The challenge of managing patients with generalized short root anomaly: A case report

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The case of an 18-year-old woman with generalized short root anomaly (SRA) is presented, and the clinical management and challenges associated with this rare condition are discussed. The patient was referred for restoration of the edentulous maxillary left anterior region. Due to the SRA, the patient had previously received limited orthodontic treatment for the ectopic maxillary canines. The maxillary left canine failed during orthodontic traction, the left lateral incisor migrated in its region and had poor prognosis due to severe mobility. Therefore, it was extracted and the region was restored with an implant-supported cantilever metal-ceramic fixed partial denture.

Radiographic examination revealed generalized SRA along with other dental anomalies. From the patient’s family history, as well as from the physical, clinical, radiographic, biochemical, and histologic evaluations, we were unable to identify the etiology behind this unique combination of dental anomalies. Esthetics and function were successfully restored and a strict recall system was implemented in order to monitor and maintain the short-rooted teeth. The need to prevent tooth and periodontal tissue deterioration in patients with generalized short roots is emphasized. (Quintessence Int 2018;49: 673–679; doi: 10.3290/j.qi.a40481)

Key words: dental syndrome, displaced canines, short root anomaly, taurodontia, third molar agenesis, transposition

Short root anomaly (SRA) was first described by Lind1 in maxillary central incisors, and is defined by a crown-to-root ratio of 1:1 or less.2 The prevalence of isolated short-rooted maxillary central incisors has been reported to be approximately 2.4% in white populations,3 and 10% in Japanese populations.4 The anomaly is seen three times as commonly in girls as in boys.1 The maxillary central incisors are almost always affected, followed by the premolars, maxillary canines, mandibular central incisors, maxillary lateral incisors, and first molars.5 SRA has been proposed to be an intrinsic, inherent anomaly rather than a result of resorption or of an exogenous disturbance.1 Since root development

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precedes tooth eruption, the diagnosis of SRA is often delayed until 1 to 2 years after tooth emergence.5

Generalized root shortening is a very rare finding with only few reported cases.5,6 It is mainly observed in patients with syndromes or underlying systemic conditions. However, sporadic cases5-13 and familial1,5,6,14-17 occurrences of nonsyndromic SRA with multiple other dental anomalies have also been described. SRA usually occurs without any clinical symptoms except for occasionally increased tooth mobility.1 Cases with no apparent systemic etiology, particularly when the occurrence is sporadic, may present with a diagnostic dilemma. In addition, delay of proper diagnosis may increase the risk of deleterious complications.

This article presents a case with generalized SRA and discusses the clinical management and challenges associated with this rare condition.

CASE PRESENTATION

An 18-year-old woman was referred for prosthetic restoration of the edentulous maxillary left anterior region. The patient’s medical history was noncontributory. She was born full-term by nonconsanguineous parents and had not undergone any serious medical event. The first dental record available was a panoramic radiograph taken at the age of 9 (Fig 1). The dental history revealed that the patient underwent interceptive orthodontic treatment at the age of 11 to facilitate the eruption of the ectopic permanent maxillary canines. The primary maxillary canines were extracted and fixed appliances were placed in the maxillary anterior teeth. However, in consecutive panoramic radiographs at the ages of 11 and 12 (Fig 2), it was evident that the maxillary canines were severely mesially displaced and spontaneous eruption was considered highly unlikely. Therefore, orthodontic treatment with surgical exposure and orthodontic traction of the maxillary canines was performed. During traction, the left maxillary canine failed; orthodontic treatment was discontinued, and a removable Hawley appliance was constructed in order to restore esthetics in the region.

During the prosthetic consultation, the clinical examination revealed that the maxillary left canine was missing. The maxillary left lateral incisor had migrated in its region and showed Grade III mobility. A transposition was observed between the maxillary right lateral incisor and canine; the third molars were missing; and the patient had a mild Class III dental relationship. The dental crowns appeared normal, all teeth were vital, and the patient was free of caries and periodontal disease and had good oral hygiene standards. The oral tissues were within normal limits (Fig 3). A panoramic radiograph was taken prior to the initiation of the restorative treatment (Fig 4).

The review of all four panoramic radiographs (Figs 1, 2, and 4) disclosed that tooth root development was abnormal, as the roots of most teeth were short. Specifically, the maxillary incisors and premolars exhibited particularly short, blunted roots, while the molars presented a more conical-spiky root morphology. The mandibular incisors and premolars had conical-spiky
roots with a sharp edge, while the mandibular molars were taurodontic. The maxillary right third molar was impacted, while the other third molars were congenitally missing. A comparison among these radiographs showed that the appearance of the roots was the same before (at the age of 9), during (at the ages of 11 and 12), and after orthodontic treatment (at the age of 18). The radioluency seen around the crown of the impacted maxillary left canine could represent an enlarged dental follicle or a dentigerous cyst, while the radiopacities present mesially to the unerupted maxillary right first premolar and coronally to the unerupted mandibular right second molar crowns were interpreted as more consistent with calcifications in the dental follicles (Fig 2). However, none of the lesions were present for further evaluation (Fig 4).

Panoramic radiographs of the patient’s 50-year-old mother and 25-year-old sister were also available, and did not show similar findings. In addition, the patient’s parents could not recall whether any other family member had lost his/her teeth prematurely. A complete blood count (CBC) performed in the course of a regular check-up by the patient was within normal range.

As prognosis for the maxillary left lateral incisor was poor (Grade III mobility), the tooth was extracted under local anesthesia and an implant was placed in the area at the same time. The implant was immediately loaded with a temporary acrylic cantilever fixed partial denture in order to restore function and esthetics in the region until the final restoration. The extracted tooth was fixed in 10% buffered formalin solution and submitted for histologic examination. After decalcification in Surgipath decalcifier II (Leica Biosystems) and paraffin embedding, 5-μm thick sections were cut and stained with hematoxylin and eosin. Microscopic examination showed that the tooth was composed of mature dentin surrounding a pulp cavity occupied by a fibrovascular pulp, and covered by cementum (Fig 5).

Finally, an implant-supported cantilever metal-ceramic fixed partial denture was placed to restore the edentulous maxillary left anterior region (Fig 6). The transposition between the maxillary right lateral incisor and canine, which resulted from severe maxillary right canine mesial displacement, was not corrected orthodontically due to the underlying increased risks. The esthetic problem was addressed with the use of composite material on the maxillary right canine to mimic the appearance of a lateral incisor tooth. The patient was placed in a strict recall system in order to monitor the dental root condition and prevent the occurrence of dental or periodontal disease. The impacted maxillary right third molar is currently under observation and will be extracted in the future (Fig 7).

DISCUSSION

The case presented herein showed generalized SRA in association with other dental anomalies, such as taurodontia, displaced maxillary canines, and agenesis of third molars. Although canine displacement and agenesis of third molars are quite common, generalized SRA is a very rare condition, which poses diagnostic and
clinical challenges. In the present case, esthetics and function were successfully restored and a strict recall system was implemented.

The pathogenesis of short-rooted teeth is still uncertain and the diagnosis is often challenging. In general, short roots could derive from external apical root resorption or be the result of a disturbed developmental process. Root resorption has been associated with underlying systemic conditions, such as rare bone dysplasias, Papillon-Lefévre syndrome, and scleroderma. No such disease had been diagnosed in the present patient, and the available clinical and laboratory findings were not suggestive of a systemic condition. External apical root resorption is a common undesirable sequela of orthodontic treatment. Consecutive radiographic monitoring in the present case showed that root length and morphology remained rather stable throughout a 10-year observation period, ruling out that orthodontic treatment was a possible etiologic factor. In addition, teeth with resorbed roots have a different morphology compared to teeth with underdeveloped roots. In root resorption, the apical surface appears often irregular, compared to the apical surface in SRA which is radiographically smooth, and often exhibits short, rounded stumps or short, thin, pointed roots. Furthermore, histologic examination in root resorption shows multinucleated osteoclasts and resorption lacunae. The radiographic and histologic findings in the present case were not consistent with root resorption. The lack of progression also pointed towards a developmental etiology.

SRA has been reported in patients with short stature syndromes, such as in Aarskog syndrome, in Seckel bird-headed dwarfism, as well as in Hallermann-Streiff syndrome. In addition, it has been observed in patients with mental retardation, such as in the case of Laurence-Moon-Bardet-Biedl syndrome. Examples of metabolic diseases that are associated with SRA are idiopathic hypoparathyroidism and pseudohypoparathyroidism, as well as thalassemia. Since the present patient’s medical history, physical examination, and blood evaluations were all within normal range, such syndromes and conditions were excluded.

Hereditary dentin disorders, such as dentin dysplasia and dentinogenesis imperfecta, may also affect...
specifically, dentin dysplasia type I has been associated with radiographically sharp conical roots with apical constrictions or rootless teeth, while the clinical appearance of the crowns appears normal. Other radiographic features of dentin dysplasia type I in permanent teeth include pulpal obliterations, crescent-shaped pulpal remnants, and numerous periapical radiolucencies. The radiographic findings in the present case, and the presence of normal dentin in the extracted tooth, verified histologically, were not consistent with dentin dysplasia type I, or with other associated hereditary conditions. Moreover, the present patient had normal tooth color and morphology, no tooth wear, and her medical history was free of bone fractures, blue sclerae, and hearing loss. Therefore, different types of dentinogenesis imperfecta and associated osteogenesis imperfecta were excluded. Finally, the patient had never received radiation or chemotherapy, thus possible exogenous factors disturbing root development were ruled out.

Nonsyndromic SRA has been described in patients with or without other dental anomalies. Seven of those cases were sporadic, whereas in seven studies a familial occurrence was indicated. In six of those reports the patients were also shown to be of short stature. Most familial cases indicated an autosomal dominant pattern of inheritance; however, X-linked inheritance could not be excluded. As far as other dental anomalies are concerned, dens invaginatus, microdontia, taurodontia, talon cusps, reduced alveolar bone levels, and multituberculism of molars have been described with SRA. A few cases revealed an association of SRA with external root resorption, with an infected cyst, obliterated pulp chambers, widened root canals, pulp stones, tooth mobility, and anterior crossbite. It should be noted that SRA has also been described in cases with unerupted ectopic canines. In comparison to the latter studies, the present patient presented a unique combination of generalized SRA with taurodontia, agenesis of third molars, ectopic maxillary canines, and a Class III dental relationship. From the mother and sister’s panoramic radiographs, as well as from the lack of similar family history, it was concluded that a familial occurrence was unlikely in this case. However, since dental and radiographic examination of other family members was not possible, such a possibility could not definitively be excluded.

It has been suggested that SRA may result from a disturbance in the signaling pathways in Hertwig epithelial root sheath, causing premature closure of the apex. Wnt/β-catenin signaling and the Nfí gene were shown to be key regulators of dental root formation in mice. Another important finding is a continuing gelatinase (MMP-9) activity observed in the periodontium of short-rooted teeth. This might indicate the periodontal remodeling needed in order to withstand the increased stress loads due to short root structure. A common genetic cause underlying the present patient’s dental characteristics could not be determined and should be investigated in future studies.

Since SRA is not evident clinically, careful radiographic evaluation is essential for proper diagnosis. Care should also be given not to misdiagnose the SRA condition as root resorption. The importance of preventive measures as well as early treatment of caries is stressed in order to avoid root canal therapy that is often challenging in severely underdeveloped roots. In addition, it is important to prevent periodontal inflammation since short-rooted teeth cannot afford any loss of supporting bone. Further aims of dental treatment in patients with SRA could be the reestablishment and...
normalization of the occlusion and facial esthetics when teeth are lost due to severe tooth mobility. Function and esthetics have been restored in the present patient and a strict recall protocol has been implemented to monitor and maintain oral health.

There are important implications regarding the orthodontic treatment of patients with SRA. Specifically, an increased tendency towards root resorption during orthodontic therapy and due to prolonged treatment time in cases with impacted canines has been reported in SRA dentitions. Moreover, increased root resorption has been noted in patients with tooth agenesis and taurodontia. Therefore, long complex treatment and heavy orthodontic forces should be avoided in cases with such a combination of dental anomalies, and frequent radiographic monitoring is recommended. It is also necessary to deal with ectopic canines before they could lead to additional shortening of the lateral incisor roots. Nonetheless, unless an exceptionally heavy force is exerted, it has been suggested that short-rooted teeth function well. Indeed, orthodontic treatment was successful in a case with SRA without further shortening of the dental roots. In the present case, tooth movement occurred without a general deterioration of tooth root structure. However, during traction of the maxillary left ectopic canine the tooth failed, indicating the need for particular caution when applying mechanical forces to short-rooted teeth. It is speculated that the failure of the maxillary left canine during orthodontic traction was associated with the generalized SRA. However, since cone beam computed tomography (CBCT) during orthodontic treatment and information regarding the specific force levels applied during traction were not available, the etiology behind the maxillary left canine failure remains uncertain. Further research is recommended to evaluate the effects of orthodontic force application on short-rooted teeth.

CONCLUSION

A case with generalized SRA in association with other dental anomalies is presented, in which no syndromic, systemic, or other explanation could be identified as a possible etiologic factor. To the best of the present authors’ knowledge, a similar association of generalized SRA has not been previously described. The constellation of dental anomalies in this patient may represent a subgroup of SRA rather than a novel dental syndrome. Further studies are needed to test this hypothesis.

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REFERENCES