

Prevalence of Microdontia in People with Down Syndrome: A Rapid Review

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Abstract

Down syndrome is a genetic chromosomal disorder caused by errors in cell division that results in the presence of an extra copy of chromosome 21. Patients with Down syndrome show various physical characteristics, including abnormalities in craniofacial, extremity and oral cavity. Previous studies have suggested that microdontia is an oral manifestation of Down syndrome.

The purpose of this study was to determine the prevalence of microdontia in patients with Down syndrome. The study was conducted using the rapid review method of articles describing the prevalence of microdontia in Down syndrome patients. The databases used are PubMed, ScienceDirect, and Google Scholar with filters for 2007-2022. PRISMA (Preferred Reporting Items for Systematic Review and Meta-analysis) and SORT (Strength of Recommendation Taxonomy) analyzes were used in the screening and selection of feasibility studies. A total of 9 relevant articles that met the inclusion criteria were reviewed in this study. Two of them compared Down syndrome patients and non-Down syndrome control groups.

The prevalence rate of microdontia in Down syndrome shows variation in this study. Several articles reviewed stated that there was no significant difference between male and female microdontia patients in Down syndrome. This study shows that the prevalence of microdontia in patients with Down syndrome ranges from 13% to 47.7%.

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Introduction

Down syndrome known as Trisomy 21 is a genetic chromosomal disorder caused by an error in cell division that results in the presence of an extra copy of chromosome 21.¹ Down syndrome was first described by John Langdon Down in 1866.² The World Health Organization (WHO) estimates that there are 8 million people with Down syndrome worldwide.³ Down syndrome is one of the most common chromosomal disorders, occurring in one in 700-1000 live births, and the most common cause of mental retardation.⁴ Maternal age is a significant incident factor; the disease occurs in 1 in 1,550 live births in women under the age of 20, difference in woman over 45 with the accidence 1 in 25 live births.⁵

Patients with Down syndrome show a variety of physical characteristic findings, these features include abnormalities in the craniofacial, extremities and oral cavity.⁶ Craniofacial anomalies in people with Down syndrome accompany systemic manifestations along with a lack of intellectual development.² The most obvious feature of Down's syndrome is the underdeveloped middle of the face, resulting in a small nose with a flat nose bridge, prognathic mandible, and developing a class III dental and skeletal relationships caused by growth deficiency in maxilla.^{2,7} Clinically, Down syndrome is also characterized by brachicephaly with a flat back of the head, smaller head, short ears, deep epicanthic folds, feet showing sandal gap, simian crease in the hands, neurological changes, structural cardiopathy, respiratory problems, increased risk of infection, increased risk of leukemia, dental anomalies, orofacial dysmorphology, impaired bone growth and generalized muscle hypotonia.³ Down syndrome patients also show a high prevalence of gingivitis and periodontal disease.⁸ A study conducted in

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Brazil reported that in most cases there is a high incidence of dental anomalies in which the same individual has more than one dental anomaly. Various types of anomalies among research subjects were quite high, those are taurodontism, true anodontia, microdontia, and impacted teeth.⁹ Down syndrome patients have specific orofacial features such as open bite, macroglossia, hypodontia, anodontia, drooling, fissured tongue, malocclusion, low dental caries, poor oral hygiene, delayed tooth eruption, small maxilla, retained primary teeth, and microdontia.⁵

Approximately 35-55% of Down's syndrome patients have extra small teeth or microdontia.¹⁰ Microdontia is a dental disorder in the form of teeth that are smaller than normal teeth.¹¹ In 2007, Moraes et al conducted a study on 49 people with Down syndrome and found that 14.28% had conical teeth. The study of tooth size dimensions conducted by Abeleira et al. to 40 people with Down syndrome stated that the teeth in people with Down syndrome have a size that is smaller than the size of normal teeth. A similar study was also conducted by Coughi et al. to 105 people with Down's syndrome and 17 of them had microdontia.^{5,12} The prevalence of microdontia is estimated to be between 25% and 55%. Although it can affect primary and permanent teeth, microdontia is more common in permanent teeth and can cause abnormal spacing.¹² Microdontia can cause a diastema between the involved tooth and the adjacent tooth so that it has the potential to become an area for trapped food debris or food impaction that has the potential to cause caries and periodontal disease. The size of the teeth that are smaller than the surrounding teeth can also affect the aesthetic aspect.¹³

The purpose of this study was to determine the prevalence of microdontia in patients with Down syndrome. The initial search conducted by the author on a database search engine showed that there were not many studies on the prevalence of microdontia in people with Down's syndrome in the form of literature studies. So the authors are interested in conducting a study on the prevalence of microdontia in people with Down syndrome using the rapid review method.

Materials and methods

The research was conducted using the

rapid review method. The materials used are online articles. The articles in this study were obtained by searching through PubMed, ScienceDirect, and Google Scholar. The study was conducted from March to May 2022.

The search and selection of articles in the electronic database refers to the Preferred Reporting Items for Systematic Review without Meta-Analyses (PRISMA). Article searches are carried out by entering keywords in each search engine with a publication year limitation of 2007-2022. The keywords used to search the articles were ((microdontia) OR (conical teeth)) OR (peg shaped teeth)) OR (dental anomalies)) AND (prevalence) AND (down syndrome)) OR (trisomy 21)). The inclusion criteria used were full articles discussing research on the prevalence of microdontia in Down syndrome, accessible full-text articles, English or Indonesian articles. The exclusion criteria are articles in the form of case reports and articles using a review study design.

Results

A total of 77 articles were identified through a search on PubMed, 165 articles were identified through a search on ScienceDirect and 1,540 articles were identified through a search on Google Scholar, bringing the total number of articles identified to a total of 1,782 articles. Duplication checks were carried out so that 1,757 articles were obtained.

The first screening is done by reading the title and abstract. A total of 1,732 articles were selected because they were irrelevant and did not match the inclusion criteria, so that 25 articles were obtained for further screening. The next screening was done by reading the contents of the entire text, 16 articles were selected because they were not relevant, so 9 articles were obtained for review. These relevant articles are then re-filtered based on their respective study designs to see the quality or level of scientific evidence (level of evidence) using SORT (Strength of Recommendation Taxonomy). Based on the SORT there were 3 levels of study quality, with level 1 being the highest quality.¹³ There were eight articles with cross-sectional study design, and one article with a retrospective study design. Based on SORT, articles with cross-sectional and retrospective study designs are at level 2 (limited-quality patient-oriented evidence). The results of the level of evidence

assessment in the 9 articles show that the articles are relevant and will be reviewed. The results of the search and study selection in this review are described in Figure 1. A total of 9 articles that met the inclusion criteria were obtained, with the general characteristics of the selected studies listed in Table 1. The results of studies conducted on the prevalence of microdontia in Down syndrome are presented in Table 2.

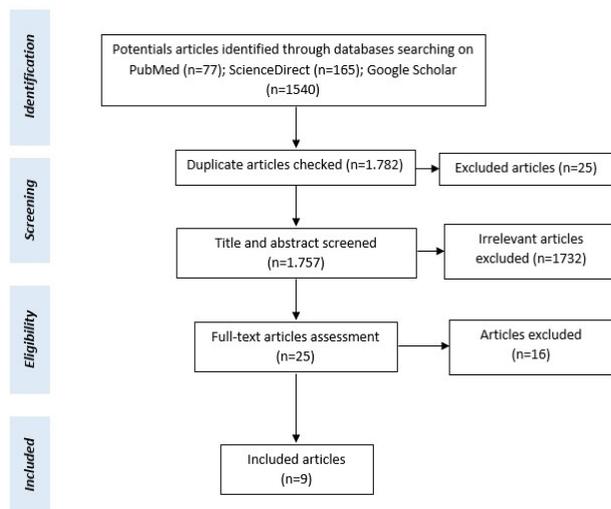


Figure 1. The results of the search and study selection.

The studies included in this study were published from 2008 to 2019, with a total of 9 articles. There was one article published in 2008¹⁴, two articles in 2014^{15,16}, three articles in 2016¹⁷⁻¹⁹, one article in 2018²⁰, and two articles in 2019^{21,22}. A total of two studies were conducted in India^{14,15}, two articles in Indonesia^{20,21}, while the other five articles were each conducted in Romania¹⁷, Turkey¹⁶, Saudi Arabia²², Brazil¹⁸, and Spain¹⁹.

The articles reviewed had a cross-sectional study design in eight articles^{14,15,17-22}, and a retrospective study design in one article¹⁶. The total sample of Down syndrome patients studied in all articles amounted to 788 people. There are two articles with a control group sample, that were Mayoral Trias M.A. et al. (2016) with 42 control groups without Down syndrome, and Hoda Fansa et al. (2019) with 50 control groups without Down's syndrome²². Eight articles grouped participants by sex^{14-19,21,22}, while one article did not mention it²⁰.

Research by Severin et al. (2016) were

conducted on different types of Down syndrome, that was the full trisomy type, Robertsonian type, and mosaic type.¹⁷ The other eight articles did not mention the type of Down syndrome in the participants.^{14-16,18-22} There was one articles with patient exclusion criteria currently undergoing dental treatment such as crowns, bridges, or orthodontic treatment.²¹ One article excluded patients with medical disorders.¹⁵ The lowest reported prevalence of microdontia was 13%¹⁶ and the highest was 47.7%²¹. A total of three articles did not mention the teeth with microdontia^{15,18,22}, and five articles mentioned the teeth with the most microdontia.^{14,16,17,19-21} The articles that were studied varied, the research was carried out on primary, mixed, and permanent teeth¹⁷; primary and permanent teeth^{15,18,22}; and permanent only.^{16,20,21}

Discussion

Based on the data in Table 2, the prevalence of microdontia in people with Down syndrome ranges from 13% to 47.7%. The different results from each study may be due to differences in the number of samples, data collection methods, age distribution, and the country in which the study was conducted. High results were found in Indonesia and India.^{15,21} Previous studies were conducted in several different countries, where different environmental factors may result in different prevalence.²⁰ Several studies that have been conducted have found that microdontia is more common in Asian and Mongoloid people compared to other races.²³ Research conducted by Mayoral Trias et al.¹⁹ and Hoda Fansa et al.²² stated that the prevalence value in the control group or the normal population was lower than the group with Down syndrome. The control group in the study of Mayoral Trias et al.¹⁹ who had microdontia was 2.4%, and 24.4% of people with Down syndrome. Hoda Fansa et al.²² stated that in their study, none of the control groups in their study had microdontia (0%), while 34% of participants with Down syndrome had microdontia. The results of these two studies showed that microdontia in patients with Down syndrome was more common than the normal population as a control group.

There was only one article that mentioned the type of Down syndrome that the participants had.¹⁷ Severin et al. stated in his study that microdontia occurred in 9 of 45 participants with

full-type Down syndrome, 4 of 4 participants with the Robertsonian type, and 2 of 2 participants with the mosaic type.¹⁷

The results showed that there was no statistically significant difference between male and female microdontia cases with Down syndrome.^{14,18,19} Research conducted by Asokan et al. found microdontia in its participants to occur in 10 of 45 women, and 7 of 57 men.¹⁴ Sekerci et al. reported that 11 of 112 men and 18 of 104 women had microdontia.¹⁶ Cuoghi et al. in his study stated that microdontia occurred in 10 of 61 men, and 7 of 44 women.¹⁸ Hoda Fansa et al. in his study stated that 11 out of 30 men and 6 out of 20 women with Down syndrome had microdontia.²² Based on the figures shown by the studies that have been presented, the incidence of microdontia in men and women was not significantly different even though some of these studies noted rates in men were higher than women and or vice versa.

There are several articles that do not mention which teeth have microdontia.^{14,15,18,22} Research conducted by C. Dara Manja et al. found that there was a variety of teeth that had microdontia, from the maxillary central incisors to the mandibular second premolars.²⁰ Sekerci et al. stated in their study, the teeth with the most microdontia were the maxillary lateral incisors.¹⁶ The same finding was also obtained in this study conducted by Mayoral Trias et al. where microdontia occurs more frequently in the maxillary lateral incisors.¹⁹ Based on a study conducted by Luly Anggraini et al. half of the participants had tooth size anomalies, and the most common tooth size anomaly found in his study was microdontia. The microdontia that occurred in the participants were true generalized microdontia which affected 16 subjects, and localized microdontia in 67 subjects. In this study, the teeth most frequently affected by microdontia were the maxillary lateral incisors, followed by the maxillary second premolars and the maxillary second molars.²¹ The different prevalence of anomalies in various tooth regions can be attributed to the development time, teeth that develop later show more variability than teeth that develop earlier in the same region. The maxillary lateral incisors are highly variable teeth, possibly due to environmental influences.²⁴ Sekerci et al. in his research mentioned microdontia which was found to occur bilaterally.¹⁶ The same thing was also stated by

Luly Anggraini et al. in his study that of all participants who experienced microdontia more occurred bilaterally than unilaterally.²¹ Severin et al. did not describe which teeth had microdontia, but the study only stated that the microdontia found in the participants were mostly localized microdontia.¹⁷ Three studies reported that microdontia occurred in the maxilla or occurred more frequently in the maxilla than in the mandible.^{16,19,21} Another study reported that microdontia that occurred in participants was more occurred in mandible.²⁰

Based on the data in table 2, it can be seen that the total number of samples of Down syndrome patients from the article reviewed was 788 people with an average age of 4 to 53 years and the study was located in India^{14,15}, Indonesia^{20,21}, Romania¹⁷, Turkey¹⁶, Saudi Arabia²², Brazil¹⁸, and Spain¹⁹ from 2008 to 2019. From the studies reviewed, several of them showed that microdontia was the most common dental disorder found in people with Down syndrome.^{18,20-22} Luly Anggraini et al. mentioning individuals with Down syndrome in their study showed a high prevalence of dental anomalies, with microdontia being the most common anomaly.²¹ O.A. Cuoghi et al. explained in his study there was a high prevalence of dental anomalies evaluated with Down's syndrome. The most frequently observed dental anomaly was microdontia.¹⁸ Research conducted by C. Dara Manja et al. showed that microdontia was the second most common finding in Down syndrome after hypodontia.²⁰ The same thing was also found by Hoda Fansa et al. in his study where microdontia was ranked as the second most common dental anomaly among his research subjects.²²

Additional genetic material in Down syndrome causes delays in the physical and mental development of children, various developmental delays in patients with Down syndrome include variability in oral and dental development, (i.e. abnormalities in tooth eruption, structure, and shape).¹⁰ Microdontia in individuals with Down syndrome is associated with decreased mitotic activity of trisomy cells in developing tooth buds, which results in fewer ectomesenchymal cells to form dentin and fewer inner enamel epithelial cells in the DEJ to differentiate into ameloblasts and secrete the enamel matrix in the proliferative phase of the tooth bud when the crown is forming.²¹

Conclusions

The conclusion of this study is that the prevalence of microdontia in patients with Down syndrome is around 13% to 47.7%.

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Declaration of Interest

The authors report no conflict of interest.

Author	Title (Research Location)	Data collection method	Research methodology
Sharath Asokan et al (2008). ¹⁴	<i>Oral findings of Down syndrome children in Chennai city, India (India)</i>	Clinical examination	Descriptive cross-sectional study
Deepika Shukla et al(2014). ¹⁵	<i>Dentofacial and Cranial Changes in Down Syndrome (India)</i>	Clinical examination	Cross-sectional study
Ahmet Ercan Sekerci et al(2014). ¹⁶	<i>Prevalence of Dental Anomalies in the Permanent Dentition of Children with Down Syndrome (Turkiye)</i>	Photo taking & radiographic examination	Retrospective study
Emilia Severin et al(2016). ¹⁷	<i>Common, Rare, and Individual Oro-dental Findings in People with Down Syndrome (Romania)</i>	Clinical and radiographic examination	Cross-sectional study
Osmar Aparecido Cuoghi et al(2016). ¹⁸	<i>Prevalence of Dental Anomalies in Permanent Dentition of Brazilian Individuals with Down Syndrome (Brazil)</i>	Radiographic examination	Cross-sectional study
Mayoral Trias M.A. et al(2016). ¹⁹	<i>Comparative study of dental anomalies assessed with panoramic radiographs of Down syndrome and non-Down syndrome patients (Spain)</i>	Radiographic examination	Retrospective cross-sectional study
C. Dara Manja dan N. Azimi(2018). ²⁰	<i>The Use of Panoramic Radiography to Determine Prevalence of Dental Anomaly in Individuals with Down Syndrome (Indonesia)</i>	Radiographic examination	Cross-sectional study
Luly Anggraini et al(2019). ²¹	<i>Prevalence of Dental Anomalies in Indonesian Individuals with Down Syndrome (Indonesia)</i>	Clinical examination	Cross-sectional study
Hoda Fansa et al(2019). ²²	<i>The Prevalence of Oral and Dental Anomalies in Down syndrome Children in Western Region, Saudi Arabia (Saudi Arabia)</i>	Clinical examination	Cross-sectional study

Table 1. General study characteristics.

Author	Research sites	Control group	Samples	Prevalence of microdontia
Sharath Asokan et al(2008) ¹⁴	India	None	Total samples: 102 Age: 15 years and under F/M: 7/70	16.7%
Deepika Shukla et al (2014) ¹⁵	India	None	Total samples: 77 Age: 6-40 years old F/M: 7/70	45.5%
Ahmet Ercan Sekerci et al (2014). ¹⁶	Turkey	None	Total samples: 216 Age: average age 13.5 years old F/M: 104/112	13%
Emilia Severin et al (2016). ¹⁷	romania	None	Total samples: 51 Age: 2-31 years old F/M: 23/28	25.53%
Osmar Aparecido Cuoghi et al (2016). ¹⁸	Brazil	None	Total samples: 105 Age: 7-42 years old F/M: 44/61	16.19%
Mayoral Trias M.A. et al(2016). ¹⁹	Spanish	Yes	Total samples: 41 Down syndrome patients and 42 control group without Down syndrome Age: 9-13 years old, with mean age of patients Down syndrome 10.6 ±1.4 Mean age of control group 11.1 ±1.3 F/M: Down syndrome patient : 19/22 Control group without Down syndrome: 26/16	24.4%
C. Dara Manja dan N. Azimi(2018). ²⁰	Indonesia	None	Total samples: 12 Age: 12-26 years old, F/M: -	18.06%
Luly Anggraini et al(2019). ²¹	Indonesia	None	Total samples: 174 Age: 14-53 tahun, F/M: 70/104	47.7%
Hoda Fansa et al(2019). ²²	Saudi Arabia	Yes	Total samples: 50 Down syndrome patients and 50 control group without Down syndrome Age: 4-14 years old F/M: Down syndrome patients: 20/30 Control group without Down syndrome : 22/28	34%

Table 2. Sample characteristics and prevalence of microdontia in Down's syndrome.

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