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The impact of orthodontic treatment on a young person's quality of life, aesthetics, and self-esteem in hypodontia; a longitudinal study
--Manuscript Draft--

Abstract:	Introduction: This research aimed to evaluate the impact of undergoing orthodontic treatment on a young person's oral health-related quality of life, self-esteem and aesthetics in relation to hypodontia. Materials and Methods: A prospective longitudinal hospital-based study recruited 97 participants with hypodontia, aged 11-18 years. Forty-one participants (42%) were originally planned to have space closure and the remainder space opening, with subsequent prosthetic replacement. The following questionnaires were completed both prior (T0) and after (T1) orthodontic treatment: child perception questionnaire (CPQ), Bristol condition-specific questionnaire for hypodontia (BCSQ), child health questionnaire (CHQ) and the Oral Aesthetic Subjective Impact Scale (OASIS). The Wilcoxon test and matched pairs t test approach were applied to compare between T1 and T0 for significant testing (p<0.05). Results: Fifteen participants were lost to follow-up, resulting in a total of 82 participants completing orthodontic treatment, with an average age of 13.8 (SD=1.71) years. A total of 282 teeth were missing in the sample. Treatment resulted in significantly lower levels of the indices (P<0.001), in relation to overall BCSQ, OASIS, appearance and how other people would treat them. In comparing the two sub-groups, those treated with space closure had significantly reduced functional limitations (CPQ), appearance concerns, self-esteem (CHQ), OASIS and overall BCSQ scores. Conclusions: Orthodontic treatment in participants with hypodontia appears to have a significant impact on a range of psychological and esthetic scales. In particular, space closure appears to significantly improve the quality of life of participants, compared to those undergoing space opening.

8th March 2023

Dear Editor,

RE: The impact of orthodontic treatment on a young person's quality of life, aesthetics, and self-esteem in hypodontia; a longitudinal study. Johal, Amin, Dean

We would be very grateful if you would consider the above manuscript for publication in the AJODFO. It is original in its concept, with previous studies limited assessing the impact of untreated hypodontia on quality of life using generic measures. Thus the objectives of the present study were to use both a generic and a newly developed condition specific quality of life instrument, developed and tested for validity and reliability for use in hypodontia patients, following the completion of orthodontic treatment, to sensitively assess the impact on the quality of life, self-esteem and aesthetics. The publication builds on the previously published manuscript in the AJODFO (2022), which evaluated untreated hypodontia patients.

The undersigned corresponding author warrants that the article is original, is not under consideration for publication by another journal and has not been previously published. I sign for and accept responsibility for releasing this material on behalf of *any* and all co-authors

Kind regards

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Reviewer #1: The authors have carefully responded to the reviewers' suggestions/comments in this revised submission.
This has resulted in an increase in the length of the Discussion to five(5) pages and the 12 Tables to six(6) pages. Ideally, this submission could benefit from the discussion being reduced in length and several of the 12 Tables combined or included in an appendix.

Response:
In response to the reviewer, I have removed 2 Tables which referred to the reliability and distribution, using the Cronbach's alpha, for the CPQ and BSCQ questionnaires to Appendices I and II. Just to note, the number of Tables has not changed from the original submission, as no reviewer requested any modification, this but I see that it is easy to move 2 of them to an Appendix
In addition, I have removed some of the discussion that repeated the results as previously suggested, thus reducing the Discussion to 4-pages.

Reviewer #2: The authors have adequately addressed the reviewers' concerns and made the appropriate amendments accordingly.

Response: We wish to thank the reviewer for his positive feedback and earlier comments which we feel have strengthened the manuscript further.

The impact of orthodontic treatment on a young person's quality of life, aesthetics, and self-esteem in hypodontia; a longitudinal study

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Running title: Impact of orthodontic treatment in hypodontia

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The impact of orthodontic treatment on a young person's quality of life, aesthetics, and self-esteem in hypodontia; a longitudinal study

Highlights

- Hypodontia remains the most prevalent genetic dental anomaly
- Untreated hypodontia in young people has a negative psychosocial impact, both in terms of social and emotional well-being.
- Orthodontic treatment in young people has a significant impact on a range of psychological and esthetic scales, influenced by the decision to close or open space.

The impact of orthodontic treatment on a young person's quality of life, aesthetics, and self-esteem in hypodontia; a longitudinal study

Abstract:

Introduction: This research aimed to evaluate the impact of undergoing orthodontic treatment on a young person's oral health-related quality of life, self-esteem and aesthetics in relation to hypodontia.

Materials and Methods: A prospective longitudinal hospital-based study recruited 97 participants with hypodontia, aged 11-18 years. Forty-one participants (42%) were originally planned to have space closure and the remainder space opening, with subsequent prosthetic replacement. The following questionnaires were completed both prior (T0) and after (T1) orthodontic treatment: child perception questionnaire (CPQ 11-14), Bristol condition-specific questionnaire for hypodontia (BCSQ), child health questionnaire (CHQ) and the Oral Aesthetic Subjective Impact Scale (OASIS). The Wilcoxon test and matched pairs t test approach were applied to compare between T1 and T0 for significant testing (p<0.05).

Results: Fifteen participants were lost to follow-up, resulting in a total of 82 participants completing orthodontic treatment, with an average age of 13.8 (SD=1.71) years. A total of 282 teeth were missing in the sample. Treatment resulted in significantly lower levels of the indices (P<0.001), in relation to overall BCSQ, OASIS, appearance and how other people would treat them. In comparing the two sub-groups, those treated with space closure had significantly reduced functional limitations (CPQ), appearance concerns, self-esteem (CHQ), OASIS and overall BCSQ scores.

Conclusions: Orthodontic treatment in participants with hypodontia appears to have a significant impact on a range of psychological and esthetic scales. In particular, space closure appears to significantly improve the quality of life of participants, compared to those undergoing space opening.

Key words: Hypodontia, quality of life, esthetics

Introduction

Non-syndromic hypodontia remains the most common dental developmental disorder, characterised by absence of one or more adult teeth, excluding the third molars, with an overall reported prevalence of 3.5-6.5%. [1,2] Hypodontia has recently been reported to negatively affect aesthetics, function and psychosocial well-being, with severe hypodontia showing greater negative impacts. [3] Various classifications have been used within the literature to describe the severity, with oligodontia the developmental absence of 6 or more teeth, excluding the third molars, and anodontia the complete absence of the dentition. Hypodontia has also been classified into mild (1-2 missing teeth), moderate (3-5 missing teeth) and severe (6 or more missing teeth). In terms of prevalence, severe hypodontia is reported to affect 0.14%, with the majority (83%) demonstrating the milder form. [1] Hypodontia may also be classified as non-syndromic or syndromic. Syndromic hypodontia is commonly linked with severe hypodontia and can be sporadic or familial within the familial ancestry. In contrast, non-syndromic hypodontia tends to vary in severity and frequently occurs within the permanent dentition. [4] The management of patients with hypodontia can be challenging, complex and timely and as such is thought to require a multi-disciplinary approach to care to achieve the best esthetic and functional outcomes and optimal treatment planning. The patient's needs must be solved in a functional and esthetic manner. An array of dental specialties are usually involved, with no single dental specialty possessing the varied expertise required to optimally treat this patient cohort, highlighting the multidisciplinary nature of care resulting in an interdisciplinary treatment plan. This includes the orthodontic, restorative, pediatric and oral and maxillofacial surgical teams. [5]

Hypodontia can be managed by orthodontic space closure or orthodontic space opening for prosthetic replacement. Both therapeutic options are effective, however orthodontic space closure has been reported as having a potential advantage over prosthodontic evaluation for congenitally missing lateral incisors and space opening has been advocated as the favoured approach for orthodontic treatment, particularly for maxillary lateral incisors. [6,7] There has been a reported trend away from space opening and prosthetic replacement towards orthodontic space closure, which may be indicative of a change in opinion towards prosthetic replacement options, as well as higher degrees of confidence with biomechanical strategies following the application of temporary anchorage devices to aid space closure. [8]

To-date, there appear to be significant shortcomings in the literature in respect of the impact of hypodontia on quality of life (QoL). Predominately, these have focussed on the impact of untreated hypodontia and furthermore are principally limited to the application of a generic scale of oral health related QoL. [9-15] There is an established need to utilise a condition-specific questionnaire to offer a tailored understanding into the effect of hypodontia, instead of assessing QoL in relation to general oral health. [16] More recently, Johal et al. [3] applied a combination of generic and a condition-specific scales for hypodontia, along with measures of self-esteem and esthetics. The authors demonstrated, in young people, the untreated condition had a negative psychosocial impact, both in terms of its presentation and planned care. [3] Of greater importance, is the lack of any studies evaluating the impact of orthodontic treatment on a young person's QoL, applying generic and/or a condition-specific measure of QoL. A study by Abu-Awwad et al. [14] applied a generic OHRQoL measure to assess the QoL of adult patients with hypodontia who completed treatment. The results of this study showed that hypodontia patients, following treatment reported higher OHRQoL average scores compared to the British population, with higher OHRQoL scores related to patient satisfaction with dental aesthetics following treatment. [14]

In terms of aesthetic self-perception, the literature has demonstrated that fixed orthodontic treatment in routine (non-hypodontia) patients, aged between 12 and 15 years, demonstrates significant improvement in the aesthetic self-perceptions. [17] Self-esteem is a key determining factor of psychological well-being. The effect of orthodontic treatment in hypodontia patients has not been well explored in relation to self-esteem and aesthetics, however a study looking at the impact of moderate and severe hypodontia on the QOL and self-esteem found no significant difference between those with hypodontia and the control group (Hashem et al., 2013). The older children become, the more aware they are of their aesthetics with them feeling unhappy with their dental appearance, resulting in aesthetics being the primary motivator for treatment. [18] This needs to be quantified in relation to the impact of orthodontic treatment in hypodontia patients.

It is essential to deduce the impact of completing orthodontic treatment on the quality of life in patients with hypodontia, especially in relation to orthodontic space closure and space opening. To-date, there have been no studies looking at the effect of completing orthodontic treatment, involving space closure or opening, in young people with hypodontia, in relation to either generic or condition-specific measure of QoL, esthetics or self-esteem. As a result, the

aim of the present study was to utilise both generic and condition-specific QoL tools to assess the impact of completing orthodontic treatment in participants with hypodontia on their QoL, self-esteem and esthetics, for both orthodontic space closure and space opening.

Materials and methods:

Participants:

This was a prospective, longitudinal hospital-based study in which participants with hypodontia were recruited from those attending a multidisciplinary clinic between June 2016 and January 2017. Participants who fulfilled the selection criteria were approached and invited to participate in the study. The inclusion criteria for this study were as follows: aged between 11 and 18 years; medically fit and well; able to speak and read English and with a radiographically confirmed diagnosis of non-syndromic hypodontia of at least 2 teeth, excluding third molars. Participants were excluded on the basis of: the presence of craniofacial anomaly; facial scarring or disfigurement; previously undergone orthodontic or restorative treatment to address the hypodontia or anterior aesthetics, or if there was any untreated dental disease, including caries or periodontal disease.

Methods:

Ethical approval was granted by the Regional Ethics Committee (reference number 15/LO/0294). Written informed consent was obtained from subjects aged 16 years and above, whereas for those aged under16 years, written parental consent and the child's assent were obtained. A data collection form was completed for all participants enrolled in the study, designed to collect the following information: age, gender, ethnicity, teeth present and missing, the presence of other dental features associated with hypodontia, for example, microdontia or hypoplasia, in addition to details of any planned orthodontic and/or restorative treatment.

All participants completed the following set of questionnaires prior to undertaking any orthodontic treatment (T0): the Child Perception Questionnaire (CPQ 11-14), a generic scale of QoL[19] and the Bristol condition-specific questionnaire for hypodontia (BCSQ) [16,20] the Child Health Questionnaire (CHQ-CF-87) [21] for measurement of self-esteem and the Oral Aesthetic Subjective Impact Scale (OASIS) [22] to assess aesthetic concerns. All participants

were seen for assessment and treatment planning on a multidisciplinary clinic. The decision as to whether to open or close space was an entirely non-randomised and based jointly on the advice given from the multidisciplinary team involved in their overall care and the participant's/parent's decision based on the former advice. Participants then completed the same series of four questionnaires, under supervision, following completion of orthodontic treatment (T1), in participants undergoing orthodontic space closure and orthodontic space opening, prior to definitive prosthetic replacement.

The CPQ (11-14) has 37 questions divided into 4 domains: oral symptoms (6 items), functional limitations (9 items), emotional well-being (9 items), and social well-being (13 items). The questionnaire is designed to establish the child's view of their dental appearance and the perceived view of their peers. It asks about the frequency of events experienced by the child in the previous 3 months. The response options were as follows: never = 0; once/twice = 1; sometimes = 2; often = 3; and every day/almost every day = 4. In addition, it included 2 general questions broadly phrased to assess oral health and its effect on daily life. The questionnaire has been shown in the past to demonstrate validity and reliability and has also been shown to be valid and reliable for the United Kingdom population. [19,22,23]

The BCSQ is newly designed to assess the relationship between hypodontia and QOL in 4 themes: treatment (4 items), activities (7 items), appearance (9 items), and the reaction of other people (10 items). The questionnaire has a few additional questions with categorical options (rather than Likert scales) that are not meant to be aggregated into indexes. The BCSQ was developed in conjunction with Royal Devon and Exeter NHS Foundation Trust and Bristol University. [20] Akram et al. [16] subsequently demonstrated its validity and reliability as an instrument for the assessment of QoL in participants with hypodontia.

The CHQ-CF-87 is a generic paediatric health-related QOL instrument that consists of 87 questions to assess children's physical and psychosocial experience aged 11-17 years. [21] A subscale of 14 questions from this questionnaire was used to assess how missing teeth affected the child's self-feeling and value in their social confidence, school activities, and self-regard over the preceding 4 weeks. It is measured on a 5-point Likert scale ranging from very good to very badly. The higher the score, the higher the self-esteem. This subscale questionnaire has been recognized as a valid and reliable measurement tool for assessing self-esteem in children. [23,24]

The OASIS questionnaire contains 5 items, scored on a 7-point Likert scale, developed by Mandall et al [22]. It can be used to measure the level of concern that participants have toward the appearance of their teeth, including positive or negative comments, teasing, and avoidance of smiling.

Statistical analysis:

A sample size calculation determined 65 participants were required to detect a mean difference of 8.2 in the CPQ scale, at the 5% level of significance, and with a power of 80%. The number was inflated to allow for dropouts/loss to follow-up by 30% to permit long-term follow-up of participants to the completion of planned restorative care. Thus, a minimum of 85 participants was to be recruited, with an effect size of 0.5. This was based on the study of Laing et al. [10] which evaluated the impact of untreated hypodontia on the oral health-related QOL, applying the CPQ scale and reported a standard deviation (SD) of 16.03.

Descriptive and analytical statistics were used to analyse the data. All questionnaire outcomes were statistically assessed, using Cronbach's α, for their internal consistency, that is, how closely related a set of items are as a group. In order to compare groups for significant testing, Wilcoxon tests were applied, which has a benefit of not assuming normality of the error terms as in an ANOVA. This test only assumes that the variables are at least on an ordinal scale. In this procedure the original values of the observations in their original units are converted to ranks and summed. The distribution of the sum of ranks is known and therefore allows the use of a Z test of to compare between two groups or a Chi-square to compare between many groups. For presentational consistency all tests presented used the Chi-Square. To estimate the differences over time (T1-T0), a matched pairs t test approach was adopted. This was also applied in both space closure and space opening groups for further analysis. The significance level was set at $\alpha = 5\%$ for all tests individually. All analysis was executed using JMP statistical software (version 14; SAS, Cary, NC).

Results:

Participant characteristics:

A total of 97 participants were initially recruited, 41 were originally planned to have space closure as part of their orthodontic plan, and the remaining 56 participants were planned for space opening. A total of 15 participants were lost to follow-up, with 82 participants completing orthodontic treatment, with an average age of 13.8 (SD=1.71) years. Changes in

the proposed orthodontic treatment plan occurred, resulting in 40 participants (49%) having orthodontic space closure and 42 (51%) undergoing space opening. From the 82 participants, 59% were classified as mild (2 missing teeth), with the remainder demonstrating moderate-severe hypodontia and overall the sample was predominantly female (n=62).

In assessing the characteristics of the sample at T1 (n=82), 51 (62%) were female (Table 1). Within this total sample, 48 (30 female) participants demonstrated mild (2 absent teeth) and 34 (21 female) moderate to severe (>2 absent teeth) hypodontia. A total of 282 teeth were missing in the sample. Thirty-five (42%) participants were missing anterior teeth, with the maxillary lateral incisors predominating, whilst 24 (29%) were missing posterior teeth, with the absence of mandibular second premolars predominating. Twenty-three (28%) participants were missing both anterior and posterior teeth. The mean age of participants was 13.8 (SD, 1.6) years in the mild group and 13.8 (SD, 1.9) years in the moderate to severe hypodontia group. In terms of ethnicity, there was a high proportion of White British (34%) participants, followed by Pakistani, Indian, and Bangladeshi (27%); other White (16%); Black British (10% each) and other (13%), which was selected when participants were unable to assign themselves to any of the ethnic groupings.

Child perception questionnaire (CPQ):

In terms of the global CPQ rating score, determined from separate general questions, 11 (13%) participants rated the oral health of their teeth, lips, and mouth as being fair or poor and were all from the space opening group, whereas 71 participants (87%) thought it was good or above. In addition, 22 (27%) of the space closure and 17 (21%) of the space opening groups responded very little or not at all, to the statement: 'How much does the condition of your teeth, lips, jaws, or mouth affect your life overall.'

Assessing the reliability of the CPQ as a whole and per theme, Appendix 1 presents Cronbach's α for all 37 components and each of the 4 themes. For the overall CPQ score, emotional and social well-being, the index demonstrated high reliability, in the range of 0.9-0.9. Nevertheless, and for the sake of wholesome reporting, oral symptoms and functional limitations were kept for further analysis. Ranked in descending order, the overall CPQ index and its themes had medians as follows: oral symptoms (1.0), functional limitations (0.7), emotional well-being (0.6), mean CPQ (0.7), and social well-being (0.3).

Table 2 presents a rank-sum test for each CPQ index across hypodontia, missing sites, and gender groups. No significant differences were detected. Following analysis of the CPQ overall index, as well as its partial components, significant differences were present between the treatment plans in all aspects but oral symptoms (Table 3). The space closure group had significantly lower levels of the indices: overall CPQ ($p<0.001$), functional limitations ($p<0.001$), emotional well-being ($p<0.001$) and, social well-being ($p<0.001$).

The Bristol condition-specific questionnaire for hypodontia (BCSQ):

Estimating the reliability of the BCSQ as a whole and per theme, Appendix 2 provides the Cronbach's α and the distribution characteristics for the separate indexes. Four of the indexes, namely overall BCSQ, treatment, activities, appearance, and other people's reaction, had above sufficient reliability, with a in the range of 0.8-0.9. In contrast, the index for treatment, with its 4 items, fell short of sufficiency. Bearing this in mind, we report this index for the rest of the procedure as a matter of completeness.

Rank-sum tests for each BCSQ index across hypodontia, missing sites, and gender groups are presented in Table 4. For hypodontia, 2 out of the 5 tests proved to be significant, namely overall BCSQ ($p=0.03$) and appearance ($p=0.02$). In both indexes, the moderate to severe group reported higher levels of negative impact. In relation to gender, treatment was found to be significantly different ($p=0.03$), with males showing lower values of the index. For missing sites, none of the indices were found to be significantly different. Testing for differences across treatment plans, with the BCSQ indices (Table 5), the space closure group had significantly lower values in all dimensions: overall BCS ($p<0.001$), treatment ($p=0.04$), activities ($p<0.001$) and, appearance ($p<0.001$).

Child health questionnaire (CHQ):

For the CHQ-self-esteem questionnaire, the reliability of the 14 items was high, with an estimated Cronbach's α of 0.9. The difference between the treatment plans, in respect of self-esteem, was insignificant.

In contrast, for the OASIS scale, the space closure group had significantly lower values, when compared with the space opening group ($p<0.001$; Table 6).

Averaging the items yield for the CHQ-self-esteem index, the score ranged between 1-4, with a mean of 2.9 (SD, 0.8) and a median of 3 at T0. At T1 the index had a mean of 3.0 (SD, 0.8) and a median of 3. Table 7 presents the rank-sum tests for this index at both time points across hypodontia, missing sites, and gender groups, with no significant differences detected.

Oral Aesthetic Subjective Impact Scale (OASIS):

For the 5 items of the OASIS, reliability was high and estimated at Cronbach's α of 0.9. The average OASIS index score was distributed in the range of 1-7, with a mean of 3.7 (SD 1.8) and a median of 3.4 at T0. At T1, the index had a mean of 3.0 (SD, 1.8) and a median of 2.5. Testing for differences across hypodontia groups, a significant difference (p=0.024) was found at T1 only otherwise, across missing sites and gender no significant difference was detected (Table 8).

Overall effect of orthodontic treatment:

Estimating the effect of orthodontic treatment (T0 and T1), a matched pairs t test approach was used (Table 9). The following observations were detected in relation to the scores: a decrease in overall BCSQ score (p<0.001); an increase in treatment (p<0.001); a decrease in appearance (p<0.001); a decrease in how other people will react (p<0.001) and a decrease in OASIS score (p=0.004).

In testing for differences between the sub-groups (space opening vs. space closing), a significant difference was found for the following indices (Table 10): the space closure group had a greater reduction over time in respect of functional limitations (p=0.011); overall BCSQ score (p <0.001); activities (p=0.004); appearance (p<0.001) and in terms of the OASIS score (p=0.002). The space opening group had a bigger reduction over time for CHQ – self-esteem (p=0.023).

Discussion:

The prospective longitudinal hospital-based study looked at the impact of orthodontic treatment, following orthodontic space closure and orthodontic space opening, on varying degrees of hypodontia severity.

Overall, a lower dropout rate (n=15) was observed over this longitudinal clinical study, which may be explained by the fact that these participants demonstrated a significant impact of their malocclusion in terms of their quality of life prior to treatment. [3] Of the final sample of 82 participants, 59% were classified as mild (2 missing teeth), with the remainder demonstrating moderate-severe hypodontia and overall the sample was predominantly female (n=62). The results demonstrate that in a small proportion of participants, the planned care, that is space opening versus space closure changed during the course of orthodontic treatment. This was explained in terms of the clinical treatment response observed in the hypodontia patients, with one participant proving to be challenging to close space. Such participants were then reviewed by the MDT and agreement to plan for space maintenance and prosthetic replacement, whilst no change in treatment goals were observed in the planned space opening group.

The participant demographics are similar to those reported in previous studies in terms of gender distribution. [1,2] The most common missing teeth present in the sample, excluding the third molars, were maxillary lateral incisors, which compares to other studies, except for those looking at the overall prevalence data in hypodontia. [1,2] This is most likely attributable to the fact that participants were recruited from those referred to a hospital setting, whereby the absence of mandibular second premolars would be less apparent and may not warrant treatment.

In terms of the severity of hypodontia, significant differences were detected following orthodontic treatment (T1), when comparing mild versus moderate-severe categories, in relation to the average OASIS index score, with the latter group reporting higher scores along with higher levels of negative impact in relation to the overall BCSQ and appearance. In comparison to the baseline (T0) scores, those with moderate-severe hypodontia had higher levels of negative impact in relation to the overall CPQ, emotional and social well-being. No significant differences were found in relation to the location of teeth absent (anterior or posterior), which echoes the results found at T0. When looking at gender, males had lower values for treatment within the BCSQ scores. In relation to gender differences, Abu-Awwad et al. [14] also found that females reported higher scores of OHRQoL than males following dental treatment to manage their hypodontia.

In terms of the global CPQ rating score, only participants in the space opening group (13%) rated the oral health as fair or poor at T1, with a reduction in scores being observed from T0. Interestingly, there were no participants from the space closure group within the fair or poor

categories, which would indicate a perceived increase in the oral health of those treated with orthodontic space closure.

When comparing the overall changes from T0 and T1, significant differences were detected, with T1 having reduced scores in relation to the overall BCSQ, appearance, how people will react, and an increase in treatment scores within the BCSQ and OASIS score. In relation to how people would react, there was an overall reduced concern that people may think it was weird to have false teeth, and that participants at T1 were generally less embarrassed in meeting people for the first time, as well as being less reluctant to their friends knowing about their missing teeth. However, in the absence of any other prospective longitudinal study assessing the impact of orthodontic treatment on hypodontia, the present findings were compared to and supported by a meta-analysis by Andiappan et al [25] who found that Oral Health Impact Profile scores were significantly lower after receiving orthodontic treatment for their malocclusion, and Feu et al [17] who found that that young patients who had fixed orthodontic treatment had significantly improved aesthetic self-perceptions.

When comparing the two sub-groups post-orthodontic treatment (space closure vs. space opening for prosthetic replacement), considerable differences were found. In terms of overall CQP index and its individual components, there were significant differences between the groups, with participants undergoing space closure demonstrating significantly reduced levels of indices, in relation to the overall CQP, functional limitations, emotional well-being and social well-being. The orthodontic space closure group also had significantly lower values in most dimensions in relation to the BCSQ index (the overall BCS, treatment, functional limitations, activities and appearance) and significantly lower values for the OASIS index. In terms of the treatment domain, these participants felt that their treatment would be less complicated and take less longer than that of their friends. In addition, the space closure group were also less worried about how their teeth would look at the end of treatment and were less worried about having to wear false teeth, with a reduced impact on their daily activity. In terms of the appearance domain, they felt less embarrassed about their absent teeth, of which they felt looked less out of proportion than the space opening group. In addition, they were less concerned of additional gaps occurring from their baby teeth falling out, and had reduced concerns about how other people would react to their missing teeth. There was no statistically significant difference for the CHQ-self-esteem index between the two sub-groups at T1. However, the space opening group demonstrated a larger reduction

over time (T0 to T1) with the CHQ – self-esteem. Unfortunately, there is a lack of published evidence for comparison with the results of the present study following orthodontic treatment in hypodontia patients in relation to QoL. Abu-Awwad et al [14] reported that following completion of dental treatment, hypodontia patients reported higher OHRQoL average scores compared to the British population, with higher scores relating to patients' satisfaction with dental aesthetics after treatment. [14] Orthodontic space closure has been reported as having a potential advantage over prosthodontic rehabilitation for those with congenitally missing lateral incisors, with patients reporting to be more satisfied with their appearance. [6,26] However the latter study had significant design limitations and must be interpreted with caution. In contrast, others have reported orthodontic space closure and implant replacement for missing maxillary incisors to produce similar well-accepted results. [27] Furthermore, the studies are notably restricted to those with hypodontia of the missing upper lateral incisors only.

Thus, the impact of undergoing orthodontic treatment in participants with moderate to severe hypodontia, reveals that they experience significant negative impacts in terms of their overall quality of life as well as their level of concern towards the appearance of their teeth, teasing and avoidance of smiling. In terms of the impact of orthodontic treatment in those requiring space opening for eventual prosthetic replacements we can surmise that this will be associated with higher levels of negative impact in terms of functional limitation, emotional and social well-being and greater aesthetic concerns. As clinicians, we therefore need to support our patients in adapting to these challenges by better informing them that undergoing orthodontic treatment will be difficult but that that they are embarking on a life-long journey and in establishing the correct positioning of their teeth, the end result will be significantly enhanced and more predictable.

In terms of the outcome scales applied in the present study, the combination of a generic and condition-specific measure for hypodontia, in conjunction to a measure of self-esteem and dental aesthetics offered a more comprehensive evaluation of the impact of this prevalent condition.. This also allows for contextualisation of the use of different measures within QoL and for those participants with hypodontia. By applying this in the future, it can decrease study heterogeneity as well as permitting for future use, for instance within meta-analysis.

Aside from a single study reporting the T0 findings of this study (Johal et al., 2022), to-date there has been no previous studies using CHQ to assess the self-esteem in hypodontia patient.

The orthodontic space opening group had a significantly increased reduction over time in their self-esteem, when compared to the orthodontic space opening groups, when looking at T1 and T0.

Furthermore, there have been no studies utilising the BCSQ questionnaire to look at the impact of orthodontic treatment in hypodontia, with reliability and validation testing, other than the initial development of the tool. [16,20] By using this condition-specific quality of life measure, it gives us an important understanding of the patients' concerns, post-orthodontic treatment. [16,20]

The 5 items of the OASIS index score looked to quantify the degree of concern that the cohort had in relation to their dental appearance, in terms of both negative and positive comments received including any teasing, and if they avoided smiling. A significant difference was found in relation to a reduction in OASIS scores, when comparing the difference between T1 and T0. There were no significant differences within the independent variable categories of hypodontia severity, missing site or gender groups. The orthodontic space closure group had a significantly increased reduction over time, when compared to the orthodontic space opening groups. The OASIS questionnaire was developed (Mandall et al., 2000) to allow for the children to determine their worries with the dental appearance of their anterior teeth, whereby subjects in the study with lower orthodontic treatment needs had lower scores and perceived themselves to be less worse off than those with higher treatment needs. [22] The Mandall et al [22] study had a lower reported mean than seen in the present study, and looked at all types of malocclusion representative of the general population, whereas the current study recruited participants with hypodontia attending a hospital-setting, with teeth missing teeth anteriorly, which can affect their dental appearance concerns.

When looking at the impact of hypodontia on a patient's quality of life, aesthetics and self-esteem, consideration must also be taken in relation to the potential effect of ethnicity, patient compliance and socioeconomic status on the uptake of treatment and treatment outcome. The effect of social determinants can be considered in future studies to establish the interaction comprehensively.

There is limited evidence within the literature to-date, in relation to the exact impact of restorative arms of treatment in patients with hypodontia following orthodontic treatment for

space opening to permit prosthetic replacement. The participants from this research study will be followed up to assess the impact of restorative treatment in this cohort.

Conclusion:

Orthodontic treatment in participants with hypodontia appears to have a significant impact in a range of psychological and aesthetic scales. Orthodontic space closure appears to significantly improve the quality of life of participants, compared to those undergoing space opening for a prosthetic replacement. Hypodontia patients in whom prosthetic replacement is planned following orthodontic space opening appear to have very different concerns than those for whom space closure is planned. As a result, we must ensure that all clinicians involved within the management of hypodontia are aware of these implications, particularly with the orthodontic management, to facilitate shared-decision making with patients and to in turn address any concerns that they may have.

Author credit statement:

Ama Johal contributed study conceptualization, investigation, supervision, data analysis and validation, manuscript drafting; Rabia Dean contributed to data collection, data analysis, and drafting of the manuscript; Mandana Amin contributed to participant recruitment.

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Appendices:

Appendix I: Reliability and distribution of CPQ indexes

Appendix II: BCSQ indices - distribution and reliability.

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Tables:

Table 1: Sample characteristics (n=82).

	Gender					
	Female		Male		Combined	
Hypodontia	N	%	N	%	N	%
Mild	30	63	18	38	48	59
Moderate and Severe	21	62	13	38	34	41
Missing site						
Anterior	23	66	12	34	35	42
Posterior	14	58	10	42	24	29
Combination	14	61	9	39	23	28

	missing site					
	anterior		combination		posterior	
Hypodontia	N	%	N	%	N	%
Mild	28	58	4	8	16	33
Moderate and Severe	7	21	19	56	8	24
All	35	42	23	28	24	29

Table 2: Rank sum tests for Child Perception Questionnaire (CPQ) across hypodontia, missing site and gender groups after completion of orthodontic treatment (T1).

	Hypodontia: Mild/Moderate/Severe		Missing site: Anterior/ Posterior/ Both			Gender: Male/Female	
	Z	P>|Z|	χ^2	DF	P> χ^2	Z	P>|Z|
Overall CPQ	1.5	0.1	4.5	2	0.103	-1.6	0.103
Oral symptoms	-0.3	0.7	3.4	2	0.182	-1.0	0.334
Functional limitations	0.7	0.5	2.2	2	0.329	-1.6	0.105
Emotional well being	2.0	0.1	3.9	2	0.141	-1.9	0.063
Social well being	1.9	0.1	5.0	2	0.081	-1.1	0.276

Table 3: Child Perception Questionnaire (CPQ) index by orthodontic treatment plan (space opening vs. space closing). Wilcoxon test.

Dependent variable	Orthodontic Plan	n	Mean	Std Dev	χ^2	DF	$P > \chi^2$
Overall CPQ	Close space	40	0.5	0.4	14.1	1	0.0002
	Open space	42	1.0	0.6			
Oral symptoms	Close space	40	0.9	0.6	3.0	1	0.086
	Open space	42	1.1	0.5			
Functional limitations	Close space	40	0.5	0.5	14.1	1	0.0002
	Open space	42	1.1	0.7			
Emotional well-being	Close space	40	0.6	0.7	12.8	1	0.0003
	Open space	42	1.3	1.0			
Social well-being	Close space	40	0.3	0.3	9.2	1	0.0024
	Open space	42	0.7	0.7			

Table 4: Rank-sum tests for Bristol condition-specific questionnaire for hypodontia (BCSQ) across hypodontia, missing site and gender groups after completion of orthodontic treatment (T1).

	Hypodontia: Mild/Moderate/Severe		Missing site: Anterior/ Posterior/ Both			Gender: Male/Female	
	Z	P>|Z|	χ^2	DF	$P > \chi^2$	Z	P>|Z|
Overall BCSQ	2.2	0.031	4.5	2	0.108	-1.6	0.100
Treatment	1.4	0.151	0.3	2	0.878	-2.2	0.030
Activities	1.7	0.087	4.4	2	0.111	-1.0	0.314
Appearance	2.3	0.020	3.5	2	0.176	-1.2	0.247
How other people will react	1.6	0.102	3.1	2	0.207	-2.0	0.048

Table 5: Bristol condition-specific questionnaire for hypodontia (BCSQ) indices by orthodontic treatment plan (space opening vs. space closing). Wilcoxon test.

Dependent variable	Orthodontic Plan	n	Mean	Std Dev	χ^2	DF	P>χ2
Overall BCSQ	Close space	40	1.09	0.54	22.30	1	<0.001
	Open space	42	1.86	0.73			
Treatment	Close space	40	2.32	0.92	4.25	1	0.039
	Open space	42	2.75	0.75			
Activities	Close space	40	0.88	0.58	15.62	1	<0.001
	Open space	42	1.59	0.83			
Appearance	Close space	40	0.96	0.77	24.82	1	<0.001
	Open space	42	2.12	0.97			

Table 6: Child health questionnaire (CHQ) for self-esteem and oral aesthetic subjective impact scale (OASIS) indices by orthodontic treatment plan groups (space opening vs. space closing). Wilcoxon test.

Dependent variable	Orthodontic Plan	n	Mean	Std Dev	χ^2	DF	P> χ^2
CHQ - Self Esteem	Close space	40	3.2	0.7	3.7	1	0.0543
	Open space	42	2.8	0.8			
OASIS	Close space	40	2.3	1.4	13.5	1	0.0002
	Open space	42	3.7	1.8			

Table 7: Child health questionnaire (CHQ) for self-esteem index before (T0) & after (T1) orthodontic treatment, by hypodontia condition, missing site and gender groups. Wilcoxon test.

	Hypodontia: Mild/Moderate/Severe		Missing site: Anterior/ Posterior/ Both			Gender: Male/Female	
	Z	P>|Z|	χ^2	DF	P> χ^2	Z	P>|Z|
CHQ- Self Esteem index T1	-0.2	0.861	3.0	2	0.217	1.7	0.080
CHQ- Self Esteem index T0	-1.5	0.15	0.8	2	0.677	-1.8	0.076

Table 8: Oral aesthetic subjective impact scale (OASIS) index before (T0) & after (T1) orthodontic treatment, by hypodontia condition, missing site and gender groups. Wilcoxon test.

	Hypodontia: Mild/Moderate/Severe		Missing site: Anterior/ Posterior/ Both			Gender: Male/Female	
	Z	P>|Z|	χ^2	DF	P> χ^2	Z	P>|Z|
OASIS T1	2.3	0.024	2.5	2	0.288	-1.4	0.163
OASIS T0	1.8	0.071	1.5	2	0.466	-1.5	0.130

Table 9: Questionnaire indices comparing before and after orthodontic treatment. Matched pairs t tests.

	N	Mean	Std Dev	S	P>|S|
Overall CPQ T0	82	0.9	0.6		
Overall CPQ T1	82	0.8	0.6		
Overall CPQ T1-T0	82	-0.1	0.6	-370	0.087
Oral symptoms T0	82	1.1	0.5		
Oral symptoms T1	82	1.0	0.6		
Oral symptoms T1-T0	82	-0.1	0.7	-182	0.401
Functional limitations T0	82	0.9	0.6		
Functional limitations T1	82	0.8	0.7		
Functional limitations T1-T0	82	-0.1	0.7	-188.5	0.386

Emotional well-being T0	82	1.2	1.1		
Emotional well-being T1	82	1.0	0.9		
Emotional well-being T1-T0	82	-0.3	1.2	-393	0.069
Social well-being T0	82	0.6	0.6		
Social well-being T1	82	0.5	0.6		
Social well-being T1-T0	82	-0.1	0.6	-225	0.300
Overall BCSQ T0	82	1.8	0.6		
Overall BCSQ T1	82	1.5	0.8		
Overall BCSQ T1-T0	82	-0.3	0.7	-718.5	<0.001
Treatment T0	82	2.2	0.8		
Treatment T1	82	2.5	0.9		
Treatment T1-T0	82	0.4	1.0	734	<0.001
Activities T0	82	1.3	0.7		
Activities T1	82	1.2	0.8		
Activities T1-T0	82	-0.1	0.9	-131	0.547
Appearance T0	82	2.1	0.9		
Appearance T1	82	1.6	1.1		
Appearance T1-T0	82	-0.5	1.0	-961	<0.001
How other people will react T0	82	1.7	0.7		
How other people will react T1	82	1.2	0.7		
How other people will react T1-T0	82	-0.5	0.8	-1073	<0.001
CHQ - Self Esteem T0	82	2.9	0.8		
CHQ - Self Esteem T1	82	3.0	0.8		
CHQ - Self Esteem T1-T0	82	0.1	1.0	73.5	0.736
OASIS T0	82	3.7	1.8		
OASIS T1	82	3.0	1.8		
OASIS T1-T0	82	-0.6	2.0	-611	0.004

Table 10: The influence of the treatment plan in the 2 sub-groups (space opening vs. space closing) over time. Matched pairs t tests.

	Close space			Open space			χ^2	DF	$P > \chi^2$
	N	Mean	Std Dev	N	Mean	Std Dev			
Overall CPQ	40	0.52	0.41	42	1.00	0.62	3.65	1	0.056
Overall CPQ T0	40	0.82	0.58	42	0.94	0.58			
Overall CPQ T1-T0	40	-0.30	0.66	42	0.06	0.56			
Oral symptoms	40	0.89	0.55	42	1.11	0.54			
Oral symptoms T0	40	1.04	0.54	42	1.06	0.54			
Oral symptoms T1-T0	40	-0.15	0.62	42	0.05	0.69	1.21	1	0.271
Functional limitations	40	0.53	0.52	42	1.05	0.66			
Functional limitations T0	40	0.77	0.53	42	0.93	0.56			
Functional limitations T1-T0	40	-0.24	0.66	42	0.12	0.65	6.50	1	0.011
Emotional well-being	40	0.57	0.70	42	1.31	1.01			
Emotional well-being T0	40	1.12	1.14	42	1.26	1.04			
Emotional well-being T1-T0	40	-0.56	1.17	42	0.05	1.07	2.70	1	0.100
Social well-being	40	0.31	0.29	42	0.70	0.69			
Social well-being T0	40	0.55	0.52	42	0.68	0.62			
Social well-being T1-T0	40	-0.24	0.63	42	0.02	0.53	3.00	1	0.083
Overall BCS	40	1.09	0.54	42	1.86	0.73			
Overall BCS T0	40	1.61	0.59	42	1.90	0.63			
Overall BCS T1-T0	40	-0.52	0.68	42	-0.04	0.67	10.85	1	<0.001
Treatment	40	2.32	0.92	42	2.75	0.75			
Treatment T0	40	1.97	0.83	42	2.35	0.68			
Treatment T1-T0	40	0.35	1.09	42	0.40	0.85	0.57	1	0.451
Activities	40	0.88	0.58	42	1.59	0.83			
Activities T0	40	1.25	0.68	42	1.34	0.78			
Activities T1-T0	40	-0.37	0.83	42	0.25	0.86	8.44	1	0.004
Appearance	40	0.96	0.77	42	2.12	0.97			
Appearance T0	40	1.84	0.79	42	2.28	0.85			

Appearance T1-T0	40	-0.88	0.90	42	-0.16	0.87	14.91	1	<0.001
How other people will react	40	0.92	0.51	42	1.45	0.79			
How other people will react T0	40	1.53	0.72	42	1.78	0.66			
How other people will react T1-T0	40	-0.61	0.76	42	-0.33	0.77	2.90	1	0.089
CHQ - Self Esteem	40	3.16	0.71	42	2.80	0.81			
CHQ - Self Esteem T0	40	2.87	0.81	42	2.94	0.74			
CHQ - Self Esteem T1-T0	40	0.29	0.98	42	-0.14	0.92	5.15	1	0.023
OASIS	40	2.29	1.37	42	3.73	1.79			
OASIS T0	40	3.61	1.79	42	3.72	1.85			
OASIS T1-T0	40	-1.33	1.91	42	0.01	1.78	10.07	1	0.002

Appendix I: Reliability and distribution of Child Perception Questionnaire (CPQ) index.

Index	No. of items	Cronbach α	Mean $\pm$ SD	Median
Overall CPQ	37	0.9	0.8 $\pm$ 0.6	0.7
Oral symptoms	6	0.6	1.0 $\pm$ 0.6	1.0
Functional limitations	9	0.8	0.8 $\pm$ 0.7	0.7
Emotional well-being	9	0.9	1.0 $\pm$ 0.9	0.6
Social well-being	13	0.9	0.5 $\pm$ 0.6	0.3

Note: scale range (0-4) n=82.

Appendix II: Bristol condition-specific questionnaire for hypodontia (BCSQ) index distribution and reliability.

Index	No. of items	Cronbach α	Mean $\pm$ SD	Median
Overall BCS	30	0.9	1.5 $\pm$ 0.8	1.4
Treatment	4	0.7	2.5 $\pm$ 0.9	2.6
Activities	7	0.8	1.2 $\pm$ 0.8	1.1
Appearance	9	0.9	1.6 $\pm$ 1.1	1.4
How other people will react	10	0.8	1.2 $\pm$ 0.7	1.1

Note: scale range (0-4) n=82.

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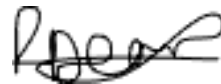
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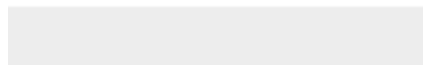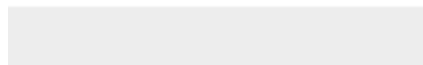
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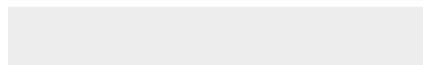**ICMJE Statement on Conflict of Interest for each author**
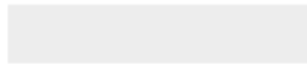coi_disclosure AJ.pdf

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Credit Author Statement:

Ama Johal contributed study conceptualization, investigation, supervision, data analysis and validation, manuscript drafting.

Rabia Dean contributed to data collection, data analysis, and drafting of the manuscript.

Mandana Amin contributed to participant recruitment.