused as one, usually with a single root and a single pulp canal. In the case of three tooth entities, the terms “triple tooth,” “triple teeth,” or

FIGURE 1  Intraoral view of the maxillary triple teeth and the gingival laceration
“triplication” are used and have the advantage of covering all types of unions. In fact, the clinical aspect of fusion or gemination of normal or supernumerary teeth is similar.

The prevalence of triple teeth in primary dentition is rare (0.02%) and shows a predilection for the male sex and Asian populations. It occurs more frequently in the upper arch than in the lower arch. A number of etiological hypothesis have been suggested: Close developing tooth buds, insufficient space in the dental arch, and physical pressure or trauma can cause contact between tooth germs that results in necrosis of the epithelial tissue that separates tooth germs and leads to fusion; genetic factors (dominant autosomal heredity); and disturbances in the prenatal period or environmental factors.
such as viral infection during pregnancy, intake of thalidomide, and lack of vitamins. However, none of them proved satisfactory.

Shilpa and Nuvvula classified triple teeth into two types and subtypes. Type I corresponds to fusion with three pulp chambers and three root canals, which includes type Ia: fusion of two normal teeth with a supernumerary tooth and type Ib: fusion of three normal teeth. Type II corresponds to fusion with two pulp chambers and two root canals, which includes type IIa: combination of one geminated tooth and a supernumerary tooth, and type IIb: combination of one geminated tooth and a normal tooth. However, it is difficult to determine the type even with intraoral radiography.

This paper aimed to report a rare case of a young patient presenting large triple teeth in the right maxillary incisor region and double teeth in the left mandibular incisal region, which has not been reported in the literature before, and further discuss it by a comprehensive literature search on triple teeth.

2 | CASE PRESENTATION

A 3-year-old male patient of Cambodian origin was referred for extraction of an unusual right maxillary incisor after a traumatic injury. According to his mother, there were no such anomalies in the other family members. The anamnesis revealed that the child was born premature and actually presented with delayed growth, hyperlaxity, and cerebellar atrophy. Moreover, his built was small for his age. These symptoms led to genetic exploration of syndromic diseases, but with no remarkable findings.

The extraoral examination did not show any alterations. Intraoral examination revealed a gingival laceration next to a decayed triple tooth, in which was a double crown in place of the maxillary central incisor fused with the lateral incisor (Figure 1). These triple teeth were affected by a large carious lesion at the junction between the double crown and an incipient carious lesion in the groove between the double crown and the lateral incisor crown. These teeth presented no mobility or fracture. In the region of the right central incisor, a submucosal abscess was observed, suggesting infected pulp necrosis. Moreover, the left mandibular central and lateral incisors appeared fused, with a unique but larger crown (Figure 2). No other findings were reported.

An intraoral periapical radiograph of the triple teeth revealed two distinct structures, an upper right incisor with a possibly unique pulp chamber (difficult to confirm because of the carious lesion) and a large root canal (possibly dividing into two canals in the middle-third), and a lateral incisor with separate pulp chamber and separate root canal (Figure 3). It also showed a radiolucent area around the apex of the large central incisor, but the lateral incisor seemed unaffected. It was not possible to take a radiograph of the double teeth due to noncompliance of the patient.

The diagnosis was gingival laceration due to trauma and abscess on the triple teeth due to an advanced carious lesion. Because of the difficulty in performing root canal treatment in such teeth and the poor cooperation of the young patient, extraction of the triple teeth was planned. Due to the difficulty in extracting such teeth and for the patient's comfort, this procedure was performed under nitrous oxide/oxygen inhalation. To prevent functional, esthetic, and phonetic problems, the missing teeth should have been replaced with a transitional partial denture. However, the replacement was not possible at this stage due to insufficient cooperation by the patient.

A 1-year follow-up showed good healing (Figure 4). It was however not possible to perform a radiograph to check the underlying tooth germs, because of insufficient cooperation.

The extracted teeth had three separate crowns and roots conjoined from the crown to the apex and possibly from the incisal edge to the apex in the large central incisor (Figure 5). Macroscopically, almost no root resorption was detected. The teeth were analyzed using micro-computed tomography to obtain a three-dimensional model and a two-dimensional cross-sectional slice (Figure 6). It showed two separate pulp chambers in the large central incisor, which were joined together at the cervical area, and one root canal. The lateral incisor presented separate pulp chamber and root canal, but its root canal was connected to the root canal of the large central incisor. These observations suggested gemination of the central incisor fused with the lateral incisor (type IIb of the classification by Shilpa and Nuvvula).

3 | DISCUSSION

To our knowledge, coexistence of triple teeth and double teeth in two different arches has never been reported. To address this issue, an extensive literature review was conducted to identify all reported cases of triple teeth in the English language. A search was performed on the PubMed database using keywords related to triple teeth and primary dentition according to the following equation search: (triplication OR "triple teeth" OR "triple tooth" OR "three teeth" OR tripled OR "three tooth fusion") AND (primary OR decidual OR deciduous OR temporary). It was manually completed with the references of the selected articles and a search on Google Scholar in order to find nonindexed publications. Inclusion criteria were all types of articles reporting or discussing triple teeth in primary dentition, written in English language, published up to December 2019, and availability of the full text. A table was created to collect details regarding age and sex of patients, teeth implicated, patient origin, familial history, medical context, diagnostic tools, radiographic interpretation, resorption trouble/delay of eruption, absence
| Author, date                     | Patient’s age and sex | Teeth implicated | Origin          | Familial history | Medical context | Diagnostic tools                                                                 |
|---------------------------------|-----------------------|-----------------|-----------------|-----------------|----------------|----------------------------------------------------------------------------------|
| Gultekin et al10                 | 5-y-old female        | 81, 82, sny     | Probably Turkish| None            | Noncontributory | Periapical radiograph, panoramic radiograph, CBCT                                  |
| Jeong et al11                    | Case 1: 1-y, 7-mo-old male   | Case 1: 51, spy, 52 Case 2: 81, 81, sny     | Case 1: probably Korean Case 2: probably Korean | Not mentioned | Not mentioned | Case 1 and Case 2: Intraoral periapical radiography                                  |
| Malikarjun et al12               | 9-y-old male          | 72, sny, 73     | Probably Indian | Noncontributory | Noncontributory | Periapical radiography, postavulsion histological examination with stereomicroscope |
| Nagaveni et al13                 | 7-y-old male          | 81, 82, 83 and 71, 72 | Probably Indian | Noncontributory | Premature birth | Intraoral periapical radiography                                                   |
| Thakkar et al14                  | Case 1: 5-y-old male   | Case 1: 61, 62, sny Case 2: 51, 52, sny      | Probably Indian | Not mentioned | Not mentioned | Case 1: Intraoral periapical radiography, CBCT Case 2: Intraoral periapical radiography |
| Shanthraj et al15                | 7.5-y-old female      | 71, sny, 72     | Probably Indian | Not mentioned | Not mentioned | Intraoral periapical radiography                                                   |
| Juneja et al16                   | 9-y-old male          | 61, sny, 62     | Probably Indian | Noncontributory | Noncontributory | Periapical and occlusal radiography, postavulsion radiography, histological examination |
| Yadav et al17                    | 10-y-old male         | 51, sny, 52     | Probably Indian | None            | Mentally challenged | Periapical radiography, histological examination with stereomicroscope            |
| Shilpa et al9                    | 5-y-old male          | 61, 62, sny     | Probably Indian | Noncontributory | Noncontributory | Occlusal radiograph                                                              |
| Sharma et al18                   | 7-y-old male          | 61, 62, sny     | Probably Indian | Noncontributory | Noncontributory | Intraoral periapical radiograph, histological examination                         |
| Babaji et al19                   | 6-y-old male          | 81, sny, 82     | Probably Indian | Noncontributory | Noncontributory | Intraoral periapical radiograph                                                   |
| Radiography interpretation | Resorption troubles/delay of eruption | Absence of successional tooth | Other dental anomalies | Treatment performed |
|----------------------------|--------------------------------------|-------------------------------|------------------------|---------------------|
| Fusion of 81 and 82 with a sny tooth (three separate pulp chambers joining in one pulp canal) | None | None | None | Regular follow-up until normal exfoliation |
| Case 1: Fusion of 51 and 52 with a sny tooth (two separate canals and pulp chambers for 51 and 52, sny tooth between them with root in developmental state) | None | Case 1: Agenesis of 12 | Case 1: None | Case 1: Pulpotomy, Case 2: Extraction |
| Case 2: Pulp chamber structure was obvious, but a canal structure was not clear for the sny tooth. The two primary teeth had clear canals | | | | |
| Fusion of 72 and 73 with a sny tooth (gemination of 72 fused with 73) | Slow root resorption, 31, 32 already erupted | None | 22 and 12 in labial version | Extraction |
| Fusion of 81,82 and 83 and fusion of 71 and 72 (separate pulp chamber and root canals in both) | None | Agenesis of 42 | None | Regular follow-up until normal exfoliation |
| Case 1: Fusion of 61 and 62 with a sny tooth (three distinct crowns, one root with two equidistant grooves, but distinct root canals) | None | None | None | Case 1: Extraction (abscess) + transitional partial denture, Case 2: Restorative treatment (decay) |
| Case 2: Fusion of 51 and 52 with a sny tooth (separate pulp chambers and root canals) | | | | |
| Fusion of 71 and 72 with a sny tooth (all along the crown and the root: each tooth with distinct pulp chambers and root canals) | Yes, but resorption at 3 mo follow-up | None | None | No treatment, root resorption until exfoliation |
| Fusion of 61 and 62 with a sny tooth (three separate pulp chambers and root canals) | Slow root resorption, 21 palatally erupted | None | 21 palatally erupted, in Crossbite, 22 unerupted with space deficiency | Extraction, composite inclined plane on opposing mandibular teeth to correct the crossbite |
| Fusion of 51 and 52 with a sny tooth (separate pulp chambers, separate root canals merging in the apical third) | Resorption only of the 52 (presence of 11) | Agenesis of 12 | 11 palatally erupted, in crossbite | Extraction (tooth retained) |
| Fusion of 61 and 62 a with sny tooth | None | Agenesis 22 | None | Regular follow-up |
| Concrescence of 61 and 62 with a sny tooth (separate pulp chamber and root, union by cementsal part) | Resorption only of the 61 (presence of 21) | None | 21 palatally erupted | Extraction (tooth retained, palatal eruption of 21) |
| Fusion of 81 and 82 with a sny tooth (separate pulp chamber and root canals) | Eruption delay (presence of 31 and 32) | Agenesis of 41 | None | Recall examination until exfoliation |
(Continues)
| Author, date | Patient’s age and sex | Teeth implicated | Origin | Familial history | Medical context | Diagnostic tools |
|-------------|-----------------------|-----------------|--------|-----------------|----------------|-----------------|
| Mohapatra et al<sup>20</sup> | 10-y-old male | 51, sny, 52 | Probably Indian | Not mentioned | Noncontributory | Intraoral periapical and occlusal radiograph, histological examination with stereomicroscope |
| Schultz-Weidner et al<sup>21</sup> | 4-y-old male | 51, sny, 52 and 61, sny, 62 | Thai | None | Noncontributory | Intraoral periapical radiography |
| Prabhakar et al<sup>22</sup> | 6-y-old male | 51, sny, 52 | Probably Indian | Not mentioned | Not mentioned | Intraoral periapical radiography, histological examination |
| Erdem et al<sup>23</sup> | 2-y-old male | 61, sny, 62 | Probably Turkish | Not mentioned | Noncontributory | Intraoral periapical radiography, light microscope |
| Aguiló et al<sup>24</sup> | Case 1: 3-y-old female; Case 2: 2-y-old male | Case 1: 61, sny, 62; Case 2: 51, sny, 52 | Caucasian | None | Noncontributory | Intraoral periapical radiography, post-avulsion radiography, histological examination, CT images |
| Rao<sup>25</sup> | 6-y-old female | 61, 62, sny | Probably Indian | Father with similar teeth | None | Intraoral periapical radiography, orthopantomogram |
| Mochizuki et al<sup>26</sup> | 2-y., 8-mo-old female | 52, 51, 61 | Japanese | None | None | Orthopantomogram, intraoral occlusal radiography |
| Riesenberger et al<sup>27</sup> | 2-y., 8-mo-old male | 61, 62, sny and 51, 52 | Not mentioned | Father with “double baby teeth” | Asthma | Intraoral occlusal radiography |
| Trubman et al<sup>28</sup> | 3-y-old male 6-y-old male | Case 1: 81, 82, sny; Case 2: 81, 82, sny | Case 1: “black”; Case 2: “white” | Not obtainable | None | Intraoral periapical radiography |
| Knapp et al<sup>1</sup> | 6.5-y-old female | 61, 62 and sny | “White” | None | None | Intraoral occlusal and periapical radiography, post-avulsion radiography |
| Dhoria et al<sup>29</sup> | 10-y-old male | 61, 62, sny | Probably Indian | None | None | Intraoral periapical radiography, postavulsion radiography |
| Radiography interpretation                                                                 | Resorption troubles/ delay of eruption     | Absence of successional tooth          | Other dental anomalies                        | Treatment performed                                                                 |
|------------------------------------------------------------------------------------------|-------------------------------------------|----------------------------------------|-----------------------------------------------|------------------------------------------------------------------------------------|
| Fusion and gemination (three separate crowns with separate pulp chambers at the crown, three joined roots with separate pulp canals at the middle third, progressively joined to form a common apical canal, dentinal fusion) | Eruption delay (presence of 21)            | Agenesis of 12                          | Midline diastema                                                                 | Extraction (permanent retained), removal prosthesis                                 |
| Fusion of 51 and 52 with a sny tooth; fusion of 61 and 62 with a sny tooth (both with separate pulp chambers and root canals) | No root resorption                        | None                                    | None                                           | Opening chamber, then extraction, and transitional partial denture                    |
| Fusion of 61 and 62 with a sny tooth (separate pulp chambers and root canals, fusion at enamel and cementum, only) | None                                       | None                                    | None                                           | Extraction, then removal of prosthesis                                               |
| Fusion of 61 and 62 with a sny tooth (separate pulp chambers and root canals, fusion by dentin and cement) | None                                       | Agenesis of 22                          | None                                           | Extraction (abscess) + transitional partial denture                                  |
| Case 1: Fusion of 61 and 62 with a sny tooth (separate pulp chambers and root canals, fused in apical) Case 2: Fusion of 51 and 52 with a sny tooth (separate pulp chamber and root canals, fused in the middle for two, then separate again) | Case 1: None (physiological resorption)   | Case 1: None                            | None                                           | Extraction (Case 1: trauma and Case 2: abscess)                                      |
| Fusion of 61 and 62 with a sny tooth (separate pulp chambers and root canals)              | None (physiological resorption)            | Agensis of 22                           | Diastema in maxillary and mandibular arch      | Restoration of decay teeth and seal the deep grooves                                |
| Fusion of 52, 51, and 61 (separate pulp chambers, root canal fused ¼th of the way from apex, one root) | Not mentioned                              | Underdeveloped 12, fused with 11       | Width and length of dental arch less than the Japan national average | Sealant therapy, fluoride application, and monitoring                               |
| Fusion of 61 and 62 with a sny and fusion of 51 with 52                                    | None                                       | None                                    | None                                           | Extraction of 61, 62, and the sny tooth (abscess); Extraction of 52 and restoration of 51 |
| Case 1: Gemination 81, fusion 82 Case 2: Gemination 81, fusion 82 (Both cases: 81 with one root canal and two crowns, 82 with one crown and one root canal) | None                                       | Case 1: None                            | None                                           | Monitoring                                                                         |
| Gemination of 61 or early fusion with sny tooth, and fusion with 62 (central and mesial elements with shared pulp chamber and separate root canal, distal element with distinct pulp chamber and root canal) | Eruption delay                             | Lower development rate of 22 as compared to 12 | None                                           | Monitoring, followed by extraction (tooth retained) and space maintainer             |
| Fusion of 61 and 62 with a sny tooth (separate pulp chambers and root canals)             | Slow root resorption, delay of resorption | None                                    | Successor teeth already erupted (21 and 22)    | Extraction                                                                         |

(Continues)
TABLE 1  (Continued)

| Author, date | Patient’s age and sex | Teeth implicated | Origin | Familial history | Medical context | Diagnostic tools |
|--------------|-----------------------|-----------------|--------|----------------|----------------|----------------|
| Burley et al\textsuperscript{30} | Case 1: 4-y-, 10-mo-old female | Case 1: 61, 62, sny | Not mentioned | Sister and brother with similar dental features | None | Case 1: intraoral periapical radiography |
| | Case 2: 2-y-, 11-mo-old male | Case 2: 61 62 | | | | Case 2: intraoral occlusal radiography |
| Long\textsuperscript{31} | 7-y-old male | 71, 72, sny | Not mentioned | Not mentioned | Not mentioned | Post-avulsion radiography |

Abbreviations: CBCT, cone beam computed tomography; CT, computed tomography; mo, months; sny, supernumerary; y, years.

Most studies only use intraoral periapical or occlusal radiographs, limiting the distinction between fusion and gemination as well as the relationship and proximity between the triple teeth and adjacent and underlying teeth. Panoramic radiographs help in better examination of the entire dental situation, especially to detect potential agenesis of underlying permanent teeth. In fact, 10 cases\textsuperscript{9,11,13,17,19,20,23-25,31} reported missing successional teeth and one case reported presence of two mesiodens.\textsuperscript{11} The cone beam computed tomography (CBCT) avoids image distortions and superimpositions, allowing easy observation of root canal and precise determination of resorption areas. However, it is difficult to perform panoramic radiography in very young patients and CBCT is irradiating. Only three cases underwent panoramic radiography,\textsuperscript{10,25,26} while CBCT was used in two cases.\textsuperscript{10,14}

Furthermore, several cases reported crossbite or malalignment,\textsuperscript{12,16-18,29,31} underlying the importance to monitor tooth resorption and its timely exfoliation.

In most cases, monitoring was implemented\textsuperscript{3,9,10,13,15,19,26,28} or extraction was indicated,\textsuperscript{11,12,14,16-18,20,22-24,27,29-31} as in our case. Only three cases had undergone restoration procedure\textsuperscript{11,25,27} and one case underwent pulpotomy.\textsuperscript{11} Because of the complexity of the root canal system, reliable root canal treatment is almost impossible; thus, all efforts should be made to avoid carious lesions. Sealants should be placed in the grooves of the occlusal surfaces, followed by regular monitoring. Only two cases reported sealing of the grooves,\textsuperscript{25,26} whereas 10 cases were monitored. In cases of deep pulpal involvement or periapical lesions, extraction is inevitable. In cases of delayed exfoliation, extraction is also recommended to avert malocclusion.

4  |  CONCLUSION

Fused teeth are initially asymptomatic and rarely seen in children. Aside from esthetic concerns, they can develop carious
lesions in their grooves, pulpal inflammation, or abscesses. Root canal therapy is not a reliable treatment. Fused teeth could also lead to delayed exfoliation, resulting in space problems, occlusal disturbances, and delayed eruption of the permanent successors. Therefore, early identification is crucial to implement preventive/simple restorative treatment followed by careful monitoring until exfoliation. In cases of delayed physiological root resorption, extraction at the age of normal exfoliation should be implemented to prevent late eruption of the permanent teeth.

CONFLICT OF INTEREST
None declared.

AUTHOR CONTRIBUTIONS
ML: treated the patient, conducted the literature search, and wrote the manuscript. AB: performed the 3D analysis and gave final approval. ND and MT: treated the patient and gave final approval. ED: directed the work, analyzed the literature search, and wrote the manuscript.

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