

Dento-Occlusal and Skeletal Anomalies in Nigerian Individuals with Down Syndrome

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Abstract

Background: Dento-skeletal and occlusal anomalies have been associated with Down syndrome (DS). The aim of this study was to determine the dental, skeletal and occlusal anomalies in untreated Down Syndrome (DS) and untreated non-Down syndrome individuals aged 13 years and above.

Methods: Individuals with untreated DS were selected from special schools in Southwest Nigeria and untreated non-Down Syndrome (controls), were matched for age and sex from public schools. A questionnaire was used to collect data on socio-demographic characteristics and findings from oral examination carried out in their respective institutions. Descriptive characteristics such as Angle classification, missing or impacted teeth, crossbites, open bites, and other anomalies were recorded. Orthodontic treatment need was assessed using the Dental Aesthetic Index (DAI).

Result: There were 102 individuals in each group with mean age of 21 ± 6.2 years and 22.7 ± 7.6 years in the DS and non-DS groups respectively. Forty nine (48%) DS individuals compared with 90 (88.2%) non-DS individuals had Angle's Class I malocclusion, 1 (1%) of DS individuals had Angle's Class II Division 1 compared with 7 (6.9%) of non-DS individuals. About 3% of individuals with DS and non-DS presented with Angle's Class II Division 2 while 48.1% of individuals with DS presented with Angle's Class III compared with only 2% of non-DS individuals. Skeletal pattern III was found in 77.5% of DS individuals, while 92.3% of non-DS had skeletal pattern I. Class III incisal relationship was predominant in DS group (55.9%) compared with 2.0% of non-DS. Retained primary teeth were found in 31.4% of DS and in 5.9% of non-DS individuals while 39.2% of DS and 9.8% of non-DS had un-erupted teeth. Fifty two (51%) subjects in the DS group had anterior, posterior, unilateral and bilateral cross-bite compared with 3 (2.9%) of the non-DS individuals.

Conclusion: It was concluded from these findings that Angle's Class III malocclusion and severe dental and skeletal discrepancies were more predominant in the DS than in the non-DS subjects. It is therefore recommended that orthodontic treatment should be subsidized for individuals with Down Syndrome (DS) and encourages inclusion in orthodontic practice.

Key words: Down syndrome, Special needs, dento-skeletal, occlusal, malocclusion

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Introduction

Down syndrome (DS) is a well-known genetic syndrome named after John Langdon Down, the British doctor who first published and described the syndrome in 1866.¹ It was identified as a chromosomal disorder, specifically chromosome 21 trisomy by Jerome Lejeune in 1959.¹ Also referred to as Trisomy 21, the condition is characterised by generalized hypotonia, causing intellectual impairment and physical abnormalities including short stature and a broad facial profile. About 95% of the DS cases have an

extra chromosome 21, 3% have translocation and 2% exhibit mosaicism or partial trisomy.²⁻³

Down Syndrome (DS) leads to impairments in both cognitive ability and physical growth that range from mild to moderate developmental disabilities and therefore are more likely to require assistance for their disabilities. Such individuals with 'special needs' are therefore referred to as those with physical or developmental disability including Down Syndrome (DS), autistic spectrum disorder, medically compromised and those who may require special attention.⁴

The oral condition of individuals with special health needs have been reported in literature to be more prevalent and are influenced by various socio-demographic factors, including living conditions and severity of impairment when compared to people with closest age range.⁵⁻⁸

Down syndrome was initially believed to be very rare in Nigeria with an incidence of 1:865 per 1000 live births reported.⁹ This apparent low incidence might be the result of incomplete ascertainment at birth, high mortality in infancy and short period of case collection.⁹ The perception remains that a DS infant is still not openly accepted by parents and society, thus creating many ethical challenges in

treating such babies starting with foetal diagnosis of the disorder in the womb and moving through early stages after the birth of the baby.¹⁰

Dental characteristics have been described in children with DS and they include macroglossia (enlarged tongue), fissured tongue, underdeveloped maxilla, tongue thrusting, congenitally missing teeth, malocclusion, high arched palate and microdontia.¹¹⁻¹⁴ The skeletal features in DS include small midface, pseudoprognathism and high arched palate. A small oral cavity with relatively large tongue leads to mouth breathing, which is a common cause of chronic periodontitis and xerostomia.¹⁵ Individuals with DS are thus more vulnerable to loss of teeth from periodontal disease resulting in derangement of the dentition.¹⁶ Similarly, hypodontia is prevalent in individuals with DS and this leads to underdevelopment of alveolar bone, resulting in decreased maxillary length and bone height. These features tend to affect function, appearance and inclusion of people with DS into the society.¹⁶

In a recent report, 63% of the DS group had one or more missing teeth,¹⁴ while in a normal population study, a prevalence of missing teeth were reported in only 1.8% of the study population.¹⁸ Most frequently missing teeth in DS group were the maxillary and mandibular third molars; maxillary and mandibular lateral incisors, canines and mandibular left second premolar.

Special need individuals tend to have definitive malocclusion compared to general population of comparable age.^{14, 18-19} Usually mandibular overjet and anterior open-bite are associated with DS²⁰ which otherwise are very rare in a healthy population. Vigild,²¹ reported that among the DS individuals found in his study, 41% had mandibular overjet, 54% had mesial occlusion, 38% had frontal open bite and 65% had crossbite.²¹ In contrast however, deep bite and maxillary prognathism are very rarely observed in individuals with DS.²² A report of 65% of individuals with DS who presented with posterior crossbite was made by Musich.²³ The tendency towards Class III malocclusion is very high in individuals with DS. This was reported in 47% of the study population compared to 5% of the control group.¹³

The high incidence of malocclusion in these individuals remained present into old age, mainly due to lack of treatment and the need to employ non-conventional orthodontic treatment.¹⁸ Though it was concluded that the prevalence of malocclusion among people with disabilities is higher when compared with normal population,²⁴⁻²⁵ dental and

orthodontic commitment to patients with DS is less evident.²³

The Dental Aesthetic Index (DAI) is an occlusal index; universally acceptable, designed for use in epidemiological surveys to categorize malocclusion into groups according to the level and priority of orthodontic treatment need.²⁶ The DAI relates the clinical and aesthetic components to mathematically calculate a single score that reflects the malocclusion severity. By using cut-off points, the score is used to determine the need for orthodontic treatment. The DAI has been demonstrated to be simple, valid, reliable, and easy to use.²⁷⁻²⁹ Several previous studies have determined the prevalence of malocclusion and need for orthodontic treatment among persons with disabilities using DAI.^{25,30-34}

The main objective of this study was to determine the prevalence of malocclusion and need for orthodontic treatment among individuals with DS using the Dental Aesthetic Index (DAI) in Southwest region of Nigeria.

Materials and Methods

The study was conducted after obtaining ethical approval from Health Research Ethics Committee of the Lagos University Teaching Hospital, and all the institutions that participated in the study. Verbal and written consents were also sought from subjects and from subjects' parents/guardians. Only those with positive responses were examined. Institutionalized individuals in Southwest region of Nigeria and from selected institutions in Lagos, Ogun and Oyo states, clinically diagnosed with DS aged 13 years and above were included in this study.

A non-probability sampling method was used due to the rarity of the condition and the limited accessibility to affected individuals. A convenient sample size of 102 individuals with DS was therefore examined and an equal number of non-DS population was matched for age and sex and included in the study. All the individuals with DS who fulfilled the inclusion and exclusion criteria were invited to participate in the study. The following inclusion criteria were used: (1) diagnosed with DS at any level of intelligence quotient, (2) no history of previous orthodontic therapy, and (3) willingness to participate in the study. All individuals with cerebral palsy and cleft lip and palate, those with severe mental disability and were uncooperative during oral examinations and those not willing to participate were excluded. Refusal to sign the consent form by a parent/guardian also excluded the individual from the study.

Each subject with DS was seated on an ordinary

chair. The examiner (OJB) performed a simple clinical oral examination, without the use of dental probe and mirror to ensure cooperation. Other persons with DS, if present, were allowed to observe the procedure for others to gain their interaction, confidence, and cooperation. The clinical oral examination included dental, skeletal and occlusal parameters including molar relationship, skeletal relationship, overjet and overbite were assessed. Unerupted teeth, retained primary teeth and other dental anomalies were also noted during the examination. All items used in the examination were under strict infection-control procedures.

The DAI measurements were done by the same examiner (OJB) following the sequence of the DAI components (Appendix A). Measurements were done three times; the mean was used. Intra-examiner reliability was determined by reassessing 10 randomly selected DS individuals at a 1-week interval. Kappa analysis showed good intra-examiner reliability with a score of 1.

Other data collected were recorded on a data collection sheet and included age, gender, level of education and age of parents. Others were demographic information of the parents and family history of DS. This was done through a questionnaire completed by subjects, parent/guardian of subjects in the case of DS group.

The SPSS (Statistical Package for Social Science software for Windows version 16.0, (SPSS, Chicago, Ill) was used for data entry and analysis. Descriptive statistics were performed, including frequencies and percentages for categorical variables, means and standard deviations were calculated for quantitative variables.

Results

A total of 204 subjects participated in the study and this comprised two major groups. Group 1 consisted of 102 individuals with DS with mean age of 21.3 ± 6.2 years (Table I). Group 2 also consisted of 102 individuals in the non-DS with mean age of 22.7 ± 7.6 years (Table I). There was no statistical significant difference in age and gender in the two groups.

Most of the subjects' fathers were reported to be above 40 years, while most of them had education up to the tertiary level in all groups of study. The same trend was reported for the subjects' mothers and the level of education. The socio-demographic data of both parents and subjects were also recorded (Table II).

Table I: Demographic Characteristics of Subjects (N= 204)

Variable	DS N (%)	non-DS N (%)	χ^2	df	p
Age (Years)					
< 20	50 (49.0)	50 (49.0)	0.87	4	0.93
20-29	39 (38.2)	40 (39.2)			
≥ 30	13 (12.8)	12 (11.8)			
Mean age	22.7 ± 7.6	21 ± 6.2			
Gender					
Male	51 (50.0)	57 (55.9)	0.74	2	0.69
Female	51 (50.0)	45 (44.1)			
Total	102 (100.0)	102 (100.0)			

Table II: Demographic Characteristics of Parents of Subjects (N=204)

Variables	DS N (%)	non-DS
Father's age (Year)		
< 20	0 (0)	0 (0)
20-29	0 (0)	0 (0)
30-39	3 (2.9)	5 (4.9)
≥ 40	99 (97.1)	97 (95.1)
Mother's age (Years)		
< 20	0 (0)	0 (0)
20-29	3 (2.9)	3 (2.9)
30-39	7 (6.9)	14 (13.7)
≥ 40	92 (90.2)	85 (83.4)
Father's Education		
Primary	1 (1.0)	8 (7.8)
Secondary	34 (33.3)	48 (47.1)
Tertiary	67 (65.7)	45 (44.1)
Mother's Education		
No formal	1 (1.0)	3 (2.9)
Primary	3 (2.9)	8 (7.8)
Secondary	38 (37.3)	49 (48.0)
Tertiary	60 (58.8)	42 (41.2)
Family History of DS		
Yes	5 (4.9)	5 (4.9)
No	97 (95.1)	97 (95.1)
Total	102 (100.0)	102 (100.0)

Angle's Class I molar relationship was recorded in 49 (48%) and in 90 (80.2%) of the population of individuals with DS and non-DS respectively; Angle's Class II, division 1 molar relationship in only 1% and 6.9% of DS and non-DS groups respectively; Angle's Class II, division 2 in only 2.9% each in populations with DS and non-DS and Angle's Class III in 48.1% and 2.0% of DS and non-DS respectively. The correlation was found to be statistically significant ($p < 0.001$).

The population with DS predominantly had skeletal pattern 3 and those in the non-DS had predominantly skeletal pattern 1. Class III incisal

Table III: Distribution of Occlusal Pattern of Subjects in the Study Population (N=204)

Variables	DS N (%)	non--DS N (%)	Fisher's exact test p
Angle's Classification			
Class I	49 (48.0)	90 (88.2)	0.001*
Class II division 1	11 (1.0)	7 (6.9)	
Class II division 2	3 (2.9)	3 (2.9)	
Class III (True)	22 (21.6)	1 (1.0)	
Class III (Pseudo)	27 (26.5)	1 (1.0)	
Skeletal Pattern			
Class 1	22 (21.6)	94 (92.3)	0.0001*
Class 2	1 (1.0)	4 (3.9)	
Class 3	79 (77.5)	4 (3.9)	
Incisal Classification			
Class I	45 (44.1)	93 (91.2)	0.00*
Class II	0 (0)	7 (6.9)	
Class III	57 (55.9)	2 (2.0)	
Overbite			
Normal	9 (8.8)	79 (77.5)	0.001*
Increased	2 (2.0)	7 (6.9)	
Decreased	70 (68.6)	9 (8.8)	
Edge to edge	1 (1.0)	5 (4.9)	
AOB	20 (19.6)	2 (2.0)	
Overjet			
Normal	13 (12.7)	69 (67.6)	0.00*
Increased	5 (4.9)	23 (22.5)	
Decreased	84 (82.4)	10 (9.8)	
Crossbite			
Absent	50 (49.0)	98 (96.0)	0.00*
Anterior	36 (35.3)	2 (2.0)	
Posterior	7 (6.9)	1 (1.0)	
Unilateral	3 (2.9)	0 (0.0)	
Bilateral	6 (5.9)	1 (1.0)	
Unerrupted teeth			
Present	40 (39.2)	10 (9.8)	0.00*
Absent	62 (60.8)	92 (90.2)	
Supernumerary teeth			
Present	5 (4.9)	12 (11.8)	0.13
Absent	97 (95.1)	90 (88.2)	
Oral habits			
None	49 (48.0)	87 (85.2)	0.0001*
Digit	14 (13.7)	2 (2.0)	
Tongue thrusting	34 (33.3)	5 (4.9)	
Ant. resting posture	2 (2.0)	0 (0.0)	
Lip sucking	1 (1.0)	1 (1.0)	
Others	2 (2.0)	7 (6.9)	
Retained deciduous teeth			
Present	32 (31.4)	6 (5.9)	0.00*
Absent	70 (68.6)	96 (94.1)	
Tooth fracture			
Present	14 (13.7)	14 (13.7)	0.04
Absent	88 (86.3)	88 (86.3)	
Total	102 (100.0)	102 (100.0)	
Fisher's exact $p \leq 0.05$, $p \leq 0.05$			

relationship was predominant in the group with DS (55.9%) compared to 2.0% in non-DS. Decreased overbite was found in 68.6% of the group with DS, while normal overbite was found in 77.5% in non-DS. Anterior open bite was predominant (19.6%) in the population with DS and in only 2% in the non-DS (Table III).

Overjet was found to be normal in 67.6% of the non-DS while normal overjet was found in 12.7% of the group with DS. Overjet was found to decrease in 82.4% of the DS group in the study, and in 9.8% of the non-DS. Conversely, an increase in overjet was found in 4.9% and 22.5% of DS and non-DS respectively (Table III).

Fifty two individuals with DS in the study population had anterior, posterior, unilateral and bilateral crossbite while only 4 individuals in the non-DS group had crossbite. This was found to be statistically significant ($p < 0.001$, Table III).

Unerrupted teeth were found in 39.2% and 9.8% of the DS and non-DS groups respectively. Supernumerary teeth were found in 11.8% and 5% of non-DS and DS respectively. There was no statistical significance found ($p = 0.09$, Table III).

Oral habits were reported in 52.0% and 14.7% of individuals with DS and in the non-DS respectively. Tongue thrusting habit was seen to be predominant in the group with DS. This was found to be statistically significant ($p < 0.001$).

Retained deciduous teeth were recorded in 31.4% of individuals with DS, while only 5.9% of non-DS had retained deciduous teeth. This was found to be statistically significant ($p < 0.001$). Tooth fracture was reported in only 14 individuals (13.7%) each of DS and non-DS groups. This was also found to be statistically significant ($p < 0.001$) (Table III).

Dental anomalies observed in the DS group included peg shaped lateral incisors, Hutchinson's incisor, Talon cusp and mesiodens. In the non-DS group were reported Talon cusp and peg shaped lateral incisors (Table IV).

Table IV: Distribution of Dental Anomalies

Anomalies	DS N (%)	non-DS N (%)
None	89 (87.3)	99 (97.1)
Peg shaped lateral incisor	7 (6.9)	1 (1.0)
Hypoplasia	3 (2.9)	0 (0.0)
Hutchinson's incisor	1 (1.0)	0 (0.0)
Talon cusp	1 (1.0)	2 (2.0)
Mesiodens	1 (1.0)	0 (0.0)
Total	102 (100.0)	102 (100.0)

Fisher's exact $p = 0.05^*$

Normal or minor malocclusion was found to be 52% and 23.5% in non-DS individuals and in individuals with DS respectively. In this case, no need or slight need for treatment was required. Definite malocclusions or where elective treatment is required was found in 28.4% and 21.6% of non-DS and DS groups respectively; and severe malocclusion and orthodontic treatment highly desirable was recorded in 11.8% and 24.5% in non-DS and in those with DS respectively. Very severe or handicapping malocclusion and where treatment is mandatory was reported in 7.8% of the non-DS while 30.4% was found in DS group. This was found to be statistically significant (Table V). The mean DAI score in the two groups was found to be 32.1 ± 8.0 and 26.6 ± 5.8 respectively.

Table V: Distribution of Dental Aesthetic Index in Study Population

DAI	DS N (%)	non-DS N (%)
No treatment need	24 (23.5)	53 (52.0)
Treatment elective	22 (21.6)	29 (28.4)
Treatment highly desirable	25 (24.5)	12 (11.8)
Treatment mandatory	31 (30.4)	8 (7.8)
Mean	32.1 ± 8.0	26.6 ± 5.8

F statistic = 15.30, $p = 0.00^*$

Table VI: Comparing Dental Aesthetic Index in Subjects with Non-down Syndrome and with Down Syndrome

DAI	DS N (%)	non-DS N (%)	χ^2	p
No treatment need	24 (23.5)	53 (52.0)	30.01	0.00*
Treatment elective	22 (21.6)	29 (28.4)		
Treatment highly Desirable	25 (24.5)	12 (11.8)		
Treatment mandatory	31 (30.4)	8 (7.8)		
Mean	32.1 ± 8.0	26.6 ± 5.8		

Student's t statistic = 5.5, $p = 0.00^*$

Discussion

The association between physical abnormalities and mental impairment has been known for ages³⁵ and it has been affirmed that abnormal conditions of the skeleton occur with extreme frequency in amentia.³⁶ The findings in this study revealed that malocclusion is higher in Down syndrome population when compared to normal individuals (non-DS); and it is in agreement with previous authors.^{19,30-31,37-38} The normal population in this study

had predominantly Angle's Class I malocclusion. This is in agreement with the reports of other authors.^{25, 30, 39-43} The group with Down Syndrome however had more subjects with Angle's Class III

malocclusion than the non-DS group. This is comparable with other studies.^{13, 25, 30} Similar result was reported in Croatia, where Class III malocclusion was most frequently present in population of subjects with Down Syndrome.²²

Furthermore, skeletal pattern 1 was predominantly found in the non-DS population, whereas skeletal pattern 3 was reported to be found more in DS and much less in non-DS populations. This finding is also in agreement with reports of other studies^{1, 44} and similar result was reported for incisal relationship in DS population with a large number having reverse overjet or class III incisal relationship while the non-DS group had very few with Class III relationship.

African population has been known to have variation in reported cases of anterior open bite. The prevalence of anterior open bite has been reported in Nigerians to vary from 4.6% to 10%.^{39-40, 45} However, 1% of the study population. Moalence varies from 1.1% to 8.0%.⁴⁶⁻⁴⁷ A higher prevalence was reported in individuals with DS, than was reported in the non-DS. This finding is not consistent with previous studies in normal populations in Nigeria.^{30, 39-40, 43, 48} but in agreement with reports in a Sudanese population.⁴⁷ This was found to be statistically significant when the various groups were compared ($p=0.00^*$).

Majority of individuals in the DS group had decreased overjet, while overjet was decreased in a few in the 10% of non-DS. This accounted for the high percentage of anterior crossbite present in the former group.

Severe or handicapping malocclusion in this study was found more in the population with DS than in non-DS. This is in agreement with various previous studies.^{19, 25, 49-51} The student t test and the analysis of variance were found to be valid in both instances. Thus the proportion of unmet orthodontic treatment need in Nigeria is still relatively high particularly among the mentally challenged. This requires that priority should be given to individuals with DS in publically funded health programme. Enlightenment programmes should be organised among dentists, especially orthodontists, parents particularly those with children with special needs on the need and importance of aesthetics and oral function in the development of self-esteem of children with special needs.

Dental treatment, especially orthodontic treatment for individuals with special needs should be highly subsidised to encourage better patronage and dental practitioners should be motivated to take up cases with provision of adequate human and

material resources to meet the demand of this group of people. The establishment of inclusive units in both public and private schools in all local government areas in Nigeria should be encouraged. These will further the course of teaching, research, prioritization in publicly financed programmes and practice of orthodontics in Nigeria.

Conclusion

The difference in occlusal pattern and orthodontic treatment need in DS individuals might be attributed to genetic predisposition. Significantly, higher dental anomalies were recorded in the population with DS with predominantly class III incisal relationship and Angle's class III malocclusion on skeletal pattern III. Variation in growth of especially the facial skeleton could affect the development of the dental tissues, thus influencing the arrangement of dentition and occlusion. Furthermore, increase dental awareness targeting parents will help people with special needs positively.

Contributors

Bamgbose OJ was responsible for the conceptualization of the study, study design, statistical analysis, and interpretation of results. Sanu OO was responsible for the conceptualization of the study, study design, interpretation of results, critical review, editing and final approval of the study. Oredugba FA was responsible for the conceptualization of the study, study design, interpretation of results, critical review, editing and final approval of the study.

Funding

Self

Conflict of Interests

Nil

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Appendix A

Component and Weight of Dental Aesthetic Index

Component	Actual weight (wt)	Approximate wt
Missing incisor, canine and premolar teeth (number)	5.76	6
Crowding in incisal segments – Number of segment crowded	1.15	1
Spacing in the incisal segment- Number of segment spaced	1.31	1
Midline diastema (mm)	3.13	3
Largest anterior irregularity Maxilla (mm)	1.34	1
Largest anterior irregularity – Mandible (mm)	0.75	1
Anterior maxillary overjet (mm)	1.62	2
Angle's Class I molar relat (mm)	3.68	4
Vertical anterior open bite (mm)	3.69	4
Anterior-posterior relation largest deviation from Normal $\frac{1}{2} = 1$, Full cusp or more = 2	2.69	3
Constant	13.36	13