Received: 2 May 2023 | Revised: 1 June 2023 | Accepted: 11 June 2023

DOI: 10.1002/ccr3.7603

CASE REPORT

Clinical Case Reports Open Access WILEY

Solitary median maxillary central incisor in association with taurodontism and oligodontia: A case report

Yasmin Sheikhhassani[1] | **Arash Sarrafzadeh**[2] | **Soheila Jadidi**[3]

[1]Department of Pediatric Dentistry, School of Dentistry, Arak University of Medical Sciences, Arak, Iran

[2]Department of Oral and Maxillofacial Surgery, School of Dentistry, Arak University of Medical Sciences, Arak, Iran

[3]Dentistry Student, School of Dentistry, Arak University of Medical Sciences, Arak, Iran

Correspondence
Soheila Jadidi, School of Dentistry, Arak University of Medical Sciences, Arak, Markazi Provenience, Iran.
Email: s.jadidi.s@gmail.com

Key Clinical Message

In this case report, we have highlighted the probability of coinciding multiple dental anomalies, such as SMMCI, taurodontism, and oligodontia. Thus, further research is necessary to establish a correlation between oligodontia, taurodontism, and SMMCI.

KEYWORDS

central incisor, congenital abnormalities, single upper central incisor, taurodontism

1 | INTRODUCTION

Solitary median maxillary central incisor (SMMCI) with an estimated incidence of 1:50000 live births, is a rare developmental disorder, and it primarily affects females.[1,2] This anomaly may occur as an isolated trait or as a part of SMMCI syndrome (SMMCIS), wherein midline structures of the head, such as the central incisor in the maxilla, nasal airways, frenum, incisive papilla, and palate may be affected.[2,3] Moreover, developmental deficiencies in midline structures of the body, including hypotelorism, microcephaly, congenital nasal malformation, cleft lip, and/or palate have also been reported.[4,5] Autosomal dominant genetic origin and Sonic Hedgehog mutation have been linked to SMMCI.[5,3] It is worth noting that about 37% of cases with SMMCI have a history of preterm birth and low birth weight.[4] Additionally, several concomitant syndromes such as ectodermal dysplasia, oromandibular limb hypogenesis syndrome type 1, and autosomal dominant holoprosencephaly, have been associated with SMMCI.[1,5] Other concurrent anomalies, including hemifacial microsomia and fused mandibular incisor in primary dentition, have also been observed along with SMMCI.[3]

2 | CASE PRESENTATION

A 7-year-old female patient visited the pediatric department with complaining of pain in the right deciduous mandibular second molar. Radiographic examination revealed the presence of a solely permanent maxillary central incisor located in the midline, as shown in Figure 1.

The patient's medical and dental history indicated no medication during pregnancy, and the patient was born preterm at 28 weeks of pregnancy gestation. The parents reported a non specific upper respiratory tract disorder in the patient's neonatal period. No familial history or other developmental anomalies were identified, and there was no history of trauma to the primary dentition.

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.
© 2023 The Authors. *Clinical Case Reports* published by John Wiley & Sons Ltd.

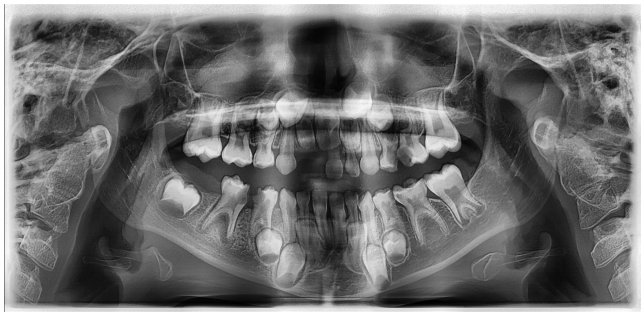

FIGURE 1 Panoramic radiograph. Note the solitary median maxillary central incisor located exactly in the midline of maxillary alveolus.

Upon examination, the patient appeared small for her age in weight and height measurements below the 5th and 50th percentile, respectively (16 kg and 120 cm).

The extraoral evaluation confirmed a distinct philtrum, asymmetric facial thirds, midfacial deficiency, slight asymmetry in the nasal septum and narrow nasal cavity, as shown in Figure 2. The patient presented with audible wheezing during breathing.

Profile examination revealed retrognathic chin, convex profile, and slight nasal bone depression (Figure 3).

Intraoral examination indicated that the patient was in the mixed dentition stage, with the SMMCI unerupted. The patient exhibited a normal labial frenulum of the upper lip and a narrow palate. The mother reported the exfoliation of the right deciduous maxillary central incisor. However, radiographic investigation revealed that the exfoliated tooth was the right deciduous maxillary lateral incisor, and a congenital dental abnormality known as SMMCI, was also present during the deciduous dentition stage. The remaining space and position of the left deciduous maxillary central incisor confirmed this finding.

Panoramic radiography revealed taurodontism in teeth numbered 16, 26, 36, 55, 64, 65, 74, 75, 84, and 85, as well as missing teeth numbered 17, 25, 27, 35, 37, 45, and 46 in the FDI numbering system.

The presence of a symmetrical single maxillary central incisor can be attributed to four differential diagnoses, namely premature tooth loss due to trauma, hypodontia, mesiodens, and SMMCI.

After reviewing the medical and dental history, conducting the radiographic evaluation, and examining extraoral evidence, a diagnosis of solitary median maxillary in association with oligodontia, taurodontism, and midfacial deficiency was made.

The parents were informed about the condition, and various treatment plans were explained. These included using orthodontics expansion appliances at an appropriate age to provide sufficient space, move the SMMCI to one side, and replace the other incisor using bridges, Maryland retainers or single-tooth implants to achieve esthetics.

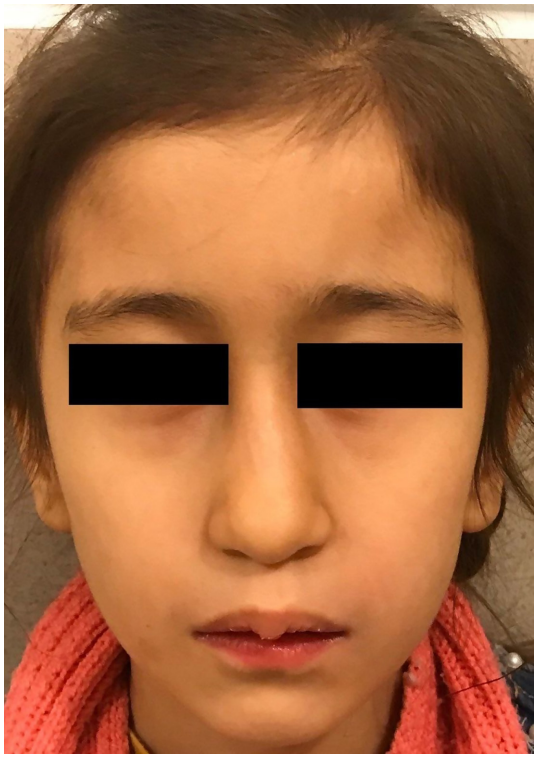

FIGURE 2 Frontal view. Distinct philtrum and nasal septum deviation is noticeable.

3 | DISCUSSION

As mentioned previously, there are four possible differential diagnoses for the presence of a symmetrical single maxillary central incisor. These include premature tooth loss due to trauma, hypodontia, mesiodens, and less commonly, SMMCI.[6] To rule out tooth loss related to trauma or a missing tooth, it is critical to determine if the tooth is precisely located in line with the mid-palatal raphe and has similar morphology to natural permanent central incisal teeth, without any history of trauma.[6] The absence of other teeth can also lead to ruling out mesiodens.[7]

SMMCI is a rare developmental disorder and may be a symptom or in association with other severe congenital or developmental anomalies and known syndromes, including congenital heart disease, microcephaly, short stature, hypopituitarism, oromandibular-limb hypogenesis type 1, velocardiofacial syndrome, ectodermal dysplasia,[6] hemifacial microsomia,[3] autosomal dominant holoprosencephaly,[5] nasal pyriform aperture stenosis, choanal

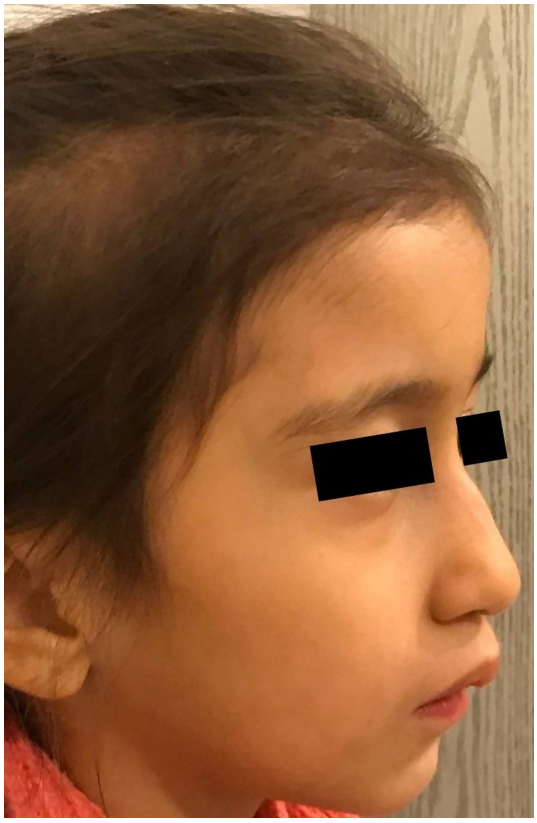

FIGURE 3 Profile view. Retrognathic chin and nasal bone depression is visible.

atresia, and mid-nasal stenosis.[2] It is important to diagnose SMMCI early,[8] as it can present in the primary and secondary dentition.[6]

In the case of isolated SMMCI, the patient concern may be aesthetics. A teamwork approach between orthodontists and prosthodontists would be necessary to achieve the desired results.[2]

Taurodontism can appear either as an isolated anomaly or in association with various syndromes and developmental anomalies, including ectodermal dysplasia and oral-facial-digital type 2.[9] Teeth affected by taurodontism have distinct morphological characteristics, such as large pulp chambers with apically displaced furcation, and greater apico-occlusal pulp chamber height than normal teeth, with minimal constriction at the level of cementoenamel junction.[9] Diagnosis of taurodontism is typically made through radiographic findings. Teeth affected by taurodontism may present unexpected challenges during various dental procedures, such as extractions due to the apically shifted furcation,[9] and endodontic treatments regarding complex root canal anatomy. They may also affect prosthetic and orthodontic procedures due to less tooth stability as an abutment.[10,9] Therefore, multiple diagnosed taurodont teeth in the presented case may create challenges during further orthodontic treatments.

4 | CONCLUSION

It is crucial to diagnose SMMCI early because of possible concomitant developmental disorders and associations with other abnormalities. As a result, regular long-term follow-up by a multidisciplinary team may be necessary for individuals with an SMMCI diagnosis. The correction of aesthetic deformity associated with isolated SMMCI may require maxillary expansion by an orthodontist followed by dental implant insertion. Therefore, early diagnosis and referral would provide more favourable outcome.

AUTHOR CONTRIBUTIONS

Yasmin Sheikhhassani: Conceptualization; data curation; investigation; project administration; validation. **Arash Sarrafzadeh:** Supervision; visualization; writing – review and editing. **Soheila Jadidi:** Writing – original draft; writing – review and editing.

CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest to declare.

DATA AVAILABILITY STATEMENT

The data that supports the findings of this study are available in the supplementary material of this article.

CONSENT

Written informed consent was obtained from the patient's father to publish this report in accordance with the journal's patient consent policy.

ORCID

Yasmin Sheikhhassani https://orcid.org/0000-0003-2898-2723
Arash Sarrafzadeh https://orcid.org/0000-0003-0026-6821
Soheila Jadidi https://orcid.org/0000-0002-7307-6108

REFERENCES

1. Shilpa G, Nuvvula S, Gokhale N, Yamini V. Concomitant solitary median maxillary central incisor and fused right mandibular incisor in primary dentition. *Contemp Clin Dent.* 2012;3(suppl 2):S203-S205.
2. Negi A, Negi A, Mohanan M. Solitary median maxillary central incisor syndrome: a rare entity. *J Oral Maxillofac Pathol.* 2020;24(2):402.
3. Utreja A, Zahid SN, Gupta R. Solitary median maxillary central incisor in association with hemifacial microsomia: a

rare case report and review of literature. *Contemp Clin Dent*. 2011;2(4):385-389.
4. Mustafa MM, Zakirulla M, AlShahrani I, Togoo RA, Alkahtani ZM, Ain TS. Clinical evaluation of solitary median maxillary central incisor syndrome. *Case Rep Dent*. 2019;2019:2637825.
5. Li J, Liu D, Liu Y, Zhang C, Zheng S. Solitary median maxillary central incisor syndrome: an exploration of the pathogenic mechanism. *Front Genet*. 2022;13:780930.
6. Nalawade TM, Mallikarjuna RM, Sogi HPS, Bhat KG, Kumbar VM. Solitary median maxillary central incisor: a case report with 3-year follow-up and literature review. *Contemp Clin Dent*. 2021;12(3):324-327.
7. Mallya SM, Lam EWN. *White and Pharoah's Oral Radiology: Principles and Interpretation*. 8th ed. Mosby; 2018.
8. Chandrasekaran D, Yezdani A, Tajir F, Saravanan B, Rajasekar L. Solitary median maxillary central incisor: a case report of a rare dental anomaly. *J Pharm Bioallied Sci*. 2015;7(suppl 1):S307-S308.
9. Dineshshankar J, Sivakumar M, Balasubramanium AM, Kesavan G, Karthikeyan M, Prasad VS. Taurodontism. *J Pharm Bioallied sci*. 2014;6(suppl 1):S13-S15.
10. Mohan RP, Verma S, Agarwal N, Singh U. Taurodontism. *BMJ Case Rep*. 2013;2013:bcr2012008490.

How to cite this article: Sheikhhassani Y, Sarrafzadeh A, Jadidi S. Solitary median maxillary central incisor in association with taurodontism and oligodontia: A case report. *Clin Case Rep*. 2023;11:e7603. doi:10.1002/ccr3.7603