International Journal of Contemporary Pediatrics
Bisht H et al. Int J Contemp Pediatr. 2025 Sep;12(9):1563-1566
http://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20252611

Case Report

Innovative endocrown restoration in a situs inversus totalis patient using CAD/CAM technology

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Received: 11 June 2025
Revised: 11 July 2025
Accepted: 11 August 2025

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ABSTRACT

Situs inversus totalis (SIT) is a rare congenital abnormality characterized by a mirror-image transposition of both the abdominal and the thoracic organs. Frequency of situs inversus is 1:10,000 and is more frequent in males: 1.5:1. SIT is inherited as an autosomal recessive pattern and is associated with multiple gene mutations. Advanced imaging modalities can be used to assess fine anatomical details, which play a crucial role in these cases to plan radiologic or surgical interventions. Diagnosis is usually made when patient presents with other medical concerns as in this case report. The concurrence of aglossia and hypoglossia has been reported with situs inversus totalis condition in some of the literatures. The following case report discusses the dental management of a young girl with Situs Inversus Totalis condition.

Keywords: Congenital abnormalities, Dextrocardia, Situs inversus totalis

INTRODUCTION

Situs inversus totalis (SIT) is a rare occurrence that is defined by the complete positional inversion of both cardiac and abdominal viscera.[1] When the heart's anatomical position is completely reversed to the right side, then it is also known as dextrocardia with situs inversus.[2] Marco Severino first recognized dextrocardia in 1643 and it is generally an autosomal recessive genetic condition.[1] The main cause of situs inversus totalis is unknown, but in 2002, Bartoloni et al studied a mutation in the gene DNAH11 that seems to responsible for the condition.[1] In addition, other genetic components that are associated with the condition are lefty genes, nodal genes, and ZIC 3, ACVR2B and Pitxz genes.[3] According to estimates, the incidence rate lies between 1 in 10,000 and 50,000 live births.[4,5] It can also be associated with other conditions like primary ciliary dyskinesia, congenital heart defects, and splenic abnormalities.[5] In 1925 by Watkin first time reported the concurrence of SIT and aglossia and their sporadic cases in the literature. Although micrognathia and malocclusion were also observed in all of these cases, few cases identified other dentofacial deformities, which led to several secondary problems (e.g. masticatory difficulty, negative perception of facial aesthetics, psychological problems, and malnutrition) and greatly decreased the patients' quality of life.[6] The existing literature on the dental management of this condition is limited; therefore, this clinical report aims to aid in both the diagnosis and dental care of affected individuals.

CASE REPORT

A 12-year-old female child presented to the Department of Pediatric and Preventive Dentistry, University College of Medical Sciences, Guru Teg Bahadur (GTB) Hospital, Delhi, with a chief complaint of spontaneous pain in the lower left back tooth region for the past 3 to 4 days, accompanied by prolonged sensitivity to hot and cold

stimuli. According to the patient's medical history, the diagnosis of situs inversus was made incidentally at the age of 10 years, when a chest radiograph, performed during evaluation for a mild upper respiratory tract infection, revealed a mirror-image orientation of the thoracic organs. As stated by the patient, there were no preceding symptoms indicative of an underlying anomaly, and subsequent clinical assessment confirmed the absence of any additional congenital abnormalities. Also, the patient exhibited no cardiopulmonary complaints such as persistent cough or nasal congestion. In the present case report, situs inversus was found in association with dextrocardia. Confirmation of the diagnosis was achieved through chest radiography (Figure 1), and echocardiography revealed no structural cardiac abnormalities.

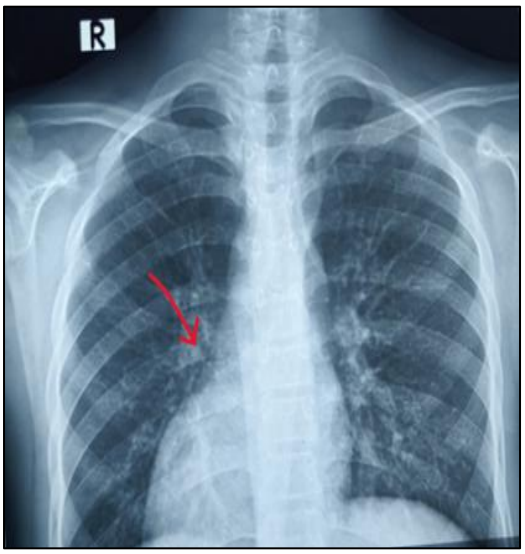

Figure 1: Radiograph of chest depicting dextrocardia.

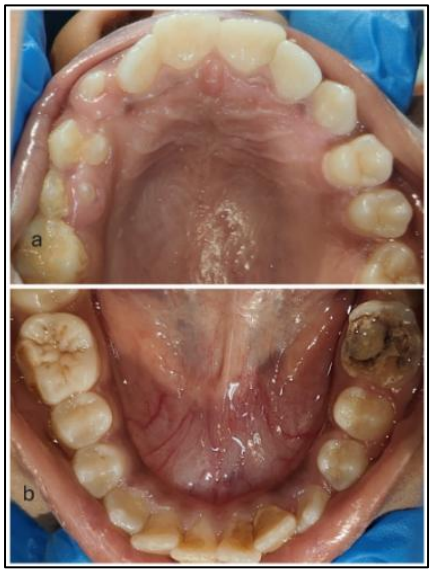

Figure 2: (a) Intraoral view of maxillary arch. (b) Intraoral view of mandibular arch showing carious tooth 36.

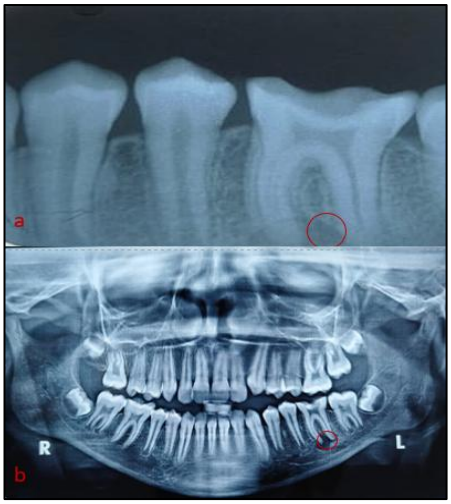

Figure [3]: (a) Pre-operative radiograph of tooth 36. (b) Orthopantomogram.

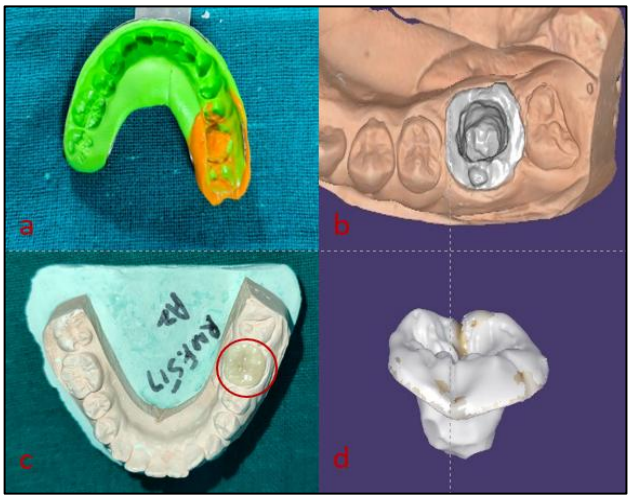

Figure 4: (a) Elastomeric impression after crown preparation. (b) Digital image of prepared crown using CAD software. (c) Endocrown fabricated with CAD-CAM on cast. (d) monolithic zirconia endocrown.

On intraoral examination, dental caries was noted in relation to tooth number 36, presenting as an extensive distinct cavity with visible dentin, consistent with ICDAS Code 6 and suggestive of possible pulpal involvement (Figure 2). Radiographic evaluation revealed extensive carious destruction involving the pulp tissue in tooth 36, reflecting an advanced stage of decay. A distinct periapical radiolucency was also noted, suggestive of chronic apical periodontitis likely secondary to pulpal necrosis and sustained periapical infection (Figure 3). Based on the clinical presentation and radiographic evidence, a diagnosis of irreversible pulpitis was established in the left mandibular first molar, indicating the need for root canal treatment.

Following physician consent for dental treatment under local anesthesia, root canal therapy was completed for

tooth 36. The tooth was then restored using an endocrown, a conservative full-coverage restoration that derives retention from the pulp chamber, thereby preserving a greater amount of natural tooth structure. The restoration was designed and milled using CAD-CAM (Computer-Aided Design and Computer-Aided Manufacturing) technology, ensuring high precision and a custom fit. This technique provided a durable, esthetic, and functionally stable outcome (Figure 4). To enhance overall oral health, a thorough oral prophylaxis was carried out. Furthermore, pit and fissure sealants were strategically applied to teeth 16, 26, and 46 to provide preventive protection against the development of occlusal caries.

Prosthetic management

An endocrown is a type of dental restoration used to restore endodontically treated (root canal treated) teeth, especially molars and premolars that have significant loss of tooth structure. It is considered a conservative and effective alternative to traditional post-and-core crowns.[7] Endocrowns represent a reliable restorative option for endodontically treated teeth, with both in vitro and in vivo studies showing success rates comparable to conventional techniques. Unlike traditional crowns, endocrowns rely primarily on adhesive cementation for retention, necessitating preparation designs suited to bonding. Their fabrication typically involves materials compatible with resin cements, such as lithium disilicate or composite resin. However, proper case selection is essential, as specific indications and contraindications must be considered to ensure clinical success.[8]

From a biomimetic standpoint, preserving tooth structure is critical for maintaining optimal biological, mechanical, adhesive, functional, and esthetic outcomes. Endocrowns offer a conservative restorative option by eliminating the need for root dentine removal, thereby minimizing recontamination risks and facilitating easier retreatment in case of endodontic failure. The natural saddle-shaped pulp floor in molars, combined with advanced adhesive materials, provides inherent stability without the need for additional preparation or posts, which can compromise tooth integrity.

Unlike conventional restorations composed of materials with varying elastic moduli that create multiple interfaces and potential stress concentrations, the monoblock structure of endocrown allows for more favorable stress distribution. Furthermore, supragingival margins support improved plaque control, clinical assessment, and periodontal health.[9] The key features of endocrowns as shown in Table 1.

Table 3: Key features of endocrown.

Features	
Monolithic structure	Made from a single block of ceramic or composite material (often lithium disilicate like *E. max*)
No post	Unlike traditional restorations, it doesn't require a post into the root canal; instead, it uses the pulp chamber for retention
Adhesively bonded	Cemented with adhesive resins for strong bonding to the tooth
Conservative	Preserves more natural tooth structure compared to traditional crown preparations

DISCUSSION

Situs inversus totalis is a veritably rare natural condition characterized by the complete glass- image reversal of thoracic and abdominal organs. It has two main forms situs inversus totalis and situs inversus with levocardia.[10] Aristotle was the earliest to document the phenomenon of situs inversus in living beings, a concept that would later be expanded upon by Leonardo da Vinci, first to recognize its association with dextrocardia. In 1788, Matthew Baillie handed a whole description of a complete glass- image reversal of thoracic and abdominal organs. Vehsemeyer was first to observe the transposition of the viscera on radiographs.[11] A mutation on murine chromosome 12 has been linked to the absence of a specific protein, although the exact cause remains unclear. Other possible contributing factors include motherly diabetes, cocaine use, conjoined twinning, and an autosomal recessive gene with deficient penetrance.[12]

From a dental and medical perspective, special considerations are demanded when managing cases with SIT. Affected individualities may have associated natural heart blights, similar as discordant atrioventricular (AV) and ventriculo-atrial (VA) connections, atrial situs solus, or congenitally corrected transposition of the great arteries (TGA).[5,10] Redundant care is needed during surgical procedures due to the atypical organ positioning. Situs inversus is frequently accompanied by primary ciliary dyskinesia (PCD). In this condition, the cilia are not performing adequately which leads to intermittent respiratory infections and bronchiectasis. In addition, males may suffer from gravidity secondary to sperm dyskinesia.[13] Around 20% of individuals with dextrocardia and situs inversus present with Kartagener's syndrome, characterized by the triad of reversed organ positioning, persistent sinus inflammation, and bronchiectasis, all stemming from defective ciliary motility. One major threat associated with situs inversus with dextrocardia is misapprehension of appertained pain

during medical extremities like myocardial infarction, cholecystitis, or appendicitis, where pain may manifest on the "wrong side". Feting the presence of condition will help in preoperative assessment and will simplify interpretation of "abnormal ECG" in case of any postoperative exigency.[5] The radiographic examination plays important part in the detection of situs inversus totalis and its associated conditions. Traditional imaging modalities like ultrasound (US) or plain film X-ray are generally the first choice of individual imaging. Advanced imaging modalities like computer tomography (CT) or glamorous resonance imaging (MRI) can also be used to assess fine anatomical details and possible pathological findings. In some cases, nuclear drug imaging, similar as cardiac SPECT, can be employed though it requires careful adaptation, like displacing of the sensor due to the reversed deconstruction.[4]

In our present case report, situs inversus was associated with dextrocardia. The opinion was verified with casket radiography. No cardiac anomalies were linked on echocardiography. While our literature review revealed several cases of situs inversus associated with aglossia, our patient presented no such findings, with neither hypoglossia nor aglossia observed.

CONCLUSION

SIT is a rare anatomical anomaly that can present challenges in clinical practice, particularly in surgical procedures. Although it does not necessarily cause specific complications or symptoms by itself, it can complicate the diagnosis and management of certain medical conditions. This case report presents a successful application of an endocrown restoration in a patient with situs inversus totalis, emphasizing the adaptability of advanced restorative techniques in individuals with uncommon anatomical conditions. The use of CAD/CAM technology enabled high-precision design and fabrication, resulting in a restoration that met both functional and esthetic demands. This clinical experience not only underscores the efficacy of conservative, digitally guided dental solutions but also enriches current understanding of tailored treatment planning in patients with rare systemic anomalies, thereby advancing knowledge in both restorative and special care dentistry.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Bisht H, Khatri A, Tyagi R, Khandelwal D, Yangdol P, Kumar A, et al. Innovative endocrown restoration in a situs inversus totalis patient using CAD/CAM technology. Int J Contemp Pediatr 2025;12:1563-6.