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CASE REPORT

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# Cone-beam computed tomography in the diagnosis of regional odontodysplasia

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**Abstract**

Regional odontodysplasia (RO) in permanent teeth is a rare developmental anomaly of mineralized tissues. Three-dimensional images and data from CBCT allowed to provide useful information on the degree of tooth calcification and consequently confirm the diagnosis of RO and establish a treatment strategy to minimize future damages and sequels.

**KEYWORDS**

cone-beam computed tomography, permanent maxillary incisors, primary teeth, regional odontodysplasia, sequelae of trauma

## 1 | INTRODUCTION

Regional odontodysplasia (RO), also known as "localized arrest tooth development" and "ghost teeth," is an unusual, non-hereditary anomaly of the dental hard tissues that usually affect the anterior maxilla and female patients.[1,2] The term "odontodysplasia" was first introduced by Zegarelli et al. in 1963 to describe an unusual dental development disorder that affects both permanent and deciduous dentitions. Since this condition tends to affect only one quadrant, Pindborg et al. added the prefix "regional".[2-4]

Only a few numbers of studies about RO have been published. The literature search via PubMed using the keywords ("Cone-beam computed tomography" OR "CBCT") AND "regional odontodysplasia" included only a few articles which highlight the importance of the current study.[2,5]

The etiology of RO remains uncertain, with many possible causes, namely local trauma, somatic mutation of the dental lamina, viral infection, medication taken during pregnancy, local circulating disorder like vascular nevi on the skin of the affected side of the face, failure of migration, and differentiation of neural crest cells. Furthermore, there are case reports of RO with epidermal nevus syndrome, hypoplasia of the affected side of the face, hypophosphatasia, hydrocephalus and mental retardation, and ipsilateral vascular nevi.[2,6,7]

Diagnosis of RO is mainly based on the clinical and radiographic findings, supported by histologic features. Teeth affected by RO show clinical and radiograph aspects useful as diagnostic criteria.[8,9]

Clinically, affected teeth are small, discolored (yellow or brown), friable, hypoplastic, hypocalcified, have an altered morphology and an irregular surface contour.[2,8,10,11]

Radiography shows a "ghostlike" appearance characteristic of RO, due to the lack of distinction between the enamel and dentin, both are less radiopaque than sound teeth. Additionally, enamel and dentin layers are thin, pulp chambers are wide, apices are open and roots are significantly shortened.[2,8,10,11]

Although several cases have been reported, there are only two published cases evaluating RO on CBCT.

Through this case, it has been demonstrated that radiographic analysis through CBCT is essential to provide an early diagnosis of RO, to minimize future damages and sequels and allow noninvasive treatment.[2,7]

The present report aimed to describe unusual radiographic findings observed on conventional and CBCT from a 6-year-old male patient with maxillary RO with history of intrusive trauma at an early age.

## 2 | CASE PRESENTATION

The present case report was prepared according to the CARE Guidelines.[12]

A 6-year-old male patient visited the Department of Pediatric Dentistry, with a chief complaint of noneruption of upper front permanent teeth.

Written informed consent was obtained from the child's parents for all imaging examinations, treatment modalities, and data publications.

The medical history of the patient showed no specific problems.

During the anamnesis, patient's father gave a history of trauma to the upper front teeth at the age of 8 months which resulted in a complete intrusion of primary maxillary central incisors 51, 52 (fully intruded). The patient had fallen down the stairs from approximatively 3 meters.

Then, he consulted a local dentist who kept him on wait and see.

No radiographic examination was carried out at that time and no other symptoms were present.

Intraoral examination at the first appointment, revealed that the maxillary right primary incisors (tooth 51 and 52), and the right primary canine (tooth 53) were absent and the alveolar ridge of the affected region was covered with normal colored mucosa (Figure 1).

A periapical radiograph of the permanent maxillary incisors (tooth 11 and tooth 21) revealed a remarkably reduced radiolucency and the demarcation between enamel and dentin was not visible (Figure 2).

Panoramic radiograph (Soredex SCANORA® 3Dx Country, Germany) at the first visit confirmed the difference between teeth 21, 11, 12 and the unaffected teeth regarding radio density and showed that the germ of the right canine (tooth13) was absent radiologically.

Teeth 12, 11, and 21 were small, enamel, and dentin layers were thin, giving the affected teeth a "germlike" appearance while the other teeth were present and normal (Figure 3).

A CBCT scan (Soredex SCANORA® 3Dx, Germany) was performed 6 months after the consultation's day in three dimensions; axial, sagittal, cross-sectional and panoramic images, with a 1 mm slice thickness. The exposure parameters were as follows: field of view (FOV) = Medium (80 × 100), Peak kilovoltage (kVp) = 90, and Milliamperage (mA) = 13.

The CBCT analysis showed that the affected teeth[11-13] were less radiopaque than the unaffected counterpart

**FIGURE 1** Intraoral view showing lack of tooth 51, 52, and 53 and normal appearance of the edentulous quadrant


**FIGURE 2** Periapical radiographs showing a "germ-like" appearance of tooth 11 and 21

tooth 22 with shortened roots and wide-open apical foramen and remarkable differences in the pulp chamber space were also reported (Figures 4-6).

The CBCT highlighted also that the affected teeth were surrounded by large hypodense areas suggestive of enlargement of the pericoronal follicle.

To provide a further volumetric comparison of the affected and the non-affected contralateral tooth 22, the zoom 3D tool of the OnDemand3D™ software (Cybermed Inc, USA) was used (Figures 7 and 8).

Three-dimensional and sagittal CT revealed that the germs of teeth 21,11, and 12 were in Nolla's mineralization stage 4 (Figures 4-7) compared to Nolla's stage 7 of tooth 22 (Figure 8).

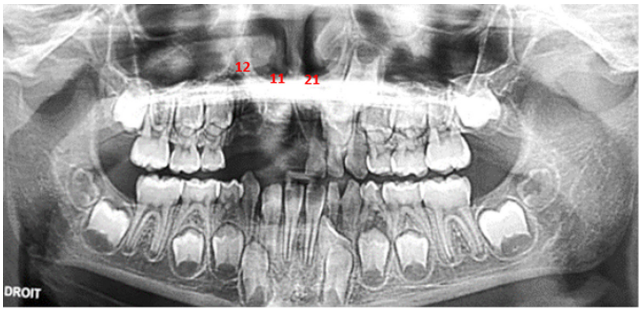


**FIGURE 3** Orthopantomography showing the appearance of "Ghost teeth" contrasts (tooth 21, tooth 11, and tooth 12) with a normal aspect of the unaffected teeth

Based on the above-mentioned clinical and radiographic findings, age and dental history of the patient, the diagnosis of RO of upper permanent incisors was confirmed.

At this stage, the treatment was to re-establish the esthetics, speech, and swallowing by making a functional space maintainer using a removable partial denture.

A second orthopantomography and a periapical radiograph were requested 18 months after the first visit and revealed no visible changes in the degree of germs calcification in the affected incisors and only an insignificant amount of root formation since the initial imaging examinations were noticed (Figures 9 and 10).

Follow-up appointments were scheduled every six months to control the eruption of the affected teeth and to readjust the removable space maintainer according to the patient's maxillary arch growth.

During the last appointment (2 years after the first visit), a laser-assisted labial frenectomy was performed and the appliance has been changed for a new one (Figure 11).

## 3 | DISCUSSION

Regional odontodysplasiais a rare non-hereditary developmental anomaly of the dental tissues derived from both the mesoderm and ectoderm.[10,14]

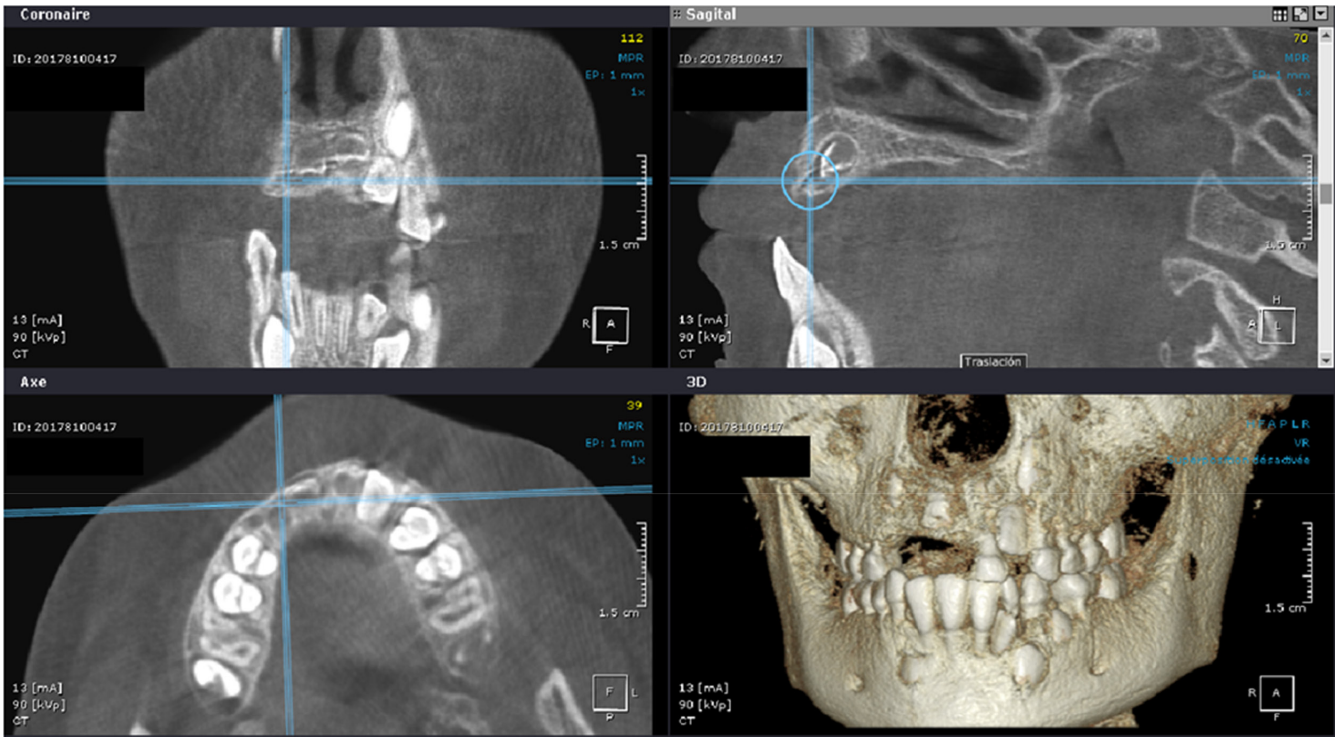


**FIGURE 4** Multiplanar reformatted (MPR), panoramic Volume Rendering (VR) views through tooth 12 (Coronal, sagittal, axial and 3-dimensional view)

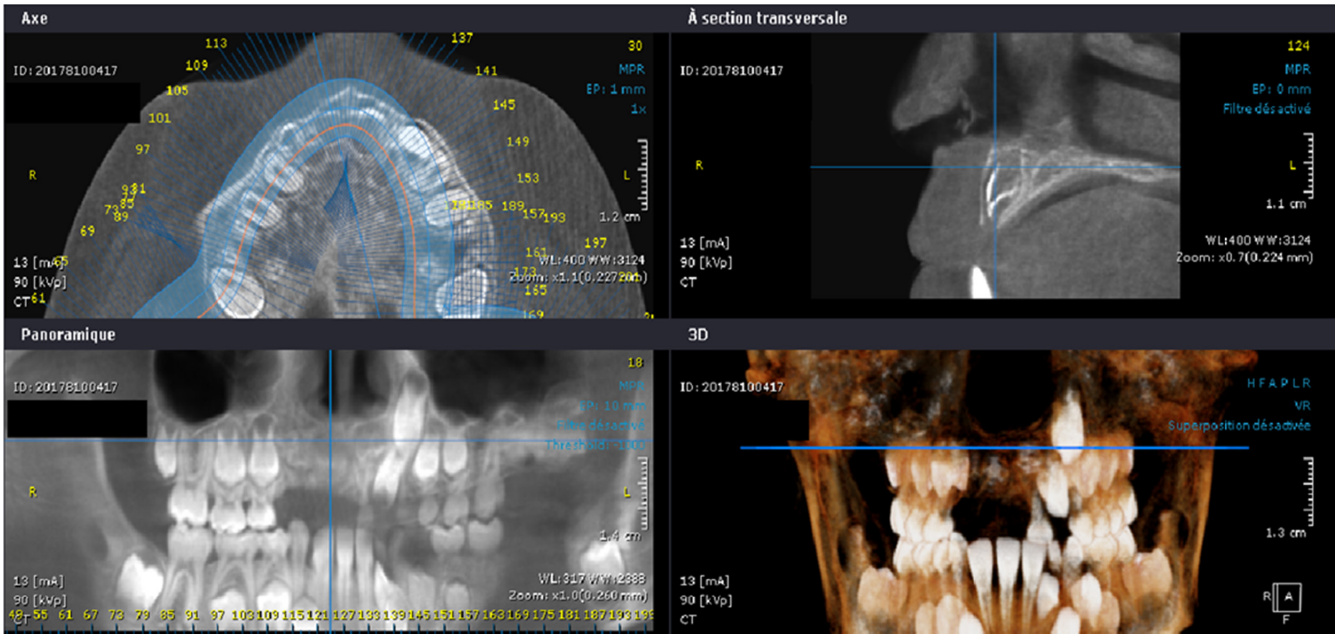


**FIGURE 5** Multiplanar reformatted (MPR), panoramic Volume Rendering (VR) views through tooth 11 (Axial transverse panoramic and 3-dimensional view are shown)

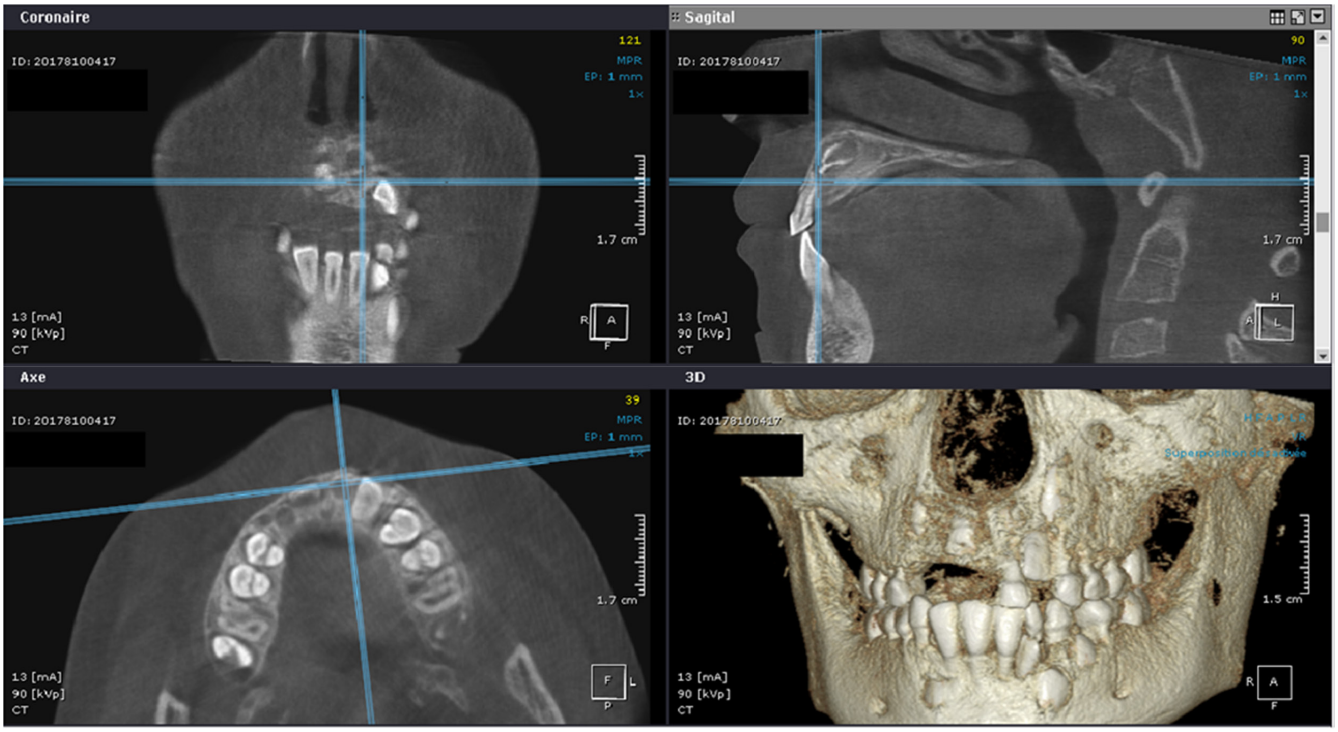


**FIGURE 6** Multiplanar reformatted (MPR) and volume rendered (VR) images through tooth 21. Coronal view through tooth 21(top left). Axial view (bottom left). Cross-sectional view (top right) and 3-dimensional representation (bottom right)

Although several theories have been proposed, the etiology of RO remains uncertain. However, a history of trauma has been suggested among the causative factors, besides Van der Waal et al[15] affirmed that a history of trauma with the involvement of more than two teeth argues with a diagnosis of RO.[4]

In the present report, according to the patient's father the intrusive injury occurred at the age of 8 months and four teeth were affected (tooth; 13, 12, 11, 21) which may explain the occurrence of such lesion. Nevertheless, the history of trauma may be coincidental association.

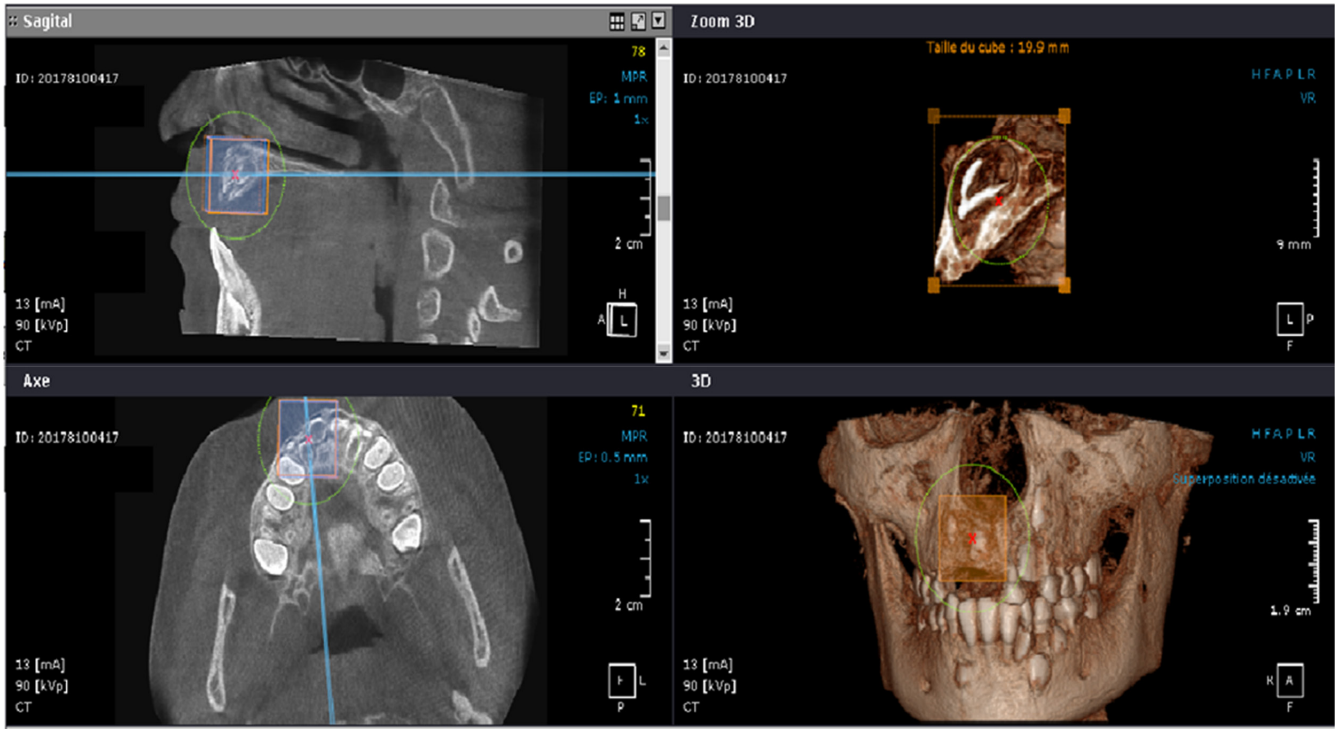


FIGURE 7 3D zoom tool (Ondemand3DApp program) showing a clear image of the RO tooth 12 which resembled Nolla's stage 4

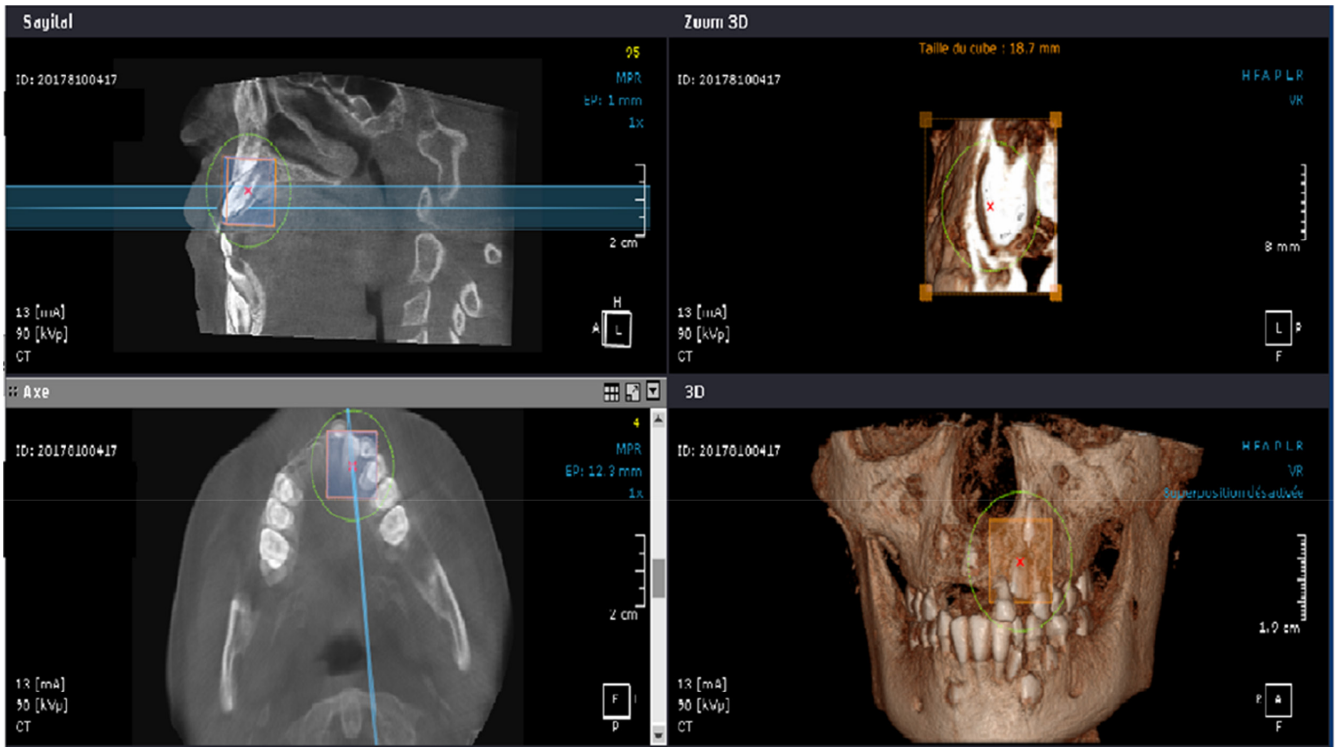


FIGURE 8 3D zoom tool (Ondemand3DApp program) showing a clear image of the contralateral sound tooth 22 which resembled Nolla's stage 7

This developmental dental disorder rarely affects more than one quadrant with a predilection to female patients (1.7/1). [2,16]

RO most often affects incisors and canines which was confirmed in the present report.[17]

In the field of oral radiology, imaging findings are strong diagnostic criteria for RO.[2,8]

Imaging examinations in this clinical case showed three "germlike" radiopacities in the maxilla crossing the midline. A lack of contrast between the enamel dentin,

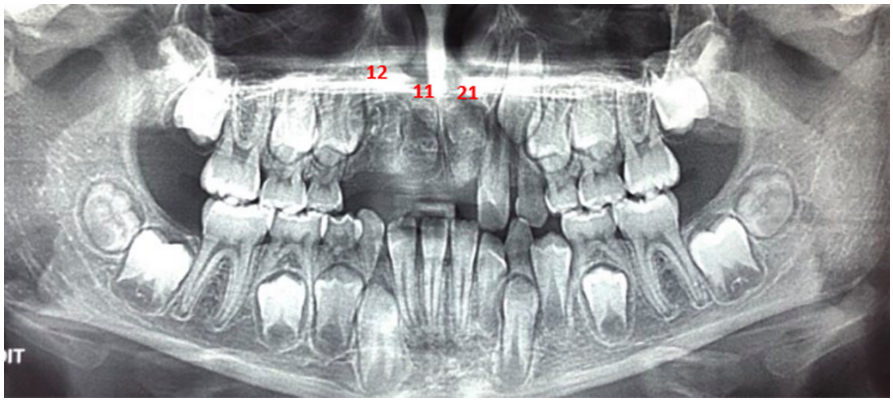


**FIGURE 9** Orthopantomography showing no visible changes in the degree of germs calcification in the affected incisors with only an insignificant amount of root formation after 18 months follow-up

less radiopaque than unaffected counterparts, was also reported.

Enamel and dentinlayers were thin, and an insignificant amount of root formation with thin dentinal walls giving the affected teeth a "ghost-like" appearance was noticed.

The pulp chambers were wide with open apices and enlarged follicles.[2,8,10,11]

The radiographic characteristics involving the permanent incisors in the maxillary right quadrant reported in this case strongly supported the diagnosis of RO.

In the current case, a rare radiographic finding which was the absence of right permanent canine (tooth 13) was described. Similarly, Cavalcante et al[2] reported the absence of right permanent mandibular central and lateral incisors in a mandibular RO case. However, a congenitally missing tooth cannot be excluded.

Accordingly, Tervonen et al[2,16] conducted an epidemiological study of 138 cases and reported only a rate of 10.7% missing teeth.

Another interesting finding noticed through sectional images and confirmed using the 3D zoom tool of the OnDemand3D™ software was the radiological aspect of the germ of tooth 12 which resembled Nolla's mineralization stage 4 while the non-affected contralateral (tooth 22) matched Nolla's stage 7.[18]

The OnDemand3D™ software is a highly-advanced 3D imaging software developed for dentists and research experts. Through the latest and best in 3D technology, this software uses DICOM data across modalities to reconstruct 3D volumes providing reconstructed images and tools for accurate and precise diagnoses.[19]

Computed tomography becomes a highly recommended examination in RO diagnosis, since conventional radiographs, corresponding to two-dimensional images of three-dimensional structures, have some restrictions in terms of imaging details and three-dimensional evaluation.[2,5]

In RO patients, CBCT enables cross-sectional, panoramic, axial–coronal–sagittal and 3D image reformat

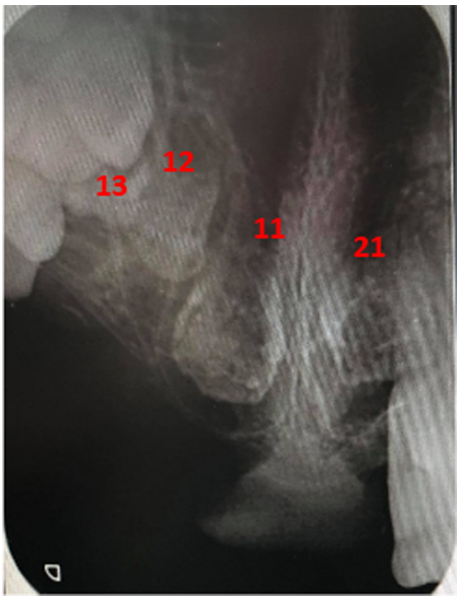


**FIGURE 10** Periapical radiograph using dental RVG after 18 months follow-up

allowing simultaneous assessment of tissue density, the volume of the pulp chamber, and the mineralization stages.[2]

In the present report, the CBCT confirmed the diagnosis of RO, thereby improving treatment planning.

To the best of our knowledge, none of RO case reports has described this anomaly following trauma in early childhood and only two reports have described the CBCT imaging findings.[2,7]

Although CBCT images were better than the conventional radiographs and allowed the diagnosis of this dental disorder, for follow-up, the CBCT images should not replace the conventional imaging methods.[20]

To monitor the development, the eruption, and the mineralization of the affected teeth, a recent orthopantomography was requested 18 months after the first visit. This orthopantomography revealed that the degree of the dental germs calcification in the affected incisors had not

**FIGURE 11** Intraoral views after labial frenectomy and esthetic rehabilitation of the patient (2 years follow-up)

evolved and only an insignificant amount of root formation occurred.

Accordingly, the literature reported delayed eruption of RO teeth and a failure in the dental eruption process.[10,13,17,21]

To confirm the etiological factors, further histological analysis of the affected teeth must be done, but at this stage, in the present report, conservative oral rehabilitation was followed.[10]

Regarding the therapeutic approach of RO, the main question was whether to remove or not the affected teeth.[8]

Surgical extraction of the affected unerupted teeth, followed by autotransplantation of tooth germs or orthodontic treatment was reported in the literature.[22-24]

The use of endosseous implants was also considered as a beneficial therapy for patients with RO but only after the pubertal growth.[8,25-27]

For both therapies, a temporary removable space maintainer type partial denture must be performed during the transitional phase.

Magalhães et al. recommended the preservation of the RO uninfected teeth in the dental arch to promote normal dental arches development.[10,25]

In the current report, a removable partial denture as functional space maintainer was performed and renewed 2 years later, and the patient was kept under a regular radiographic follow-up to monitor the calcification and the eruption of the affected teeth.[10]

In conclusion, there is currently no consensus on the best therapeutic approach for RO. However, this disorder requires an interdisciplinary approach involving pediatric, prosthodontic, and orthodontic specialists and patient with RO require a long follow-up.

## AUTHOR CONTRIBUTIONS

RM and FC performed the initial examination and collected clinical data. RM provided the clinical dental care of the patient and continued performing regular clinical and radiographic follow-up. FM, FM, and AB supervised the clinical dental care of the patient. RM and FC was responsible for the literature search and wrote the paper. FM revised and edited the manuscript and figures. HG provided comprehensive judgement and assisted in editing the final version of the manuscript. All authors read and approved the final version of the manuscript prior to submission.

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## CONFLICT OF INTEREST

None of the authors has declared any conflict of interest or financial disclosures.

## DATA AVAILABILITY STATEMENT

All data generated and analyzed which related this work are included in this published article.

## CONSENT

Written informed consent was obtained from the legal guardian of the patient for publication of this case and accompanying images.

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