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A Case Report on Apparent Endodontic Failure in a Patient having Cleidocranial Dysplasia

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Abstract

The medical history of a patient and any associated oral manifestations can greatly exacerbate misleading signs and symptoms, making a diagnosis difficult. The apex of a previously treated tooth (tooth #9) was still radiographically radiolucent in a 21-year-old female patient; endodontic failure and asymptomatic apical periodontitis were anticipated. This article describes how a patient's cleidocranial dysplasia syndrome had a significant impact on her dental history, which included the existence of numerous extra teeth. The patient's youth necessitated extensive surgical intervention to eliminate the extra teeth, which led to an endodontic misdiagnosis in her adult years. A periapical scar was identified as the patient's condition after clinical and radiological testing. The occurrence of periapical fibrous scars, which are an uncommon fibrous tissue healing process following surgical and nonsurgical procedures, ranges from 2.5% to 12%. The diagnosis, pathogenesis, risk factors, and methods for long-term monitoring of fibrous scars are all included in this paper.

Keywords: Cleidocranial Dysplasia, Fibrous Scars.

Introduction

With an incidence of 1 in 1 million, cleidocranial dysplasia or dysostosis is an autosomal dominant hereditary disease that affects dental and skeletal development. It was initially identified by Martin in 1765, and it has been noted to have an impact on intramembranous ossification brought on by a CBFA1 gene mutation, insertion, or deletion.[2] Lower stature, undeveloped or nonexistent clavicles, and anomalies of the skull including frontal and parietal bossing are typical manifestations. It's also typical to have a hypoplastic maxilla and delayed fusion of the cranial sutures, which result in a projecting jaw and midface retrusion.[4] Dental anomalies include supernumerary teeth, which can cause crowding and resorption, delayed exfoliation of primary teeth, delayed eruption of primary and permanent teeth,

and occlusal anomalies. Patients may express concerns about their teeth's poor aesthetics and functional occlusal issues, with some presenting with enamel hypoplasia, taurodontia, and dentigerous cysts[5].

The goal of this paper is to demonstrate how a patient's complicated medical history can significantly impact their dental history and present a diagnostic problem. The present research discusses diagnostic methods and variable management techniques.

Case Presentation

A 21-year-old female patient with a suspected failed root canal procedure on the maxillary left central incisor was sent to the Department of Conservative Dentistry and Endodontics, Institute of Dental Sciences, Bareilly. The patient's tooth #9, which was discoloured and did not respond well to sensibility tests, was first examined by the paediatric department two years ago. Primary endodontic therapy was finished after a diagnosis of pulpal necrosis and asymptomatic apical periodontitis. Since then, the tooth has remained symptom-free. Cleidocranial dysplasia with no known family history of the disorder is present in the patient's medical history. Her dental history includes several extractions of additional teeth under general anaesthesia when she was 10 years old.

The patient didn't describe any symptoms or indicators. The clinical examination indicated no pain to palpation/percussion testing of any of the maxillary anterior teeth and normal responses to sensibility tests of the neighbouring teeth. Sinuses, discolouration, extensive periodontal pockets, mobility, or coronal leakage were not present. A periapical radiograph was provided by the new referral (Fig. 1), which revealed well-condensed obturation and radiolucency at the tooth's apex, both of which were compatible with the presence of periapical periodontitis. The primary differential diagnosis at this point was asymptomatic apical periodontitis of tooth #9, which had already undergone treatment.

It was determined through a cone-beam computed tomography (CBCT) radiograph (Fig. 2) that tooth #9 was well obturated and had a periapical radiolucency as a result. An oral and maxillofacial radiologist who read the pictures noted a low-density apical zone that was about 5.6 mm in the mesiodistal direction and extended about 2.7 mm above the apex of tooth #9. The periapical area was said to spread through the neighbouring floor of the nose as well as the buccal and palatal cortices next to tooth #9's apex.

In order to identify the number and positions of the supernumerary teeth, previous CBCT radiographs were requested and examined due to the patient's history of supernumerary tooth extractions. A supernumerary tooth was discovered in the anterior maxillary region after additional review of the CBCT records from 8 years earlier (Fig. 3). The supernumerary tooth that was previously surgically removed from the maxillary anterior region was closely related to the location of the present radiographic radiolucency that was discovered. Additionally, CBCT images taken two years prior to the beginning of the initial endodontic therapy did not reveal any differences in the size of the radiolucency prior to and following the endodontic treatment of tooth #9 (Figs. 2 and 4).

The patient was diagnosed with an apical scar located in the anterior maxilla associated with the apex of tooth #9 that was likely caused by the surgical extraction of the supernumerary tooth in the anterior maxillary region. Because of the absence of evidence ofactive disease and no changes in radiographic appearance, no treatment was deemed necessary.

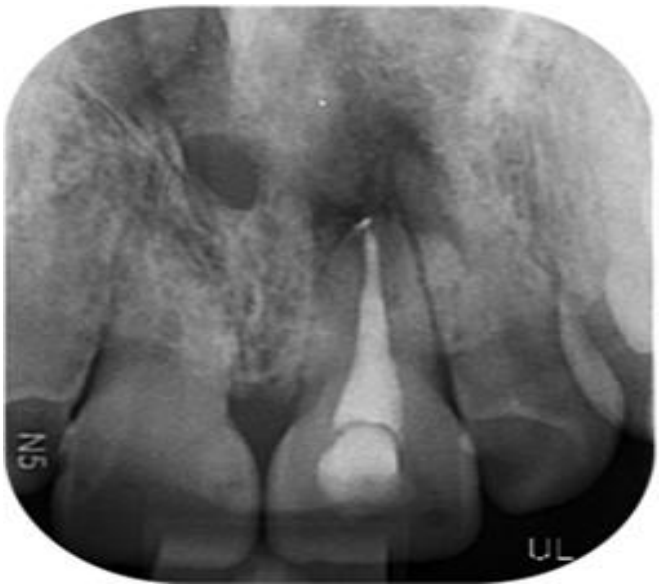

Figure 1: The periapical radiograph supplied with the original referral showing a large periapical radiolucency associated with the apex of tooth #9

Discussion

Cleidocranial dysplasia can have significant effects on patients' dental health and development considering its uncommon occurrence. The CBFA1 (also known as RUNX2) gene mutation influences chondrocyte differentiation, osteoblast function, and skeletal morphogenesis mechanisms. The RUNX2 gene controls tooth morphogenesis and histodifferentiation of the epithelial enamel organ through interactions between epithelial and mesenchymal cells. [6] Due to the disruption of these processes in cleidocranial dysplasia patients, supernumerary teeth may develop as a result of a lack of resorption and an excess of dental laminae.[5] The literature also reports a number of cranial anomalies, such as frontal bossing and delayed ossification of the sutures of the skull, the facial bones, and the cranial base.[6, 7] A multimodal therapy approach is typically used since the required treatment is complex.[8] In the majority of situations, this entails combining orthodontics with oral and maxillofacial surgery with the goal of removing the extra and deciduous teeth while also exposing the permanent teeth to aid in their emergence. Depending on the type of modality employed, such as the Toronto-Melbourne strategy or the Belfast-Hamburg method, the timing and delivery of surgical care may change.[6] The permanent teeth are then given orthodontic treatment to direct their eruption and alignment, with the option of orthognathic surgery to improve the skeletal connection if necessary.[4] Given the complexity of the situation, the course of therapy may last several years, beginning at age 5 to 6 with the Toronto-Melbourne method and concluding in maturity with orthognathic surgery.

A crucial diagnostic component of apical periodontitis is bone loss in the periapical area, which can be identified as radiographic radiolucency.[9] Radiographic decrease and resolution of the periapical lesion are signs of osseous regeneration following successful root canal therapy. [10] But it is well documented in the literature that periapical healing can be a long process, with 15% of cases taking up to 48 months and roughly 50% of cases demonstrating signs of radiographic reduction in the size of the lesion at a 6-month interval.[11] The primary cause of periapical radiolucency has been proven to be incomplete root canal system disinfection, which results in persistent intraradicular infection; however, there are a variety of other factors that must be taken into account.[13]

They involve the development of true or pocket cysts, the accumulation of cholesterol crystals, periapical scarring, and extraradicular infection brought on by periapical actinomycosis, as well as foreign body reactions to substances like gutta-percha.

As a result, it can be inaccurate to judge the effectiveness of endodontic therapy purely based on radiographic appearances. Endodontic failure diagnosis is a difficult process that combines clinical findings, historical

radiographic examination, and the patient's complaints. Furthermore, as seen in this instance, the patient's medical and dental records can offer vital details to help with the assessment.

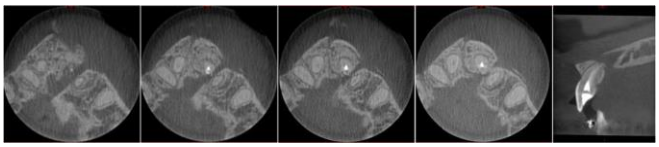

Figure 2: The latest CBCT radiograph showcasing a well-obturated tooth #9 with a large radiolucent lesion affecting both the palatal and buccal cortices.

Following endodontic treatment, periapical scars, also known as post-endodontic periapical fibrous scars, are an unusual healing process with collagenous fibrous tissue.[14] According to reports, their incidence ranges from 2.5% to 12%.[13] Histologically, they consist of dense collagenous fibrous tissue containing fibroblasts, which is often devoid of inflammation in the described cases. A periapical scar lesion with chronic inflammation was described by Lee et al., [14] but it was assumed that the imbedded amalgam particles in the lesion were to blame. Following a successful root canal procedure, the migration of osteoblasts or undifferentiated mesenchymal stem cells occurs in the presence of the proper induction or bone growth factors,[14] which initiates the bone regeneration process. The surrounding periosteum or endosteum is often where the bone-forming cells are recruited. One of the known risk factors for the development of periapical scars is the lack or destruction of surrounding cortical plates,[11] which prevents the necessary bone-forming cells from being recruited. On the contrary, fibroblasts that create connective tissue invade the region, and scar tissue forms as part of the healing process.[9] There have also been reports of other reasons, including the overproduction of type III collagen and disruptions to the re-modelling stage of wound healing.[14]

The stable periapical scar lesions are asymptomatic, show good periodontal health, and don't show signs of a root fracture.[14] Edema and fistulation have been seen in a few instances, which could occur in the event of a reinfection.[16] On radiographs of teeth with concurrent periapical scars, the size of the radiolucent periapical scar lesion indicates effective endodontic treatment/retreatment, and this lesion size is constant over time.[14] The lamina dura, which divides the tooth's apex from the periapical scar lesion, has been reported as one of the distinctive radiographic properties.[11]

The computed tomography (CBCT) technique has been used in these situations to track the radiographic size of the periapical scar due to its three-dimensional imaging capabilities, high resolution, accuracy, and accessibility. In comparison to traditional 2-dimensional radiography, the patient is exposed to more radiation, the scanning procedure takes longer, and the cost is higher. Magnetic resonance imaging (MRI) and real-time ultrasonography (US) are two more recommended monitoring methods.[11]Scar tissue can be effectively monitored and detected using magnetic resonance imaging, which doesn't release ionising radiation and provides strong soft tissue contrast.[17]

It has been demonstrated that US, a distinct nonionizing imaging option to CBCT, can give accurate periapical lesion diagnostic and monitoring capabilities.[18] The requirement that a cortical plate be absent in order for the ultrasonic waves to penetrate the lesion is one of the fundamental constraints of US. Cortical plates would typically be absent due to the type of the periapical scar lesion's creation, nevertheless this may facilitate the use of this approach in the monitoring of these patients.

Since periapical scars are diagnosed histologically, a biopsy of the lesion permits the diagnosis to be verified. The invasiveness of this method, the chance that the

material will be distorted, and the possibility of sectioning errors leading to a possible incorrect diagnosis are its drawbacks when used to collect periapical lesions. In fact, Simon et al.,[19] demonstrated that CBCT imaging may be a more reliable and practical approach than a biopsy to diagnosis big periapical lesions. However, because there was no diagnosis of a periapical scar in this study, it is important to exercise caution when evaluating the results' applicability. Alternatively, a novel approach by Zmener et al.,[20] has been suggested using a core bone biopsy needle to obtain the histologic sample of the periapical scar in a non-invasive manner.

An explanation of the potential risks and advantages with the patient helps establish if a biopsy of the lesion needs to be performed. It is less invasive for patients to monitor the suspected periapical scar area non-surgically, and invasive procedures can be performed if symptoms appear.

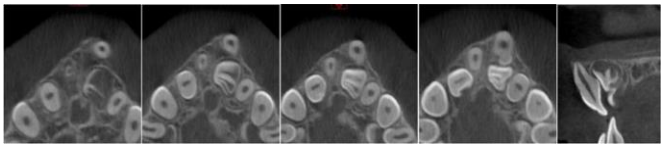

Figure 3: A CBCT radiograph taken when the patient was 10 years old showcasing a supernumerary tooth in the anterior maxilla in close proximity to the erupting tooth #9. This radiograph was taken to assess the position of the supernumerary teeth in the maxilla before their surgical removal.

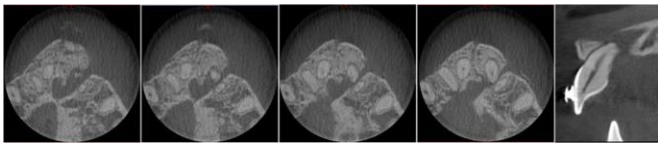

Figure 4: A CBCT radiograph taken when the patient was 16 years old showcasing a large radiolucent bony cleft extending through both the buccal and palatal cortices adjacent to the root apex of tooth #9. This radiograph was taken to assess the anatomy of tooth #9 and the associated periapical radiolucency before the nonsurgical primary endodontic treatment of tooth #9.

The purpose of this case study was to emphasise the value of taking a complete medical history, especially when a condition like cleidocranial dysplasia and the accompanying supernumerary teeth are involved. The researchers make no attempt to establish a direct causal relationship between cleidocranial dysplasia and the development of apical scars. However, there is proof that the periapical scar may have developed as a result of the presence of extra teeth and their subsequent surgical removal. No histologic examination was made in this instance because the clinical and radiographic results supported the diagnosis of a periapical scar. The magnitude of the radiographic radiolucency has remained constant for at least the past two years, and it will continue to be routinely assessed both clinically and radiographically in the future. Patients with apical scars should be given a letter or a dental passport summarising the diagnosis and outcomes of earlier tests due to the uncommon incidence of periapical scar lesions. To avoid a false diagnosis and inappropriate treatment of the afflicted tooth, this should be conveyed to the patient's present and/or future dentists.

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