

Cone Beam Computed Tomographic Characterization of Short Root Anomaly in Chloroquine-Induced Stevens-Johnson Syndrome – a Case Report

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Abstract

Stevens-Johnson syndrome (SJS) is a rare medical condition with severe mucocutaneous reaction due to infection or adverse drug reaction. The present case reports the impact of chloroquine-induced SJS on the tooth root development. A 20-year-old Indian male reported to conservative dentistry and endodontics speciality clinic with the chief complaint of food lodgement and sensitivity in maxillary and mandibular posterior teeth. He had a past medical history of severe cutaneous reaction after taking Tab. Lariago (chloroquine) for treatment of malarial fever at the age of 8 years. The acute inflammatory immune response was managed by hospitalization and administration of steroids and anti-inflammatory drugs. Clinical examination revealed dry mucosa, carious teeth with adequate oral hygiene. Panoramic X-ray showed multiple teeth with short roots. A detailed cone beam computed tomographic scan (CBCT) demonstrated a healthy bone trabecular pattern with the absence of any periapical radiolucency. Short, blunt roots with immature apex were seen in many teeth. Based upon the measurement of root to the crown ratio on the CBCT scan and correlating the development status of teeth with the medical history, a diagnosis of short root anomaly (SRA) after chloroquine-induced SJS was made. This is the first report to describe the three-dimensional features of teeth with SRA in a patient with SJS. Diagnostic, restorative, and endodontic implications of SJS are highlighted.

Keywords

rhizomycry, short root anomaly, Stevens-Johnson syndrome

INTRODUCTION

Stevens-Johnson syndrome (SJS) is a rare medical condition with a prevalence of 7.1 per million people.¹ It is characterized by exudative multiform erythema, stomatitis, conjunctivitis (sometimes leading to keratitis and panophthalmitis), and severe constitutional symptoms.² Although the aetiology includes infections like mycoplasma pneumonia, vaccine, graft vs. host reaction, and idiopathic

disease, it is most commonly associated with an adverse reaction to certain drugs.³ SJS is classified into three types: erythema multiform (smooth form), SJS (main form), and toxic epidermal necrolysis (TEN) (severe form), depending on the total body surface area affected by an adverse drug reaction.⁴ Pathogenesis and clinical presentation are similar for SJS and TEN, differing only on their extent of skin detachment following severe epidermolysis.⁵

Cutaneous sequelae are the most common long-term

complications of SJS, including post-inflammatory dyspigmentation, abnormal scarring, eruptive naevi, and nail changes.⁶ The risk of nail involvement (onychoma, dystrophy, anonychia, pterygium, ridging, streaky pigmentation, and longitudinal nail ridges) is related to the severity of the disease.⁷ Chronic ocular sequelae are the most disabling complication seen in all three forms of SJS, which can lead to partial or absolute loss of vision. Oral complication primarily includes xerostomia and difficulty in mastication or deglutination.⁶ Dental growth abnormalities due to SJS range from dental agenesis, root dysmorphia, short root, microdontia, and incomplete root apex closure, depending upon its time of onset, i.e., during primary or mixed dentition.⁸⁻¹⁰ The short root anomaly (SRA) with immature apex can present as a diagnostic dilemma and treatment challenge for endodontists. This paper aims to report 3D cone-beam computed tomographic (CBCT) features of SRA in the case of SJS and to discuss their diagnostic, restorative, and endodontic implications.

CASE REPORT

A 20-year-old Indian male was referred to the Conservative Dentistry and Endodontics specialty clinic with a chief complaint of sensitivity and food lodgement in maxillary and mandibular posterior teeth on both sides. His past medical history was remarkable for severe illness at the age of 7 years. Medical records indicated that the patient had an acute episode of erythema and skin desquamation all over the body along with conjunctivitis and oral ulceration one day after taking chloroquine tablet (Tab. Lariago™, Ipca Laboratories Pvt. Ltd., India) for the treatment of malaria. A diagnosis of

SJS due to the adverse reaction of chloroquine was made at that time. He was hospitalized for a month and prescribed topical steroids and analgesics to treat the disease condition. Although the skin lesions healed at the time of discharge, there was persistent irritation and dryness of eyes. This condition gradually progressed into partial loss of vision at the age of 12 years for which he underwent corneal transplant surgery. He had a dental history of extraction of tooth #47 due to increased mobility 18 months ago. There was no previous history of dental trauma or orthodontic treatment.

The patient's height (164 cm), weight (54 kg), BMI (20.6), and intelligent quotient were within the normal range. Clinical examination revealed mottled pigmentation on the face and bilateral blepharitis (Fig. 1A). Malformed nails with pterygium and longitudinal ridge formation were also noticed in upper and lower limbs (Figs 1B, 1C). On intraoral examination, dryness of oral mucosa was observed. Oral hygiene was satisfactory, with no signs of gingival inflammation. Hypoplastic enamel was present on the buccal surfaces of both maxillary and mandibular second premolars on the left side. Deep dentinal carious lesions were present in teeth (#17, 26, 27, 37, 36, and 46). Bilateral maxillary lateral incisors (#12 & 22) and right mandibular first premolar (#44) were congenitally missing (Figs 1D, 1F).

Orthopantomogram (OPG) (Villa system medical) revealed multiple teeth in maxillary and mandibular jaw with abnormal short roots with blunting at root apex and an impacted tooth in mandibular jaw. There was no radiographic evidence of periapical pathology, and the trabecular bone pattern was normal (Fig. 2). All teeth responded positively to cold and electric pulp sensibility tests. The periodontal probing depth and tooth mobility were within physiologic limits for all the teeth. The patient was asked to chew on paraffin



Figure 1. A. Mottled pigmentation on the face and bilateral blepharitis; B. Malformed nails with pterygium and longitudinal ridge formation in upper limbs; C. Malformed nails with pterygium and longitudinal ridge formation in lower limbs; D. Intraoral labial view; E. Maxillary occlusal view; F. Mandibular occlusal view.



Figure 2. Orthopantomogram showing short root in teeth #17, 15, 14, 24, 25, 27, 37, 35, 33, and 45. Impacted tooth #44 is also seen.

wax, and the saliva sample was collected in a measuring container for 5 minutes. The stimulated saliva flow rate of 0.5 ml/min indicated towards decreased salivation. Haematological parameters (urea, creatinine, alkaline phosphatase, SGPT, SGOT, calcium, uric acid, phosphate, total cholesterol) were within the normal range. The family history was non-contributory.

To further investigate the atypical finding of short roots in multiple teeth, a CBCT scan (16×8 FOV, 0.2 mm voxel size) was advised. The CBCT images were stored in DICOM format and viewed using Carestream 3D software (Carestream Health Inc.) on an HP 21-inch LED monitor with a resolution of 1280×1024 under ambient light conditions. The scan was evaluated in coronal, sagittal, and transverse planes using oblique slicing mode by a single operator (VY). Abnormal incompletely formed root with funnel-shaped apical root canal opening was seen with relation to teeth #17, 27, and 37. Root with blunt apices and onion-like appearance was noticed in teeth #15, 14, 24, 25, 35, 33, 42, and 45, including

the impacted tooth #44 (**Fig. 3**). Apical foramen of diameter > 1 mm was noticed with relation to teeth #17, 15, 14, 24, 25, 27, and 37. Rest all teeth had complete root formation with closed apex. The root/crown (R/C) ratio for the teeth was measured on the CBCT scan using methods given by Choi et al. (**Fig. 4**).¹¹ A reference line was drawn by joining highest point on labial and palatal CEJ in the sagittal section.

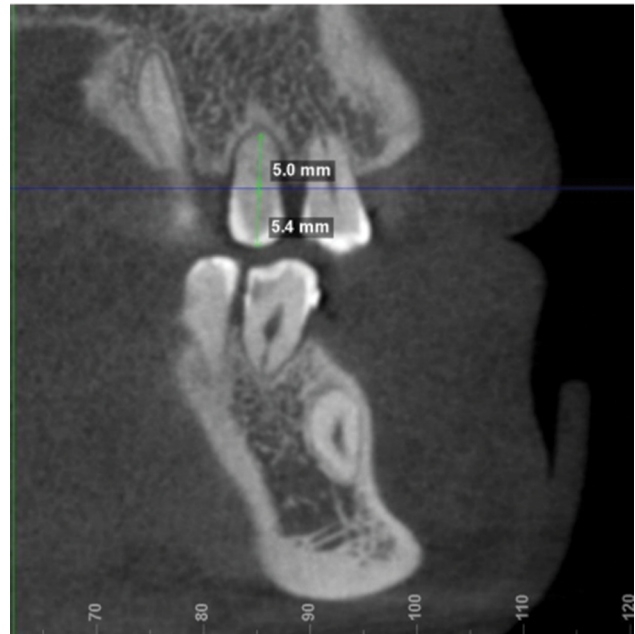


Figure 4. Measurement of root and crown length, line joining cemento-enamel junction is taken a reference line. Distance from cusp tip and root apex to the reference line is considered as crown and root length, respectively.

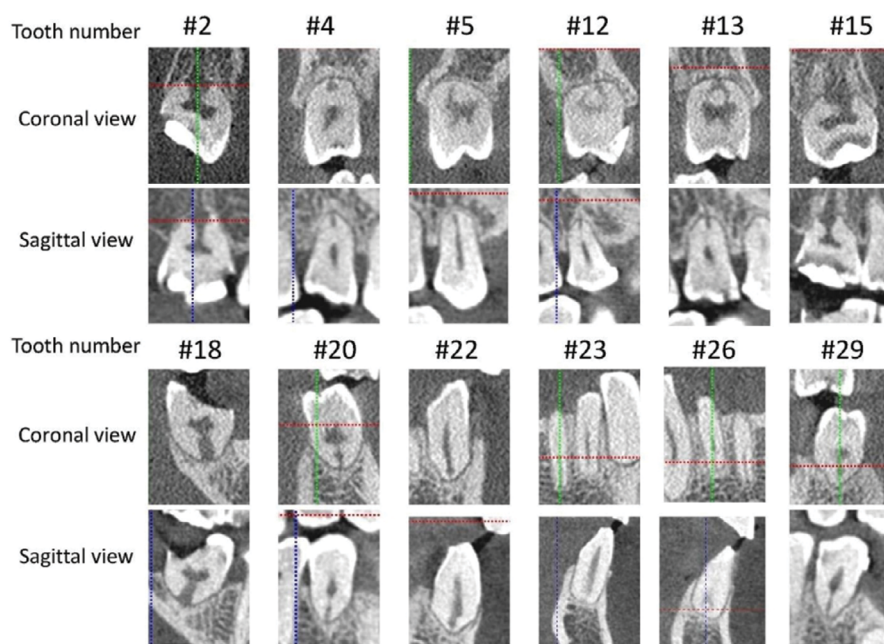


Figure 3. CBCT images of teeth with malformed root in coronal and sagittal cross section. The apical features are explained in **Table 1**.

A distance on perpendicular drawn from the reference line to the incisal tip and root apex was used to measure crown and root length respectively. Teeth #17, 14, 24, 25, 27, 37, 35, 33, 32, 26, 42, and 45 were categorized as SRA according to R/C ratio <1.1 (**Table 1**).

Based on medical history, clinical, and radiographic examination, the diagnosis of SRA in multiple teeth due to SJS syndrome was established. The conservative treatment plan was made, and all the carious lesions were restored with resin composite material (Tetric-E-Ceram Bulk Fill; Ivoclar-Vivadent, Schaan, Liechtenstein). Thereafter, a regular follow-up was planned. Informed consent was taken from the patient regarding the publication of the case records.

DISCUSSION

SRA is a disorder of tooth root development resulting in abnormal root morphology characterized by short and blunt roots.¹² The prevalence of SRA ranges from 0.6% to

10% with female predilection.^{13,14} The genetic factors (dentin dysplasia, dentinogenesis imperfecta), systemic diseases (hypoparathyroidism, hypocalcemia, thalassemia), environmental insult (trauma, chemotherapy, radiotherapy, orthodontic forces) and syndromic disorders (scleroderma, Down syndrome, and Laurence-Moon-Bardet-Biedl syndrome) can be attributed to the development of SRA.¹³⁻¹⁵ The differential diagnosis includes conditions in which the generalized root development cessation is often seen. In the absence of any other plausible explanation, SRA, in this case, could be attributed as a consequence of a fulminant attack of SJS after adverse drug reaction. This is in concurrence with literature where SRA is reported secondary to SJS.⁸ Antibacterial sulphonamides, anticonvulsants, and NSAIDs have been reported as the most common causative drug for SJS.¹⁶ In the present case, the intake of chloroquine was exclusively responsible for the attack of SJS. The most critical strategy for the management of SJS is the prevention of early disease progression. However, the disease has various long-term sequelae. The clinical findings of cuta-

Table 1. Linear measurements of root length, crown length, tooth length, root to crown ratio and apical root morphology of teeth seen on CBCT scan

Tooth no.	Tooth dimensions				Apical root end features
	Root length (mm)	Crown length (mm)	Tooth length (mm)	Root/ crown (R/C) ratio	
17*	4.7	6.3	11.0	0.74	Funnel shaped immature apex
16	12.4	5.4	17.8	2.30	Mature tapering apex
15	6.5	5.6	12.1	1.16	Immature apex with root end blunting
14*	6.3	5.8	12.1	1.08	Immature apex with root end blunting
13	16.9	8.0	24.1	2.10	Mature and tapering apex
11	11.4	8.9	20.3	1.28	Mature apex with slight blunting seen
21	11.3	8.8	20.1	1.28	Mature apex with slight blunting seen
23	14.6	7.0	21.6	2.08	Mature with tapering apex
24*	6.0	5.9	11.9	1.01	Immature apex with root end blunting
25*	5.5	5.7	11.2	0.96	Immature apex with root end blunting
26	12.3	5.3	17.6	2.3	Mature tapering apex
27*	5.4	5.9	11.3	0.91	Funnel shaped immature apex
37*	5.5	5.7	11.2	0.96	Funnel shaped immature apex
36	12.9	6.6	19.5	1.81	Mature tapering apex
35*	5.9	6.2	12.1	0.95	Mature apex with apical root blunting
34	10.2	6.2	16.4	1.64	Mature tapering apex
33*	5.8	8.0	13.8	0.72	Abrupt closure of apex with apical root blunting
32*	7.5	7.3	14.8	1.02	Abrupt closure of apex with apical root blunting
31	9.1	7.1	16.2	1.28	Mature blunt apex
41	9.2	7.1	16.3	1.29	Mature blunt apex
42*	7.3	7.2	14.3	1.01	Abrupt closure of apex with apical root blunting
43	13.8	8.0	21.8	1.7	Mature tapering apex
44*	7.0	6.7	13.7	1.04	Impacted tooth
45*	5.6	5.4	11.0	1.03	Apical root blunting
46	12.1	6.1	18.2	1.98	Mature tapering apex

neous pigmentation, scarring, and malformed nails, partial loss of vision along with dry mouth and multiple carious teeth in the present case were in consistence with the long term sequelae of SJS.

SRA and incomplete apical closure of multiple teeth was the interesting finding in the present case. In the SJS, the upper and lower epidermis is the most commonly affected. Necrosis of a few epidermal cells is seen in the mild lesion, whereas the severe lesion demonstrates the destruction of the whole epidermis. Similar histological features are seen in SJS lesions of the oral cavity, where the inner and outer enamel epithelia and root sheath of enamel follicle originate from the basal layer of the oral epithelia. The arrest of root development after SJS could be attributed to damage or destruction of keratinocytes of the Hertwig's epithelial root sheath or enamel follicle, which develops from oral epithelia.^{8,10,17,18} The developmental stage of the tooth at the time of the adverse event determines the effect on root growth. Teeth with complete or two-thirds root formation before this destruction are unlikely to demonstrate any effect except slight blunting of the root. Destruction after one-half of root formation can lead to cessation of further apical root elongation, and blunting of apex occurs due to continued deposition of secondary dentin by remaining odontoblasts.^{19,20} This is the reason for abrupt closure of apex in some of short rooted teeth. If this destruction happens before completion of one half of root, it inhibits the differentiation of odontoblasts from dental papilla cells resulting in the limited number of odontoblasts. It can result in malformation of the root dentin during root development.²⁰ Similar findings were demonstrated in the CBCT scan of the patient. However, the full root formation in the canines and left lower premolar, in this case, was an exceptional observation. It can be hypothesized that since the tooth bud of these teeth is situated very deep, they escaped large epidermal cell necrosis.²¹

The CBCT radiological assessment indicated that the growth disturbance must have occurred around the age of 8-9 years, which coincides with the patient's history of severe illness and strengthens the association between SJS and SRA in the present case. Teeth with decreased root to crown (R/C) ratio measuring <1.1 are described as having SRA.¹² The linear measurements on intraoral periapical radiograph, OPG, and CBCT scan have been used in literature to calculate the R/C ratio. The CBCT is an advanced dental imaging technology that has the advantage of distortion-free image reconstruction and visualization in three planes as compared to conventional radiography and OPG. Also, localization of cemento-enamel junction can be done more accurately on CBCT images, which is used as a reference point to obtain anatomical R/C ratio. The method proposed by Choi et al. was used in the present case to measure R/C ratio.¹¹

SRA poses treatment challenge for the dentists. Teeth with SRA are more prone to root resorption due to the generation of greater mechanical stress during tooth movement. The presence of impacted teeth can also complicate

and prolong the orthodontic treatment. The chances of immediate post-treatment relapse are more after orthodontic treatment in such teeth, and a permanent fixed splint is recommended.¹⁴ Compromised crown root ratio in SRA contraindicates the use of orthodontic extrusion or surgical crown lengthening procedures after the loss of coronal tooth structure; hence, the importance must be given to preventive and conservative methods of caries management. In this case, composite restorations were done as sufficient remaining dentin thickness was present. Apexification with mineral trioxide aggregate or bioceramic materials is essential for the endodontic treatment of non-vital teeth.

Appropriate management of patients with SRA requires accurate diagnosis that necessitates the collaboration of patient history, clinical, and radiographic examination. The routine radiographic methods have inherent limitations of 2D representation of the tooth and surrounding structures; the use of CBCT is necessary in such cases. CBCT allows for accurate assessment of the complex root canal morphology. The teeth can be simulated using the resin models, and a template can be prepared to minimize procedural errors during endodontic therapy.

CONCLUSIONS

The development of SJS at the age of tooth development can cause abnormal root formation. Multiple teeth were categorized as short root anomaly based on reduced root/crown ratio measured on CBCT scan.

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Конусно-лучевая компьютерная томографическая характеристика аномалии короткого корня при индуцированном хлорохином синдроме Стивенса-Джонсона – клинический случай

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Резюме

Синдром Стивенса-Джонсона (ССД) — это редкое заболевание с тяжёлой кожно-слизистой реакцией из-за инфекции или побочных реакций на лекарства. В данном случае показано влияние хлорохин-индуцированной ССД на развитие корня зуба. 20-летний мужчина индийского происхождения обратился в Клинику консервативной стоматологии и эндодонтии с серьёзной жалобой на застой пищи и чувствительность задних зубов верхней и нижней челюсти. В анамнезе у него была тяжёлая кожная реакция после приёма таблетки лариаго (хлорохин) для лечения малярии в возрасте восьми лет. Острый воспалительный иммунный ответ контролировали госпитализацией и введением стероидов и противовоспалительных препаратов. При клиническом осмотре выявлена сухость слизистых оболочек, кариозные зубы при адекватной гигиене полости рта. Панорамная рентгенография показала много зубов с короткими корнями. Детальная конусно-лучевая компьютерная томография (КЛКТ) показала здоровую трабекулярную структуру кости без периапикальной рентгенографии. У многих зубов наблюдаются короткие тупые корни с недоразвитыми верхушками. На основании измерения соотношения между корнем и коронкой сканера КЛКТ и соотнесения состояния развития зубов с анамнезом был поставлен диагноз аномалии короткого корня (АКК) после ССД, индуцированной хлорохином. Это первый отчёт, описывающий трёхмерные характеристики зубов с АКК у пациента с ССД. Акцент поставлен на диагностических, восстановительных и эндодонтических последствиях ССД.

Ключевые слова

ризомикрия, аномалия короткого корня, синдром Стивенса-Джонсона
