

BMJ Open

Developing an Index for the Orthodontic Treatment Need in Pediatric Patients with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool between Physicians and Orthodontists

Journal:	<i>BMJ Open</i>
Manuscript ID:	bmjopen-2014-005680
Article Type:	Protocol
Date Submitted by the Author:	12-May-2014
Complete List of Authors:	Altalibi, Mostafa; University of Alberta, Department of Dentistry, Orthodontic Graduate Program Saltaji, Humam; University of Alberta, Department of Dentistry, Orthodontic Graduate Program Roberts, Mary; University of Alberta, Department of Rehabilitation Medicine, Occupational Therapy Program Major, Michael; University of Alberta, Inter-disciplinary Airway Research Clinic (I-ARC) Macleane, Joanna; University of Alberta, Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital Major, Paul; University of Alberta, Department of Dentistry
Primary Subject Heading:	Dentistry and oral medicine
Secondary Subject Heading:	Paediatrics, Ear, nose and throat/otolaryngology, Health services research
Keywords:	PAEDIATRICS, HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Otolaryngology < SURGERY

SCHOLARONE™
Manuscripts

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Developing an Index for the Orthodontic Treatment Need in Pediatric Patients with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool between Physicians and Orthodontists

Mostafa Altalibi^a; Humam Saltaji^b; Mary Roduta Roberts^c; Michael Major^d; Joanna Maclean^e, Paul W Major^f

^a MSc Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

^b PhD Candidate and Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

^c Assistant Professor of Occupational Therapy, Faculty of Rehabilitation Medicine, University of Alberta, Edmonton, Alberta, Canada.

^d Director, Inter-disciplinary Airway Research Clinic (I-ARC), Assistant Clinical Professor, Orthodontics, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

^e Medical Director at Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital; Assistant Professor, School of Medicine, University of Alberta, Edmonton, Alberta, Canada.

^f Professor and Chair of Dentistry; Senior Associate Dean, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada.

Corresponding Author

Mostafa Altalibi DMD, MSc (Ortho)
Orthodontic Graduate Program
School of Dentistry
5-476, Edmonton Clinic Health Academy (ECHA)
University of Alberta
11405-87 Ave, Edmonton, Canada T6G 1C9
Phone: 780 885-7457
E-mail: altalibi@ualberta.ca

Word Count: 2807

Key Words: Obstructive Sleep Apnea; Pediatric; Index; Orthodontic Treatment Need; Visual Analogue Scale; Development; Validation; Reliability.

ABSTRACT

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

only

INTRODUCTION

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.¹ More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.²⁻⁸ Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.^{9 10 11-13} Other physiologic processes that can be affected include stunted growth,¹⁴ high blood pressure,¹⁵ damage to the heart: ventricular hypertrophy,¹⁶ and cor pulmonale¹⁷.

In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla¹⁸ or mandible, a long lower face height and restriction in the space of the upper airway^{19 20}. Furthermore, when evaluating the polysomnography of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances²¹⁻²⁶ can be of benefit at reducing the severity of OSA. Reverse pull headgear²⁷⁻³², and maxillary & mandibular advancement surgery³³ have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, etc.) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment.

Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

METHODS AND ANALYSIS

A) Initial Development

In accordance with the World Health Organization's recommendations on developing an index,³⁴ development of the index will be achieved through the following objectives:

- 1- Establishing a Steering Committee
- 2- Scoping the index
- 3- Reviewing the literature
- 4- Drafting the index
- 5- Organizing an Index Development Group
- 6- Organizing an External Review Group

Steering Committee

The Steering Committee will be established a priori and is responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index.

Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in question will lead to an improvement in the OSA symptoms. Priority will be given to the results of well-conducted and well-reported systematic reviews and randomized control trials. Each of the factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will be searched for the craniofacial morphology of pediatric OSA patients. All of this

1
2
3 information will be brought back to the Steering Committee for discussion of inclusion /
4
5 elimination of factors in the index.
6

7 **Drafting the Index**

8
9
10 Since this index will have visual-rating scales for each of the craniofacial and occlusal
11 factors, the steering committee will devise an outline suitable for displaying each of the factors
12 representing the index. The factors in the drafted index will also have a number of levels that will
13 divide the factor into categories of severity. The number of levels for a given factor will also be
14 determined by the Steering Committee. Details describing the nature of the illustrations, the
15 amount of levels that each of the factors will have, and the general layout of the index will be
16 agreed upon by the steering committee, and the graphic artist will then design a preliminary
17 version of the index based on the relevant factors and the feedback from the steering committee.
18
19
20
21
22
23

24 **The Index Development Meeting**

25
26
27 The Index Development Group (IDG) is formed by a group of external multidisciplinary
28 experts who provide evidence-based recommendations, on the content, layout and development
29 process of the index. This eclectic group should be small enough to be able to have effective
30 discussions, while large enough to ensure the appropriate representation from all the
31 stakeholders. In this study we plan on including a broad spectrum of medical specialists, family
32 physicians, orthodontists, and methodologists. The goals of this meeting are to gain additional
33 information to strengthen the index, as well as gaining the professional credibility of a broad
34 range of experts on the topic of pediatric OSA.
35
36
37
38
39
40

41
42 Once the members have been chosen by the Steering Committee and accept the invitation
43 to participate, the IDG members will be emailed a document 4 days prior to the meeting,
44 explaining: the purpose of the index and meeting, how the meeting will proceed, what to expect
45 in the meeting, and a brief literature review of the orthodontic techniques currently available to
46 help with the symptoms of OSA. Moreover, every member will be randomly assigned a number
47 in order to maintain anonymity of the responses.
48
49
50
51

52 **Procedure of the IDG Meeting**

53
54
55 The meeting will commence with a brief introduction summarizing the literature review,
56 the purpose of the index and its relevance. The meeting will then proceed to collect feedback
57
58
59
60

1
2
3 (APPENDIX 1) on the aforementioned chosen factors using a modified Delphi technique^{35 36},
4 which is a communication technique that structures the meeting and minimizes bias in responses.
5 Participants' responses will be collected through a web-based response portal. Each factor will
6 be explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based
7 on its importance for decision-making. Once all the feedback is received on a particular factor,
8 the summary of the results will be displayed for everyone to see. Discussion will ensue for a
9 maximum of 5 minutes, with each person talking no more than 1 minute. Then everyone will be
10 asked to re-enter their feedback on the website for the same factor in light of the discussion. This
11 cycle will continue until a consensus of greater than 80% of the members is reached. Once
12 consensus is reached, then we will move on to the next factor and the participants will be asked
13 to give feedback on this new factor in a similar manner.
14
15
16
17
18
19
20
21
22

23 In addition, two negative control factors will be used to calibrate the responses. The first
24 negative control will be of orthodontic relevance but have no effect on the amelioration of the
25 symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of
26 relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck
27 circumference).
28
29
30
31

32 Once all the factors have been discussed and consensus reached, we will get feedback
33 through the website on the: alignment - importance of the index, and if the development process
34 is appropriate; relevance – content analysis, and whether all the factors identified are important;
35 representation – if there is anything that needs to be added to the index (APPENDIX 2). Finally,
36 additional written feedback will be accepted at the very end of the session.
37
38
39
40
41

42 All the feedback from the IDM will be summarized and presented to the Steering
43 Committee, and decisions will be made to remove factors, modify factors, and /or modify the
44 outline and layout of the index. These modifications will subsequently be presented to the
45 graphic artist and a second draft will be procured.
46
47
48

49 **The External Review Group**

50 The External Review Group (ERG) is composed of end-users and interested parties. This
51 group is not responsible for any content analysis, instead it will be responsible for reviewing the
52 layout, simplicity and ease of use. It will also be responsible to assess usefulness of the index in
53 the healthcare setting and give feedback on the feasibility of implementing the index in practice.
54
55
56
57
58
59
60

1
2
3 The goal of this meeting is to gain the end-user approval for the ease and feasibility of
4 administering the index. This group should be large enough to be a representative sample of the
5 population, yet small enough to allow for ease of explanation and healthy discussion. It is not as
6 structured as the IDM, and allows for the participants to freely express their opinions in an open
7 forum.
8
9

10
11
12 In this meeting we will explain the theory behind the index by briefly reviewing the
13 literature and then explain the purpose of the index. Subsequently we will show the group a pilot
14 version of the index and a paper will be distributed to receive for their feedback based on the
15 following questions:
16
17
18

- 19 1- Do you understand the purpose of the index?
- 20 2- Do you understand what each factor is assessing?
 - 21 a. If not which one(s) do you not understand and why?
- 22 3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
- 23 4- Would you use this index in your practice?
- 24 5- Other recommendations:
25
26
27
28
29
30

31 All the feedback from the ERG will be summarized and presented to the Steering
32 Committee, and decisions will be made to modify the outline and layout of the index. These
33 modifications will subsequently be presented to the graphic artist and a third draft will be
34 procured.
35
36
37
38

39 **B) Reliability**

40
41 Reliability of the index will be tested within a group that represents the typical end-user
42 population. This includes physicians of family medicine, pediatricians, pediatric ENT, or
43 pediatric sleep physicians. Reliability will also be assessed within a group of orthodontists, who
44 by their training are experts at assessing malocclusion and craniofacial morphology. Therefore
45 10-20 physicians and orthodontists will be recruited as examiners in this study.
46
47
48
49

50
51 A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-
52 disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and
53 their intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief
54 explanation about the use and application of the index, the physicians as well as the orthodontists
55
56
57
58
59
60

1
2
3 will apply the developed index to the sample patient pool's pictures once. In order to diminish
4 recall bias, application of the index will be repeated 2 weeks later.
5
6

7 Intra-rater reliability and inter-rater reliability between the physicians, between the
8 orthodontists and between the physicians and orthodontists will be compared. The reliability will
9 be assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.
10
11

12 13 **C) Validation**

14
15 The Index will be validated using dichotomous responses from orthodontists on whether
16 specific patients would require orthodontic treatment to help their obstructive sleep apnea
17 symptoms and comparing it to the score that the index gave those same patients. This will be
18 achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric
19 OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral
20 pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic
21 diagnosed with OSA, and the orthodontists will be asked to rate these patients, using a “yes” or
22 “no” response, whether they would benefit from orthodontic treatment for their OSA symptoms.
23
24 The index will then be applied on the same patients by the principal investigator and the score of
25 each patient will be recorded. Using a stepwise multiple-logistic regression each of the identified
26 factors will be given a weight; this will represent the relative importance of the factor. Once
27 analyzed if the correlation is high between the expert scores and the cluster groups, then the
28 clusters are meaningful and valid. Furthermore, a cutoff for most efficient score above which to
29 refer will be chosen using a graph and observing the value that optimizes the sensitivity,
30 specificity and overall accuracy of the index. Finally, four grades of treatment-need will be
31 determined using the twenty-five percentile ranges. The grades will be:
32
33
34
35
36
37
38
39
40
41
42
43

- 44 1- Minimal Need
 - 45 2- Mild Need
 - 46 3- Moderate Need
 - 47 4- Severe Need
- 48
49
50
51

52 **DISCUSSION**

53
54 The development plan of this index has been conceived through a modification of the
55 WHO Handbook for guideline development,³⁴ as well as reviewing the orthodontic literature for
56
57
58
59
60

ways indices have been previously developed. The WHO provided an excellent starting point, from there modifications were made to cater to the specifics of this study, given that there are differences between developing a guideline and an index for orthodontic treatment need. The literature was useful, and among the index development protocols reviewed, certain assessed the orthodontic treatment need within the entire population,³⁷⁻⁴⁰ while others assessed it for a given subpopulation⁴¹⁻⁴⁶; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a protocol for our particular needs from the available literature and using the experience and expertise of the authors. Through this protocol we aim to develop an index that fulfills all of the following criteria:⁴⁷

1. Gradient of Numeric Values: The severity of the orthodontic treatment need within the pediatric OSA patients should be defined within a numerical scheme that demonstrates a finite and progressive gradient from low need to high need.
2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
3. Clinical Importance: The numerical scale should correspond with the clinically appraised orthodontic treatment need of pediatric OSA patients.
4. Statistical Ease: should be amendable to statistical analysis.
5. Reliability: Should have a high intra- and inter-rater reliability.
6. Practical: The instruments required to score the index should be practical to the setting in which it will be administered.
7. Minimal Judgment: Applying the index should require minimal judgment.
8. Simple: The index shouldn't have a high financial or time cost, therefore should be simple enough to administer to many patients.
9. Detect Change: The index should be able to detect changes in orthodontic treatment need in pediatric OSA patients.
10. Validity: should be valid over time.

Validity can be characterized into different types: Face Validity, Content Validity, Construct Validity, and Criterion Validity.⁴⁸ In this study, we will examine these kinds of validity at different stages of development. The first draft of the index will focus on establishing face validity. Feedback from the steering committee and IDG will assist in establishing content and construct validity. Assuming that the “gold standard” in assessing the orthodontic treatment-need in pediatric patients with OSA is an orthodontist with experience in dealing with pediatric

1
2
3 OSA patients, then the subsequent modification of the index based on the reliability tests and the
4 dichotomous responses from the orthodontists provide the index with the necessary criterion-
5 related validity evidence through statistical means.⁴⁹
6
7

8 9 **Significance**

10
11 This index will help physicians and other medical professionals identify which
12 craniofacial phenotypes may benefit from orthodontic treatment as part of their multi-
13 disciplinary OSA management. Furthermore, due to the diverse medical effects of sleep
14 deprivation, there will be a trend to make sleep apnea into a centralized service, where the main
15 focus is for a highly trained multidisciplinary team to treat a high volume of patients to a
16 standardized protocol, where meticulous documentation is exercised. This index is part of that
17 documentation process. It will allow for quality assurance, funding allocation and epidemiologic
18 studies to be performed. It will also allow long-term follow up and audit in order to enter into
19 comparisons with other centers.
20
21
22
23
24
25
26

27 28 **Dissemination Plan**

29
30 The dissemination of this index will be done through a variety of ways in order to
31 maximize its reach. Primarily, it will be published in a peer-reviewed journal, which will allow
32 its introduction to the scientific literature. The journal should be a respected medical journal with
33 broad reach, in order to allow the greatest number of physicians to be exposed to the index.
34 Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian
35 and American population of medical professionals. Moreover, the index will be presented at
36 national and international conferences to increase the awareness of the index among the
37 scientific community. Finally, the index will be used at the University of Alberta's
38 Interdisciplinary Airway Research Center, and more research, so that future research in this
39 center will incorporate it. It will also be placed on the University of Alberta's Interdisciplinary
40 Airway Research Center's website under the physician section, to further educate the doctors
41 who visit the site on the index.
42
43
44
45
46
47
48
49
50
51

52 **Funding**

53
54 No funding has been provided for this project.
55

56 **Authors' Contributions**

1
2
3 MA HS MRR MM JM PM conceived and designed the study; MA drafted the manuscript and
4 integrated critical feedback from all of the other authors. All of the authors approved the final
5 version of the manuscript.
6
7

8 9 **Ethics approval**

10
11 Ethical approval has been submitted to the Research Ethics Board at the University of Alberta
12 (expected May 2014).
13

14 15 **Competing Interests**

16
17 The authors declare no conflicts of interests.
18
19

20 21 **REFERENCES**

- 22
23
24
25 .1 O'Brien LM, Mervis CB, Holbrook CR, Bruner JL, Klaus CJ, Rutherford J, et al.
26 Neurobehavioral implications of habitual snoring in children. *Pediatrics* 2004;114(1):44-
27 9.
28
29 .2 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proceedings of*
30 *the American Thoracic Society* 2008;5(2):242-52.
31
32 .3 Gislason T, Benediktsdottir B. Snoring, apneic episodes, and nocturnal hypoxemia among
33 children 6 months to 6 years old. An epidemiologic study of lower limit of prevalence.
34 *Chest* 1995;107(4):963-6.
35
36 .4 Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds.
37 *Archives of disease in childhood* 1993;68(3):360-6.
38
39 .5 Redline S, Tishler PV, Schluchter M, Aylor J, Clark K, Graham G. Risk factors for sleep-
40 disordered breathing in children. Associations with obesity, race, and respiratory
41 problems. *American journal of respiratory and critical care medicine* 1999;159(5 Pt
42 1):1527-32.
43
44 .6Bixler EO, Vgontzas AN, Lin HM, Liao D, Calhoun S, Vela-Bueno A, et al. Sleep disordered
45 breathing in children in a general population sample: prevalence and risk factors. *Sleep*
46 2009;32(6):731-6.
47
48 .7Li AM, So HK, Au CT, Ho C, Lau J, Ng SK, et al. Epidemiology of obstructive sleep apnoea
49 syndrome in Chinese children: a two-phase community study. *Thorax* 2010;65(11):991-7.
50
51
52
53
54
55
56
57
58
59
60

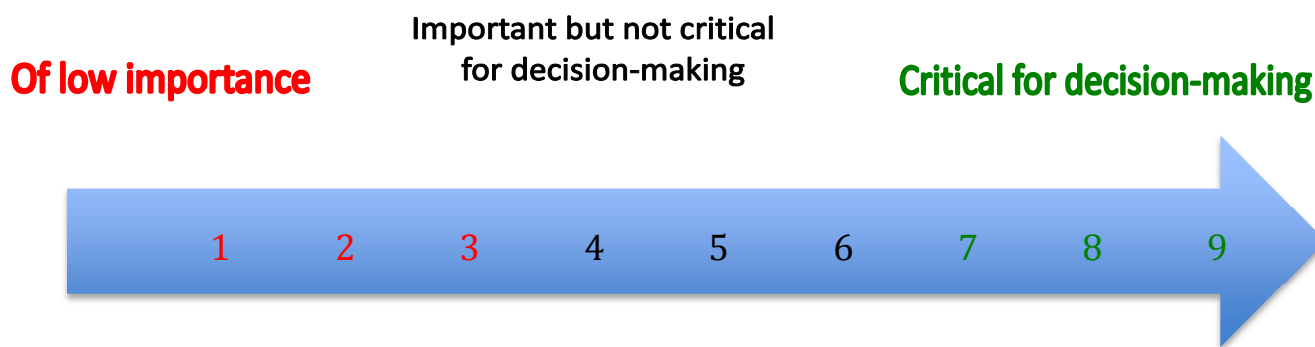
- 1
2
3 .8O'Brien LM, Holbrook CR, Mervis CB, Klaus CJ, Bruner JL, Raffield TJ, et al. Sleep and
4 neurobehavioral characteristics of 5- to 7-year-old children with parentally reported
5 symptoms of attention-deficit/hyperactivity disorder. *Pediatrics* 2003;111(3):554-63.
6
7
8 .9Huynh NT, Morton PD, Rompre PH, Papadakis A, Remise C. Associations between sleep-
9 disordered breathing symptoms and facial and dental morphometry, assessed with
10 screening examinations. *American journal of orthodontics and dentofacial orthopedics*
11 2011;140(6):762-70.
12
13
14 .10Beebe DW. Neural and neurobehavioral dysfunction in children with obstructive sleep apnea.
15 *PLoS medicine* 2006;3(8):e323.
16
17
18 .11Chervin RD, Archbold KH, Dillon JE, Panahi P, Pituch KJ, Dahl RE, et al. Inattention,
19 hyperactivity, and symptoms of sleep-disordered breathing. *Pediatrics* 2002;109(3):449-
20 56.
21
22 .12Crabtree VM, Varni JW, Gozal D. Health-related quality of life and depressive symptoms in
23 children with suspected sleep-disordered breathing. *Sleep* 2004;27(6):1131-8.
24
25 .13Halbower AC, Degaonkar M, Barker PB, Earley CJ, Marcus CL, Smith PL, et al. Childhood
26 obstructive sleep apnea associates with neuropsychological deficits and neuronal brain
27 injury. *PLoS medicine* 2006;3(8):e301.
28
29
30 .14Bar A, Tarasiuk A, Segev Y, Phillip M, Tal A. The effect of adenotonsillectomy on serum
31 insulin-like growth factor-I and growth in children with obstructive sleep apnea
32 syndrome. *The Journal of pediatrics* 1999;135(1):76-80.
33
34 .15Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea.
35 *American journal of respiratory and critical care medicine* 1998;157(4):1.103-1098:
36
37
38 .16Amin RS, Kimball TR, Bean JA, Jeffries JL, Willging JP, Cotton RT, et al. Left ventricular
39 hypertrophy and abnormal ventricular geometry in children and adolescents with
40 obstructive sleep apnea. *American journal of respiratory and critical care medicine*
41 2002;165(10):1395-9.
42
43
44 .17Sofer S, Weinhouse E, Tal A, Wanderman KL, Margulis G, Leiberman A, et al. Cor
45 pulmonale due to adenoidal or tonsillar hypertrophy or both in children. Noninvasive
46 diagnosis and follow-up. *Chest* 1988;93(1):119-22.
47
48 .18Korayem MM, Witmans M, MacLean J, Heo G, El-Hakim H, Flores-Mir C, et al.
49 Craniofacial morphology in pediatric patients with persistent obstructive sleep apnea with
50 or without positive airway pressure therapy: a cross-sectional cephalometric comparison
51 with controls. *American journal of orthodontics and dentofacial orthopedics*
52 2013;144(1):78-85.
53
54
55 .19Katyal V, Pamula Y, Martin AJ, Daynes CN, Kennedy JD, Sampson WJ. Craniofacial and
56 upper airway morphology in pediatric sleep-disordered breathing: Systematic review and
57
58
59
60

- meta-analysis. *American journal of orthodontics and dentofacial orthopedics* 2013;143(1):20-30 e3.
- .20 Flores-Mir C, Korayem M, Heo G, Witmans M, Major MP, Major PW. Craniofacial morphological characteristics in children with obstructive sleep apnea syndrome: a systematic review and meta-analysis. *Journal of the American Dental Association* 2013;144(3):269-77.
- .21 Carvalho FR, Lentini-Oliveira D, Machado MA, Prado GF, Prado LB, Saconato H. Oral appliances and functional orthopaedic appliances for obstructive sleep apnoea in children. *The Cochrane database of systematic reviews* 2007(2):CD005520.
- .22 Villa MP, Bernkopf E, Pagani J, Broia V, Montesano M, Ronchetti R. Randomized controlled study of an oral jaw-positioning appliance for the treatment of obstructive sleep apnea in children with malocclusion. *American journal of respiratory and critical care medicine* 2002;165(1):123-7.
- .23 Schutz TC, Dominguez GC, Hallinan MP, Cunha TC, Tufik S. Class II correction improves nocturnal breathing in adolescents. *The Angle orthodontist* 2011;81(2):222-8.
- .24 Cozza P, Polimeni A, Ballanti F. A modified monobloc for the treatment of obstructive sleep apnoea in paediatric patients. *Eur J Orthod* 2004;26(5):523-30.
- .25 Zhang C, He H, Ngan P. Effects of twin block appliance on obstructive sleep apnea in children: a preliminary study. *Sleep & breathing = Schlaf & Atmung* 2013;17(4):1309-14.
- .26 Pliska BT, Almeida F. Effectiveness and outcome of oral appliance therapy. *Dental clinics of North America* 2012;56(2):433-44.
- .27 Hiyama S, Suda N, Ishii-Suzuki M, Tsuiki S, Ogawa M, Suzuki S, et al. Effects of maxillary protraction on craniofacial structures and upper-airway dimension. *The Angle orthodontist* 2002;72(1):43-7.
- .28 Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway passage. *The Angle orthodontist* 2008;78(2):209-14.
- .29 Kilinc AS, Arslan SG, Kama JD, Ozer T, Dari O. Effects on the sagittal pharyngeal dimensions of protraction and rapid palatal expansion in Class III malocclusion subjects. *Eur J Orthod* 2008;30(1):61-6.
- .30 Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot study. *Eur J Orthod* 2006;28(2):184-9.
- .31 Kaygisiz E, Tuncer BB, Yuksel S, Tuncer C, Yildiz C. Effects of maxillary protraction and fixed appliance therapy on the pharyngeal airway. *The Angle orthodontist* 2009;79(4):660-7.

- 1
2
3 .32Lee JW, Park KH, Kim SH, Park YG, Kim SJ. Correlation between skeletal changes by
4 maxillary protraction and upper airway dimensions. *The Angle orthodontist*
5 2011;81(3):426-32.
6
7
8 .33Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive
9 sleep apnea: a systematic review and meta-analysis. *Sleep medicine reviews*
10 2010;14(5):287-97.
11
12 .34World Health Organization (2012) WHO Handbook for guideline development. Available:
13 http://www.who.int/hiv/topics/mtct/grc_handbook_mar2010_1.pdf. Accessed 10 Sept
14 2013.
15
16
17 .35Rowe G, Wright G. The Delphi technique as a forecasting tool: issues and analysis. *Int J*
18 *Forecasting* 1999;15(4):353-75.
19
20 .36Dalkey N, Helmer O. An Experimental Application of the Delphi Method to the use of
21 experts. *Management Science* 1963;9(3):458-67.
22
23
24 .37Richmond S, Shaw WC, O'Brien KD, Buchanan IB, Jones R, Stephens CD, et al. The
25 development of the PAR Index (Peer Assessment Rating): reliability and validity. *Eur J*
26 *Orthod* 1992;14(2):125-39.
27
28 .38Richmond S, Shaw WC, Roberts CT, Andrews M. The PAR Index (Peer Assessment
29 Rating): methods to determine outcome of orthodontic treatment in terms of
30 improvement and standards. *Eur J Orthod* 1992;14(3):180-7.
31
32
33 .39Daniels C, Richmond S. The development of the index of complexity, outcome and need
34 (ICON). *J Orthod* 2000;27(2):149-62.
35
36 .40Brook PH, Shaw WC. The development of an index of orthodontic treatment priority. *Eur J*
37 *Orthod* 1989;11(3):309-20.
38
39
40 .41Mars M, Batra P, Worrell E. Complete unilateral cleft lip and palate: validity of the five-year
41 index and the Goslon yardstick in predicting long-term dental arch relationships. *Cleft*
42 *Palate Craniofac J* 2006;43(5):557-62.
43
44 .42Mars M, Plint DA, Houston WJ, Bergland O, Semb G. The Goslon Yardstick: a new system
45 of assessing dental arch relationships in children with unilateral clefts of the lip and
46 palate. *Cleft Palate J* 1987;24(4):314-22.
47
48
49 .43Johnson N, Sandy J. An aesthetic index for evaluation of cleft repair. *Eur J Orthod*
50 2003;25(3):243-9.
51
52 .44Mossey PA, Clark JD, Gray D. Preliminary investigation of a modified Huddart/Bodenham
53 scoring system for assessment of maxillary arch constriction in unilateral cleft lip and
54 palate subjects. *Eur J Orthod* 2003;25(3):251-7.
55
56
57
58
59
60

- 1
2
3 .45Tohill C, Mossey PA. Assessment of arch constriction in patients with bilateral cleft lip and
4 palate and isolated cleft palate: a pilot study. *Eur J Orthod* 2007;29(2):193-7.
5
6
7 .46Huddart AG, Bodenham RS. The evaluation of arch form and occlusion in unilateral cleft
8 palate subjects. *Cleft Palate J* 1972;9:194-209.
9
10 .47Summers CJ. The occlusal index: a system for identifying and scoring occlusal disorders. *Am*
11 *J Orthod* 1971;59(6):552-67.
12
13 .48Streiner DL, Norman GR. *Health measurement scales: a practical guide to their*
14 *development and use*. Oxford: Oxford Medical Publications, 1995.
15
16 .49Terwee CB, Bot SDM, de Boer MR, van der Windt DAWM, Knol DL, Dekker J, et al.
17 Quality criteria were proposed for measurement properties of health status
18 questionnaires. *J Clin Epidemiol* 2007;60(1):34-42.
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Assigned Number _____



Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.

1. Do you think that this index is useful in fulfilling the goals of having an easy to use index that helps non-dentally trained professionals assess the orthodontic treatment need to help the symptoms in pediatric patients with OSA?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

2. Do you believe that the organizing committee is developing this index in the appropriate way? (i.e. Reviewing the recommendations in the literature, seeking expert opinions, gaining end-user approval and testing its reliability)

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

3. In your opinion were all the factors, identified in this meeting, important for this index?

Yes 1 2 3 4 5 6 7 8 9

No; Which factors were not important?

4. In your opinion, have all the important areas concerning orthodontic treatment need to help symptoms of OSA been identified?

Yes 1 2 3 4 5 6 7 8 9

No; Which ones were missed?

5. Is the layout of the index easy to navigate?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Please provide any other feedback below that may help improve the index:

For peer review only

BMJ Open

Developing an Index for the Orthodontic Treatment Need in Pediatric Patients with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool between Physicians and Orthodontists

Journal:	<i>BMJ Open</i>
Manuscript ID:	bmjopen-2014-005680.R1
Article Type:	Protocol
Date Submitted by the Author:	28-Jul-2014
Complete List of Authors:	Altalibi, Mostafa; University of Alberta, School of Dentistry, Orthodontic Graduate Program Saltaji, Humam; University of Alberta, School of Dentistry, Orthodontic Graduate Program Roduta Roberts, Mary; University of Alberta, Department of Occupational Therapy, Faculty of Rehabilitation Medicine Major, Michael; University of Alberta, Inter-disciplinary Airway Research Clinic (I-ARC) Macleane, Joanna; University of Alberta, Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital Major, Paul; University of Alberta, School of Dentistry, Orthodontic Graduate Program
Primary Subject Heading:	Dentistry and oral medicine
Secondary Subject Heading:	Paediatrics, Ear, nose and throat/otolaryngology, Health services research
Keywords:	PAEDIATRICS, HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Otolaryngology < SURGERY

SCHOLARONE™
Manuscripts

1
2
3 **Developing an Index for the Orthodontic Treatment Need in Pediatric Patients**
4 **with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool**
5 **between Physicians and Orthodontists**
6
7
8
9

10
11 **Mostafa Altalibi^a; Humam Saltaji^b; Mary Roduta Roberts^c; Michael Major^d; Joanna**
12 **Maclean^e, Paul W Major^f**
13

14
15
16 ^a MSc Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta,
17 Edmonton, Alberta, Canada.

18
19 ^b PhD Candidate and Resident, Orthodontic Graduate Program, School of Dentistry, University of
20 Alberta, Edmonton, Alberta, Canada.

21
22 ^c Assistant Professor, Department of Occupational Therapy, Faculty of Rehabilitation Medicine,
23 University of Alberta, Edmonton, Alberta, Canada.

24
25 ^d Director, Inter-disciplinary Airway Research Clinic (I-ARC), Assistant Clinical Professor,
26 Orthodontics, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

27
28 ^e Medical Director at Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital;
29 Assistant Professor, School of Medicine, University of Alberta, Edmonton, Alberta, Canada.

30
31 ^f Professor and Chair of Dentistry; Senior Associate Dean, Faculty of Medicine and Dentistry,
32 University of Alberta, Edmonton, Alberta, Canada.
33
34
35
36

37 **Corresponding Author**

38 Mostafa Altalibi DMD, MSc (Ortho)
39 Orthodontic Graduate Program
40 School of Dentistry
41 5-476, Edmonton Clinic Health Academy (ECHA)
42 University of Alberta
43 11405-87 Ave, Edmonton, Canada T6G 1C9
44 Phone: 780 885-7457
45 E-mail: altalibi@ualberta.ca
46
47
48
49
50

51
52 **Word Count:** 2807

53
54 **Key Words:** Obstructive Sleep Apnea; Pediatric; Index; Orthodontic Treatment Need; Visual
55 Analogue Scale; Development; Validation; Reliability.
56
57
58
59
60

ABSTRACT

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

INTRODUCTION

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.¹ More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.²⁻⁸ Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.^{9 10 11-13} Other physiologic processes that can be affected include stunted growth,¹⁴ high blood pressure,¹⁵ damage to the heart: ventricular hypertrophy,¹⁶ and cor pulmonale¹⁷.

In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla¹⁸ or mandible, a long lower face height and restriction in the space of the upper airway^{19 20}. Furthermore, when evaluating the polysomnography of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances²¹⁻²⁶ can be of benefit at reducing the severity of OSA. Reverse pull headgear²⁷⁻³², and maxillary & mandibular advancement surgery³³ have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, ect) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment.

Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

METHODS AND ANALYSIS

This research is classified as an applied interdisciplinary medical research. The overall study design involves components of qualitative analysis and quantitative analysis. The qualitative components comprise focus groups and committee meetings. The quantitative components comprise reliability tests and a cross sectional validity test.

A) Initial Development

In accordance with the World Health Organization's recommendations on developing an index,³⁴ development of the index will be achieved through the following objectives:

- 1- *Establishing a Steering Committee*
- 2- *Scoping the index*
- 3- *Reviewing the literature*
- 4- *Drafting the index*
- 5- *Organizing an Index Development Group*
- 6- *Organizing an External Review Group*

Steering Committee

The Steering Committee will be established a priori and is responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index.

Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in question will lead to an improvement in the OSA symptoms. Priority will be given to the results of

1
2
3 well-conducted and well-reported systematic reviews and randomized control trials. Each of the
4 factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will be searched
5 for the craniofacial morphology of pediatric OSA patients. All of this information will be brought
6 back to the Steering Committee for discussion of inclusion / elimination of factors in the index.
7
8
9

10 **Drafting the Index**

11
12 Since this index will have visual-rating scales for each of the craniofacial and occlusal
13 factors, the steering committee will devise an outline suitable for displaying each of the factors
14 representing the index. The factors in the drafted index will also have a number of levels that will
15 divide the factor into categories of severity. The number of levels for a given factor will also be
16 determined by the Steering Committee. Details describing the nature of the illustrations, the amount
17 of levels that each of the factors will have, and the general layout of the index will be agreed upon
18 by the steering committee, and the graphic artist will then design a preliminary version of the index
19 based on the relevant factors and the feedback from the steering committee.
20
21
22
23
24
25
26

27 **The Index Development Meeting**

28
29 The Index Development Group (IDG) is formed by a group of external multidisciplinary
30 experts who provide evidence-based recommendations, on the content, layout and development
31 process of the index. This eclectic group should be small enough to be able to have effective
32 discussions, while large enough to ensure the appropriate representation from all the stakeholders.
33 In this study we plan on including a broad spectrum of medical specialists, family physicians,
34 orthodontists, and methodologists. The goals of this meeting are to gain additional information to
35 strengthen the index, as well as gaining the professional credibility of a broad range of experts on
36 the topic of pediatric OSA.
37
38
39
40
41
42
43

44 Once the members have been chosen by the Steering Committee and accept the invitation to
45 participate, the IDG members will be emailed a document 4 days prior to the meeting, explaining:
46 the purpose of the index and meeting, how the meeting will proceed, what to expect in the meeting,
47 and a brief literature review of the orthodontic techniques currently available to help with the
48 symptoms of OSA. Moreover, every member will be randomly assigned a number in order to
49 maintain anonymity of the responses.
50
51
52
53
54

55 **Procedure of the IDG Meeting**

1
2
3 The meeting will commence with a brief introduction summarizing the literature review, the
4 purpose of the index and its relevance. The meeting will then proceed to collect feedback
5 (APPENDIX 1) on the aforementioned chosen factors using a modified Delphi technique^{35 36},
6 which is a communication technique that structures the meeting and minimizes bias in responses.
7
8 Participants' responses will be collected through a web-based response portal. Each factor will be
9 explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based on
10 its importance for decision-making. Once all the feedback is received on a particular factor, the
11 summary of the results will be displayed for everyone to see. Discussion will ensue for a maximum
12 of 5 minutes, with each person talking no more than 1 minute. Then everyone will be asked to re-
13 enter their feedback on the website for the same factor in light of the discussion. This cycle will
14 continue until a consensus of greater than 80% of the members is reached. Once consensus is
15 reached, then we will move on to the next factor and the participants will be asked to give feedback
16 on this new factor in a similar manner.
17
18
19
20
21
22
23
24
25

26 In addition, two negative control factors will be used to calibrate the responses. The first
27 negative control will be of orthodontic relevance but have no effect on the amelioration of the
28 symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of
29 relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck
30 circumference).
31
32
33
34
35

36 Once all the factors have been discussed and consensus reached, we will get feedback
37 through the website on the: alignment - importance of the index, and if the development process is
38 appropriate; relevance – content analysis, and whether all the factors identified are important;
39 representation – if there is anything that needs to be added to the index (APPENDIX 2). Finally,
40 additional written feedback will be accepted at the very end of the session.
41
42
43
44

45 All the feedback from the IDM will be summarized and presented to the Steering
46 Committee, and decisions will be made to remove factors, modify factors, and /or modify the
47 outline and layout of the index. These modifications will subsequently be presented to the graphic
48 artist and a second draft will be procured.
49
50
51

52 **The External Review Group Meeting**

53
54
55 The External Review Group (ERG) is composed of end-users and interested parties. This
56 group is not responsible for any content analysis, instead it will be responsible for reviewing the
57
58
59
60

1
2
3 layout, simplicity and ease of use. It will also be responsible to assess usefulness of the index in the
4 healthcare setting and give feedback on the feasibility of implementing the index in practice. The
5 goal of this meeting is to gain the end-user approval for the ease and feasibility of administering the
6 index. This group should be large enough to be a representative sample of the population, yet small
7 enough to allow for ease of explanation and healthy discussion. It is not as structured as the IDM,
8 and allows for the participants to freely express their opinions in an open forum.
9
10
11
12

13
14 In this meeting we will explain the theory behind the index by briefly reviewing the
15 literature and then explain the purpose of the index. Subsequently we will show the group a pilot
16 version of the index and a paper will be distributed to receive for their feedback based on the
17 following questions:
18
19
20

- 21 1- Do you understand the purpose of the index?
- 22 2- Do you understand what each factor is assessing?
 - 23 a. If not which one(s) do you not understand and why?
- 24 3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
- 25 4- Would you use this index in your practice?
- 26 5- Other recommendations:
27
28
29
30
31

32 All the feedback from the ERG will be summarized and presented to the Steering
33 Committee, and decisions will be made to modify the outline and layout of the index. These
34 modifications will subsequently be presented to the graphic artist and a third draft will be procured.
35
36
37

38 **B) Reliability**

39
40 Reliability of the index will be tested within a group that represents the typical end-user
41 population. This includes physicians of family medicine, pediatricians, pediatric ENT, or pediatric
42 sleep physicians. Reliability will also be assessed within a group of orthodontists, who by their
43 training are experts at assessing malocclusion and craniofacial morphology. Therefore 10-20
44 physicians and orthodontists will be recruited as examiners in this study.
45
46
47
48
49

50 A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-
51 disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and their
52 intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief
53 explanation about the use and application of the index, the physicians as well as the orthodontists
54 will apply the developed index to the sample patient pool's pictures once. In order to diminish
55
56
57
58
59
60

1
2
3 recall bias, application of the index will be repeated 2 weeks later.
4

5 Intra-rater reliability and inter-rater reliability between the physicians, between the
6 orthodontists and between the physicians and orthodontists will be compared. The reliability will be
7 assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.
8
9

10 11 **C) Validation** 12

13 The Index will be validated using dichotomous responses from orthodontists on whether
14 specific patients would require orthodontic treatment to help their obstructive sleep apnea
15 symptoms and comparing it to the score that the index gave those same patients. This will be
16 achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric
17 OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral
18 pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic diagnosed
19 with OSA, and the orthodontists will be asked to rate these patients, using a “yes” or “no” response,
20 whether they would benefit from orthodontic treatment for their OSA symptoms. The index will
21 then be applied on the same patients by the principal investigator and the score of each patient will
22 be recorded. Using a stepwise multiple-logistic regression each of the identified factors will be
23 given a weight; this will represent the relative importance of the factor. Once analyzed if the
24 correlation is high between the expert scores and the cluster groups, then the clusters are
25 meaningful and valid. Furthermore, a cutoff for most efficient score above which to refer will be
26 chosen using a graph and observing the value that optimizes the sensitivity, specificity and overall
27 accuracy of the index. Finally, four grades of treatment-need will be determined using the twenty-
28 five percentile ranges. The grades will be:
29
30
31
32
33
34
35
36
37
38
39
40

- 41 1- Minimal Need
 - 42 2- Mild Need
 - 43 3- Moderate Need
 - 44 4- Severe Need
- 45
46
47
48

49 **DISCUSSION** 50

51 The development plan of this index has been conceived through a modification of the WHO
52 Handbook for guideline development,³⁴ as well as reviewing the orthodontic literature for ways
53 indices have been previously developed. The WHO provided an excellent starting point, from there
54 modifications were made to cater to the specifics of this study, given that there are differences
55
56
57
58
59
60

1
2
3 between developing a guideline and an index for orthodontic treatment need. The literature was
4 useful, and among the index development protocols reviewed, certain assessed the orthodontic
5 treatment need within the entire population,³⁷⁻⁴⁰ while others assessed it for a given subpopulation⁴¹⁻
6
7⁴⁶; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a
8
9 protocol for our particular needs from the available literature and using the experience and expertise
10 of the authors. Through this protocol we aim to develop an index that fulfills all of the following
11
12 criteria:⁴⁷
13
14

- 15 1. Gradient of Numeric Values: The severity of the orthodontic treatment need within
16 the pediatric OSA patients should be defined within a numerical scheme that
17 demonstrates a finite and progressive gradient from low need to high need.
- 18 2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
- 19 3. Clinical Importance: The numerical scale should correspond with the clinically
20 appraised orthodontic treatment need of pediatric OSA patients.
- 21 4. Statistical Ease: should be amendable to statistical analysis.
- 22 5. Reliability: Should have a high intra- and inter-rater reliability.
- 23 6. Practical: The instruments required to score the index should be practical to the
24 setting in which it will be administered.
- 25 7. Minimal Judgment: Applying the index should require minimal judgment.
- 26 8. Simple: The index shouldn't have a high financial or time cost, therefore should be
27 simple enough to administer to many patients.
- 28 9. Detect Change: The index should be able to detect changes in orthodontic treatment
29 need in pediatric OSA patients.
- 30 10. Validity: should be valid over time.

31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60
Validity can be characterized into different types: Face Validity, Content Validity, Construct
Validity, and Criterion Validity.⁴⁸ In this study, we will examine these kinds of validity at different
stages of development. The first draft of the index will focus on establishing face validity.
Feedback from the steering committee and IDG will assist in establishing content and construct
validity. Assuming that the “gold standard” in assessing the orthodontic treatment-need in pediatric
patients with OSA is an orthodontist with experience in dealing with pediatric OSA patients, then
the subsequent modification of the index based on the reliability tests and the dichotomous
responses from the orthodontists provide the index with the necessary criterion-related validity

1
2
3 evidence through statistical means.⁴⁹
4

5 **Significance**

6
7 This index will help physicians and other medical professionals identify which craniofacial
8 phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA
9 management. Furthermore, due to the diverse medical effects of sleep deprivation, there will be a
10 trend to make sleep apnea into a centralized service, where the main focus is for a highly trained
11 multidisciplinary team to treat a high volume of patients to a standardized protocol, where
12 meticulous documentation is exercised. This index is part of that documentation process. It will
13 allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will
14 also allow long-term follow up and audit in order to enter into comparisons with other centers.
15
16
17
18
19
20

21 **Dissemination Plan**

22
23 The dissemination of this index will be done through a variety of ways in order to maximize
24 its reach. Primarily, it will be published in a peer-reviewed journal, which will allow its
25 introduction to the scientific literature. The journal should be a respected medical journal with
26 broad reach, in order to allow the greatest number of physicians to be exposed to the index.
27 Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian
28 and American population of medical professionals. Moreover, the index will be presented at
29 national and international conferences to increase the awareness of the index among the scientific
30 community. Finally, the index will be used at the University of Alberta's Interdisciplinary Airway
31 Research Center, and more research, so that future research in this center will incorporate it. It will
32 also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website
33 under the physician section, to further educate the doctors who visit the site on the index.
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Funding

No funding has been provided for this project.

Authors' Contributions

MA HS MRR MM JM PM conceived and designed the study; MA drafted the manuscript and integrated critical feedback from all of the other authors. All of the authors approved the final version of the manuscript.

Ethics approval

The proposed research has received ethical approval numbered Pro00045067 from the University of Alberta Ethics Board.

Competing Interests

The authors declare no conflicts of interests.

REFERENCES

- 1 O'Brien LM, Mervis CB, Holbrook CR, et al. Neurobehavioral implications of habitual snoring in children. *Pediatrics* 2004;114(1):44-9.
- 2 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the American Thoracic Society* 2008;5(2):242-52.
- 3 Gislason T, Benediktsdottir B. Snoring, apneic episodes, and nocturnal hypoxemia among children 6 months to 6 years old. An epidemiologic study of lower limit of prevalence. *Chest* 1995;107(4):963-6.
- 4 Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds. *Archives of disease in childhood* 1993;68(3):360-6.
- 5 Redline S, Tishler PV, Schluchter M, et al. Risk factors for sleep-disordered breathing in children. Associations with obesity, race, and respiratory problems. *American journal of respiratory and critical care medicine* 1999;159(5 Pt 1):1527-32.
- 6 Bixler EO, Vgontzas AN, Lin HM, et al. Sleep disordered breathing in children in a general population sample: prevalence and risk factors. *Sleep* 2009;32(6):731-6.
- 7 Li AM, So HK, Au CT, Ho C, et al. Epidemiology of obstructive sleep apnoea syndrome in Chinese children: a two-phase community study. *Thorax* 2010;65(11):991-7.
- 8 O'Brien LM, Holbrook CR, Mervis CB, et al. Sleep and neurobehavioral characteristics of 5- to 7-year-old children with parentally reported symptoms of attention-deficit/hyperactivity disorder. *Pediatrics* 2003;111(3):554-63.
- 9 Huynh NT, Morton PD, Rompre PH, et al. Associations between sleep-disordered breathing symptoms and facial and dental morphometry, assessed with screening examinations. *American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics* 2011;140(6):762-70.
- 10 Beebe DW. Neural and neurobehavioral dysfunction in children with obstructive sleep apnea. *PLoS medicine* 2006;3(8):e323.
- 11 Chervin RD, Archbold KH, Dillon JE, et al. Inattention, hyperactivity, and symptoms of sleep-disordered breathing. *Pediatrics* 2002;109(3):449-56.
- 12 Crabtree VM, Varni JW, Gozal D. Health-related quality of life and depressive symptoms in children with suspected sleep-disordered breathing. *Sleep* 2004;27(6):1131-8.

- 1
2
3 .13Halbower AC, Degaonkar M, Barker PB, et al. Childhood obstructive sleep apnea associates
4 with neuropsychological deficits and neuronal brain injury. *PLoS medicine* 2006;3(8):e301.
5
6 .14Bar A, Tarasiuk A, Segev Y, et al. The effect of adenotonsillectomy on serum insulin-like
7 growth factor-I and growth in children with obstructive sleep apnea syndrome. *The Journal*
8 *of pediatrics* 1999;135(1):76-80.
9
10 .15Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea.
11 *American journal of respiratory and critical care medicine* 1998;157(4 Pt 1):103-109.
12
13 .16Amin RS, Kimball TR, Bean JA, et al. Left ventricular hypertrophy and abnormal ventricular
14 geometry in children and adolescents with obstructive sleep apnea. *American journal of*
15 *respiratory and critical care medicine* 2002;165(10):1395-9.
16
17 .17Sofer S, Weinhouse E, Tal A, Wanderman KL, et al. Cor pulmonale due to adenoidal or
18 tonsillar hypertrophy or both in children. Noninvasive diagnosis and follow-up. *Chest*
19 1988;93(1):119-22.
20
21 .18Korayem MM, Witmans M, MacLean J, et al. Craniofacial morphology in pediatric patients with
22 persistent obstructive sleep apnea with or without positive airway pressure therapy: a cross-
23 sectional cephalometric comparison with controls. *American journal of orthodontics and*
24 *dentofacial orthopedics : official publication of the American Association of Orthodontists,*
25 *its constituent societies, and the American Board of Orthodontics* 2013;144(1):78-85.
26
27 .19Katyal V, Pamula Y, Martin AJ, et al. Craniofacial and upper airway morphology in pediatric
28 sleep-disordered breathing: Systematic review and meta-analysis. *American journal of*
29 *orthodontics and dentofacial orthopedics : official publication of the American Association*
30 *of Orthodontists, its constituent societies, and the American Board of Orthodontics*
31 2013;143(1):20-30 e3.
32
33 .20Flores-Mir C, Korayem M, Heo G, Witmans M, Major MP, Major PW. Craniofacial
34 morphological characteristics in children with obstructive sleep apnea syndrome: a
35 systematic review and meta-analysis. *Journal of the American Dental Association*
36 2013;144(3):269-77.
37
38 .21Carvalho FR, Lentini-Oliveira D, Machado MA, et al. Oral appliances and functional
39 orthopaedic appliances for obstructive sleep apnoea in children. *The Cochrane database of*
40 *systematic reviews* 2007(2):CD005520.
41
42 .22Villa MP, Bernkopf E, Pagani J, et al. Randomized controlled study of an oral jaw-positioning
43 appliance for the treatment of obstructive sleep apnea in children with malocclusion.
44 *American journal of respiratory and critical care medicine* 2002;165(1):123-7.
45
46 .23Schutz TC, Dominguez GC, Hallinan MP, et al. Class II correction improves nocturnal
47 breathing in adolescents. *The Angle orthodontist* 2011;81(2):222-8.
48
49 .24Cozza P, Polimeni A, Ballanti F. A modified monobloc for the treatment of obstructive sleep
50 apnoea in paediatric patients. *Eur J Orthod* 2004;26(5):523-30.
51
52
53
54
55
56
57
58
59
60

- 1
2
3 .25Zhang C, He H, Ngan P. Effects of twin block appliance on obstructive sleep apnea in children:
4 a preliminary study. *Sleep & breathing = Schlaf & Atmung* 2013;17(4):1309-14.
5
6 .26Pliska BT, Almeida F. Effectiveness and outcome of oral appliance therapy. *Dental clinics of*
7 *North America* 2012;56(2):433-44.
8
9 .27Hiyama S, Suda N, Ishii-Suzuki M, et al. Effects of maxillary protraction on craniofacial
10 structures and upper-airway dimension. *The Angle orthodontist* 2002;72(1):43-7.
11
12 .28Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway
13 passage. *The Angle orthodontist* 2008;78(2):209-14.
14
15 .29Kilinc AS, Arslan SG, Kama JD, et al. Effects on the sagittal pharyngeal dimensions of
16 protraction and rapid palatal expansion in Class III malocclusion subjects. *Eur J Orthod*
17 2008;30(1):61-6.
18
19 .30Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot
20 study. *Eur J Orthod* 2006;28(2):184-9.
21
22 .31Kaygisiz E, Tuncer BB, Yuksel S, et al. effects of maxillary protraction and fixed appliance
23 therapy on the pharyngeal airway. *The Angle orthodontist* 2009;79(4):660-7.
24
25 .32Lee JW, Park KH, Kim SH, et al. Correlation between skeletal changes by maxillary protraction
26 and upper airway dimensions. *The Angle orthodontist* 2011;81(3):426-32.
27
28 .33Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive
29 sleep apnea: a systematic review and meta-analysis. *Sleep medicine reviews* 2010;14(5):287-
30 97.
31
32 .34World Health Organization (2012) WHO Handbook for guideline development. Available:
33 http://www.who.int/hiv/topics/mtct/grc_handbook_mar2010_1.pdf. Accessed 10 Sept 2013.
34
35 .35Rowe G, Wright G. The Delphi technique as a forecasting tool: issues and analysis. *Int J*
36 *Forecasting* 1999;15(4):353-75.
37
38 .36Dalkey N, Helmer O. An Experimental Application of the Delphi Method to the use of experts.
39 *Management Science* 1963;9(3):458-67.
40
41 .37Richmond S, Shaw WC, O'Brien KD, et al. The development of the PAR Index (Peer
42 Assessment Rating): reliability and validity. *Eur J Orthod* 1992;14(2):125-39.
43
44 .38Richmond S, Shaw WC, Roberts CT, et al. The PAR Index (Peer Assessment Rating): methods
45 to determine outcome of orthodontic treatment in terms of improvement and standards. *Eur*
46 *J Orthod* 1992;14(3):180-7.
47
48 .39Daniels C, Richmond S. The development of the index of complexity, outcome and need
49 (ICON). *J Orthod* 2000;27(2):149-62.
50
51
52
53
54
55
56
57
58
59
60

- 1
2
3 .40Brook PH, Shaw WC. The development of an index of orthodontic treatment priority. *Eur J*
4 *Orthod* 1989;11(3):309-20.
5
6 .41Mars M, Batra P, Worrell E. Complete unilateral cleft lip and palate: validity of the five-year
7 index and the Goslon yardstick in predicting long-term dental arch relationships. *Cleft*
8 *Palate Craniofac J* 2006;43(5):557-62.
9
10 .42Mars M, Plint DA, Houston WJ, et al. The Goslon Yardstick: a new system of assessing dental
11 arch relationships in children with unilateral clefts of the lip and palate. *Cleft Palate J*
12 1987;24(4):314-22.
13
14 .43Johnson N, Sandy J. An aesthetic index for evaluation of cleft repair. *Eur J Orthod*
15 2003;25(3):243-9.
16
17 .44Mossey PA, Clark JD, Gray D. Preliminary investigation of a modified Huddart/Bodenham
18 scoring system for assessment of maxillary arch constriction in unilateral cleft lip and palate
19 subjects. *Eur J Orthod* 2003;25(3):251-7.
20
21 .45Tohill C, Mossey PA. Assessment of arch constriction in patients with bilateral cleft lip and
22 palate and isolated cleft palate: a pilot study. *Eur J Orthod* 2007;29(2):193-7.
23
24 .46Huddart AG, Bodenham RS. The evaluation of arch form and occlusion in unilateral cleft palate
25 subjects. *Cleft Palate J* 1972;9:194-209.
26
27 .47Summers CJ. The occlusal index: a system for identifying and scoring occlusal disorders. *Am J*
28 *Orthod* 1971;59(6):552-67.
29
30 .48Streiner DL, Norman GR. *Health measurement scales: a practical guide to their development*
31 *and use*. Oxford: Oxford Medical Publications, 1995.
32
33 .49Terwee CB, Bot SDM, de Boer MR, et al. Quality criteria were proposed for measurement
34 properties of health status questionnaires. *J Clin Epidemiol* 2007;60(1):34-42.
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3 **Developing an Index for the Orthodontic Treatment Need in Pediatric Patients**
4 **with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool**
5 **between Physicians and Orthodontists**
6
7
8
9

10
11 **Mostafa Altalibi^a; Humam Saltaji^b; Mary Roduta Roberts^c; Michael Major^d; Joanna**
12 **Maclean^e, Paul W Major^f**
13

14
15
16 ^a MSc Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta,
17 Edmonton, Alberta, Canada.

18
19 ^b PhD Candidate and Resident, Orthodontic Graduate Program, School of Dentistry, University of
20 Alberta, Edmonton, Alberta, Canada.

21
22 ^c Assistant Professor, Department of Occupational Therapy, Faculty of Rehabilitation Medicine,
23 University of Alberta, Edmonton, Alberta, Canada.

24
25 ^d Director, Inter-disciplinary Airway Research Clinic (I-ARC), Assistant Clinical Professor,
26 Orthodontics, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

27
28 ^e Medical Director at Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital;
29 Assistant Professor, School of Medicine, University of Alberta, Edmonton, Alberta, Canada.

30
31 ^f Professor and Chair of Dentistry; Senior Associate Dean, Faculty of Medicine and Dentistry,
32 University of Alberta, Edmonton, Alberta, Canada.
33
34
35
36

37 **Corresponding Author**

38 Mostafa Altalibi DMD, MSc (Ortho)
39 Orthodontic Graduate Program
40 School of Dentistry
41 5-476, Edmonton Clinic Health Academy (ECHA)
42 University of Alberta
43 11405-87 Ave, Edmonton, Canada T6G 1C9
44 Phone: 780 885-7457
45 E-mail: altalibi@ualberta.ca
46
47
48
49
50

51
52 **Word Count:** 2807

53
54 **Key Words:** Obstructive Sleep Apnea; Pediatric; Index; Orthodontic Treatment Need; Visual
55 Analogue Scale; Development; Validation; Reliability.
56
57
58
59
60

ABSTRACT

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

INTRODUCTION

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.¹ More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.²⁻⁸ Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.^{9 10 11-13} Other physiologic processes that can be affected include stunted growth,¹⁴ high blood pressure,¹⁵ damage to the heart: ventricular hypertrophy,¹⁶ and cor pulmonale¹⁷.

In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla¹⁸ or mandible, a long lower face height and restriction in the space of the upper airway^{19 20}. Furthermore, when evaluating the polysomnography of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances²¹⁻²⁶ can be of benefit at reducing the severity of OSA. Reverse pull headgear²⁷⁻³², and maxillary & mandibular advancement surgery³³ have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, ect) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment.

Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

METHODS AND ANALYSIS

This research is classified as an applied interdisciplinary medical research. The overall study design involves components of qualitative analysis and quantitative analysis. The qualitative components comprise focus groups and committee meetings. The quantitative components comprise reliability tests and a cross sectional validity test.

A) Initial Development

In accordance with the World Health Organization's recommendations on developing an index,³⁴ development of the index will be achieved through the following objectives:

- 1- *Establishing a Steering Committee*
- 2- *Scoping the index*
- 3- *Reviewing the literature*
- 4- *Drafting the index*
- 5- *Organizing an Index Development Group*
- 6- *Organizing an External Review Group*

Steering Committee

The Steering Committee will be established a priori and is responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index.

Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in question will lead to an improvement in the OSA symptoms. Priority will be given to the results of

1
2
3 well-conducted and well-reported systematic reviews and randomized control trials. Each of the
4 factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will be searched
5 for the craniofacial morphology of pediatric OSA patients. All of this information will be brought
6 back to the Steering Committee for discussion of inclusion / elimination of factors in the index.
7
8
9

10 **Drafting the Index**

11
12 Since this index will have visual-rating scales for each of the craniofacial and occlusal
13 factors, the steering committee will devise an outline suitable for displaying each of the factors
14 representing the index. The factors in the drafted index will also have a number of levels that will
15 divide the factor into categories of severity. The number of levels for a given factor will also be
16 determined by the Steering Committee. Details describing the nature of the illustrations, the amount
17 of levels that each of the factors will have, and the general layout of the index will be agreed upon
18 by the steering committee, and the graphic artist will then design a preliminary version of the index
19 based on the relevant factors and the feedback from the steering committee.
20
21
22
23
24
25
26

27 **The Index Development Meeting**

28
29 The Index Development Group (IDG) is formed by a group of external multidisciplinary
30 experts who provide evidence-based recommendations, on the content, layout and development
31 process of the index. This eclectic group should be small enough to be able to have effective
32 discussions, while large enough to ensure the appropriate representation from all the stakeholders.
33 In this study we plan on including a broad spectrum of medical specialists, family physicians,
34 orthodontists, and methodologists. The goals of this meeting are to gain additional information to
35 strengthen the index, as well as gaining the professional credibility of a broad range of experts on
36 the topic of pediatric OSA.
37
38
39
40
41
42
43

44 Once the members have been chosen by the Steering Committee and accept the invitation to
45 participate, the IDG members will be emailed a document 4 days prior to the meeting, explaining:
46 the purpose of the index and meeting, how the meeting will proceed, what to expect in the meeting,
47 and a brief literature review of the orthodontic techniques currently available to help with the
48 symptoms of OSA. Moreover, every member will be randomly assigned a number in order to
49 maintain anonymity of the responses.
50
51
52
53
54

55 **Procedure of the IDG Meeting**

1
2
3 The meeting will commence with a brief introduction summarizing the literature review, the
4 purpose of the index and its relevance. The meeting will then proceed to collect feedback
5 (APPENDIX 1) on the aforementioned chosen factors using a modified Delphi technique^{35 36} ,
6 which is a communication technique that structures the meeting and minimizes bias in responses.
7
8 Participants' responses will be collected through a web-based response portal. Each factor will be
9 explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based on
10 its importance for decision-making. Once all the feedback is received on a particular factor, the
11 summary of the results will be displayed for everyone to see. Discussion will ensue for a maximum
12 of 5 minutes, with each person talking no more than 1 minute. Then everyone will be asked to re-
13 enter their feedback on the website for the same factor in light of the discussion. This cycle will
14 continue until a consensus of greater than 80% of the members is reached. Once consensus is
15 reached, then we will move on to the next factor and the participants will be asked to give feedback
16 on this new factor in a similar manner.
17
18
19
20
21
22
23
24
25

26 In addition, two negative control factors will be used to calibrate the responses. The first
27 negative control will be of orthodontic relevance but have no effect on the amelioration of the
28 symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of
29 relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck
30 circumference).
31
32
33
34
35

36 Once all the factors have been discussed and consensus reached, we will get feedback
37 through the website on the: alignment - importance of the index, and if the development process is
38 appropriate; relevance – content analysis, and whether all the factors identified are important;
39 representation – if there is anything that needs to be added to the index (APPENDIX 2). Finally,
40 additional written feedback will be accepted at the very end of the session.
41
42
43
44

45 All the feedback from the IDM will be summarized and presented to the Steering
46 Committee, and decisions will be made to remove factors, modify factors, and /or modify the
47 outline and layout of the index. These modifications will subsequently be presented to the graphic
48 artist and a second draft will be procured.
49
50
51

52 **The External Review Group Meeting**

53
54
55 The External Review Group (ERG) is composed of end-users and interested parties. This
56 group is not responsible for any content analysis, instead it will be responsible for reviewing the
57
58
59
60

1
2
3 layout, simplicity and ease of use. It will also be responsible to assess usefulness of the index in the
4 healthcare setting and give feedback on the feasibility of implementing the index in practice. The
5 goal of this meeting is to gain the end-user approval for the ease and feasibility of administering the
6 index. This group should be large enough to be a representative sample of the population, yet small
7 enough to allow for ease of explanation and healthy discussion. It is not as structured as the IDM,
8 and allows for the participants to freely express their opinions in an open forum.
9
10
11
12

13
14 In this meeting we will explain the theory behind the index by briefly reviewing the
15 literature and then explain the purpose of the index. Subsequently we will show the group a pilot
16 version of the index and a paper will be distributed to receive for their feedback based on the
17 following questions:
18
19
20

- 21 1- Do you understand the purpose of the index?
- 22 2- Do you understand what each factor is assessing?
 - 23 a. If not which one(s) do you not understand and why?
- 24 3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
- 25 4- Would you use this index in your practice?
- 26 5- Other recommendations:
27
28
29
30
31

32 All the feedback from the ERG will be summarized and presented to the Steering
33 Committee, and decisions will be made to modify the outline and layout of the index. These
34 modifications will subsequently be presented to the graphic artist and a third draft will be procured.
35
36
37

38 **B) Reliability**

39
40 Reliability of the index will be tested within a group that represents the typical end-user
41 population. This includes physicians of family medicine, pediatricians, pediatric ENT, or pediatric
42 sleep physicians. Reliability will also be assessed within a group of orthodontists, who by their
43 training are experts at assessing malocclusion and craniofacial morphology. Therefore 10-20
44 physicians and orthodontists will be recruited as examiners in this study.
45
46
47
48

49
50 A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-
51 disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and their
52 intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief
53 explanation about the use and application of the index, the physicians as well as the orthodontists
54 will apply the developed index to the sample patient pool's pictures once. In order to diminish
55
56
57
58
59
60

1
2
3 recall bias, application of the index will be repeated 2 weeks later.
4

5 Intra-rater reliability and inter-rater reliability between the physicians, between the
6 orthodontists and between the physicians and orthodontists will be compared. The reliability will be
7 assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.
8
9

10 11 **C) Validation** 12

13 The Index will be validated using dichotomous responses from orthodontists on whether
14 specific patients would require orthodontic treatment to help their obstructive sleep apnea
15 symptoms and comparing it to the score that the index gave those same patients. This will be
16 achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric
17 OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral
18 pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic diagnosed
19 with OSA, and the orthodontists will be asked to rate these patients, using a “yes” or “no” response,
20 whether they would benefit from orthodontic treatment for their OSA symptoms. The index will
21 then be applied on the same patients by the principal investigator and the score of each patient will
22 be recorded. Using a stepwise multiple-logistic regression each of the identified factors will be
23 given a weight; this will represent the relative importance of the factor. Once analyzed if the
24 correlation is high between the expert scores and the cluster groups, then the clusters are
25 meaningful and valid. Furthermore, a cutoff for most efficient score above which to refer will be
26 chosen using a graph and observing the value that optimizes the sensitivity, specificity and overall
27 accuracy of the index. Finally, four grades of treatment-need will be determined using the twenty-
28 five percentile ranges. The grades will be:
29
30
31
32
33
34
35
36
37
38
39
40
41

- 42 1- Minimal Need
 - 43 2- Mild Need
 - 44 3- Moderate Need
 - 45 4- Severe Need
- 46
47
48

49 **DISCUSSION** 50

51 The development plan of this index has been conceived through a modification of the WHO
52 Handbook for guideline development,³⁴ as well as reviewing the orthodontic literature for ways
53 indices have been previously developed. The WHO provided an excellent starting point, from there
54 modifications were made to cater to the specifics of this study, given that there are differences
55
56
57
58
59
60

1
2
3 between developing a guideline and an index for orthodontic treatment need. The literature was
4 useful, and among the index development protocols reviewed, certain assessed the orthodontic
5 treatment need within the entire population,³⁷⁻⁴⁰ while others assessed it for a given subpopulation⁴¹⁻
6
7⁴⁶; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a
8
9 protocol for our particular needs from the available literature and using the experience and expertise
10 of the authors. Through this protocol we aim to develop an index that fulfills all of the following
11
12 criteria:⁴⁷
13
14

- 15 1. Gradient of Numeric Values: The severity of the orthodontic treatment need within
16 the pediatric OSA patients should be defined within a numerical scheme that
17 demonstrates a finite and progressive gradient from low need to high need.
18
- 19 2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
20
- 21 3. Clinical Importance: The numerical scale should correspond with the clinically
22 appraised orthodontic treatment need of pediatric OSA patients.
23
- 24 4. Statistical Ease: should be amendable to statistical analysis.
25
- 26 5. Reliability: Should have a high intra- and inter-rater reliability.
27
- 28 6. Practical: The instruments required to score the index should be practical to the
29 setting in which it will be administered.
30
- 31 7. Minimal Judgment: Applying the index should require minimal judgment.
32
- 33 8. Simple: The index shouldn't have a high financial or time cost, therefore should be
34 simple enough to administer to many patients.
35
- 36 9. Detect Change: The index should be able to detect changes in orthodontic treatment
37 need in pediatric OSA patients.
38
- 39 10. Validity: should be valid over time.
40
41
42
43

44 Validity can be characterized into different types: Face Validity, Content Validity, Construct
45 Validity, and Criterion Validity.⁴⁸ In this study, we will examine these kinds of validity at different
46 stages of development. The first draft of the index will focus on establishing face validity.
47 Feedback from the steering committee and IDG will assist in establishing content and construct
48 validity. Assuming that the "gold standard" in assessing the orthodontic treatment-need in pediatric
49 patients with OSA is an orthodontist with experience in dealing with pediatric OSA patients, then
50 the subsequent modification of the index based on the reliability tests and the dichotomous
51 responses from the orthodontists provide the index with the necessary criterion-related validity
52
53
54
55
56
57
58
59
60

1
2
3 evidence through statistical means.⁴⁹
4

5 **Significance**

6
7 This index will help physicians and other medical professionals identify which craniofacial
8 phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA
9 management. Furthermore, due to the diverse medical effects of sleep deprivation, there will be a
10 trend to make sleep apnea into a centralized service, where the main focus is for a highly trained
11 multidisciplinary team to treat a high volume of patients to a standardized protocol, where
12 meticulous documentation is exercised. This index is part of that documentation process. It will
13 allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will
14 also allow long-term follow up and audit in order to enter into comparisons with other centers.
15
16
17
18
19
20

21 **Dissemination Plan**

22
23 The dissemination of this index will be done through a variety of ways in order to maximize
24 its reach. Primarily, it will be published in a peer-reviewed journal, which will allow its
25 introduction to the scientific literature. The journal should be a respected medical journal with
26 broad reach, in order to allow the greatest number of physicians to be exposed to the index.
27 Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian
28 and American population of medical professionals. Moreover, the index will be presented at
29 national and international conferences to increase the awareness of the index among the scientific
30 community. Finally, the index will be used at the University of Alberta's Interdisciplinary Airway
31 Research Center, and more research, so that future research in this center will incorporate it. It will
32 also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website
33 under the physician section, to further educate the doctors who visit the site on the index.
34
35
36
37
38
39
40
41
42
43

44 **Funding**

45
46 No funding has been provided for this project.
47
48

49 **Authors' Contributions**

50
51 MA HS MRR MM JM PM conceived and designed the study; MA drafted the manuscript and
52 integrated critical feedback from all of the other authors. All of the authors approved the final
53 version of the manuscript.
54
55

56 **Ethics approval**

The proposed research has received ethical approval numbered Pro00045067 from the University of Alberta Ethics Board.

Competing Interests

The authors declare no conflicts of interests.

REFERENCES

- .1 O'Brien LM, Mervis CB, Holbrook CR, Bruner JL, Klaus CJ, Rutherford J, et al. Neurobehavioral implications of habitual snoring in children. *Pediatrics* 2004;114(1):44-9.
- .2 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the American Thoracic Society* 2008;5(2):242-52.
- .3 Gislason T, Benediktsdottir B. Snoring, apneic episodes, and nocturnal hypoxemia among children 6 months to 6 years old. An epidemiologic study of lower limit of prevalence. *Chest* 1995;107(4):963-6.
- .4 Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds. *Archives of disease in childhood* 1993;68(3):360-6.
- .5 Redline S, Tishler PV, Schluchter M, Aylor J, Clark K, Graham G. Risk factors for sleep-disordered breathing in children. Associations with obesity, race, and respiratory problems. *American journal of respiratory and critical care medicine* 1999;159(5 Pt 1):1527-32.
- .6 Bixler EO, Vgontzas AN, Lin HM, Liao D, Calhoun S, Vela-Bueno A, et al. Sleep disordered breathing in children in a general population sample: prevalence and risk factors. *Sleep* 2009;32(6):731-6.
- .7 Li AM, So HK, Au CT, Ho C, Lau J, Ng SK, et al. Epidemiology of obstructive sleep apnoea syndrome in Chinese children: a two-phase community study. *Thorax* 2010;65(11):991-7.
- .8 O'Brien LM, Holbrook CR, Mervis CB, Klaus CJ, Bruner JL, Raffield TJ, et al. Sleep and neurobehavioral characteristics of 5- to 7-year-old children with parentally reported symptoms of attention-deficit/hyperactivity disorder. *Pediatrics* 2003;111(3):554-63.
- .9 Huynh NT, Morton PD, Rompre PH, Papadakis A, Remise C. Associations between sleep-disordered breathing symptoms and facial and dental morphometry, assessed with screening examinations. *American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics* 2011;140(6):762-70.
- .10 Beebe DW. Neural and neurobehavioral dysfunction in children with obstructive sleep apnea. *PLoS medicine* 2006;3(8):e323.

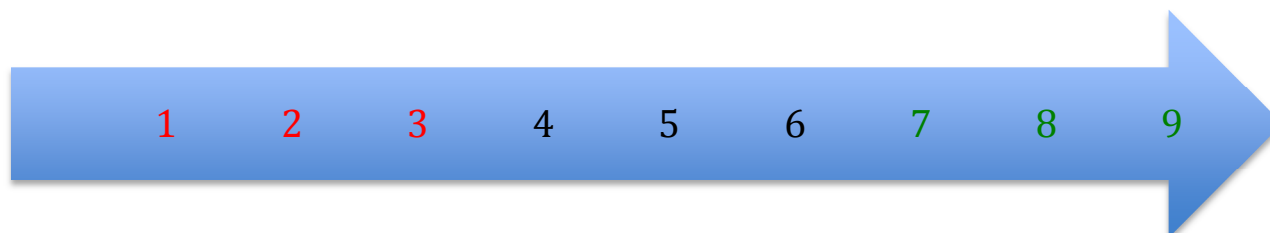
- 1
2
3 .11Chervin RD, Archbold KH, Dillon JE, Panahi P, Pituch KJ, Dahl RE, et al. Inattention,
4 hyperactivity, and symptoms of sleep-disordered breathing. *Pediatrics* 2002;109(3):449-56.
5
6 .12Crabtree VM, Varni JW, Gozal D. Health-related quality of life and depressive symptoms in
7 children with suspected sleep-disordered breathing. *Sleep* 2004;27(6):1131-8.
8
9 .13Halbower AC, Degaonkar M, Barker PB, Earley CJ, Marcus CL, Smith PL, et al. Childhood
10 obstructive sleep apnea associates with neuropsychological deficits and neuronal brain
11 injury. *PLoS medicine* 2006;3(8):e301.
12
13 .14Bar A, Tarasiuk A, Segev Y, Phillip M, Tal A. The effect of adenotonsillectomy on serum
14 insulin-like growth factor-I and growth in children with obstructive sleep apnea syndrome.
15 *The Journal of pediatrics* 1999;135(1):76-80.
16
17 .15Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea.
18 *American journal of respiratory and critical care medicine* 1998;157(4 Pt 1):103-1098:(
19
20 .16Amin RS, Kimball TR, Bean JA, Jeffries JL, Willging JP, Cotton RT, et al. Left ventricular
21 hypertrophy and abnormal ventricular geometry in children and adolescents with obstructive
22 sleep apnea. *American journal of respiratory and critical care medicine* 2002;165(10):1395-
23 9.
24
25 .17Sofer S, Weinhouse E, Tal A, Wanderman KL, Margulis G, Leiberman A, et al. Cor pulmonale
26 due to adenoidal or tonsillar hypertrophy or both in children. Noninvasive diagnosis and
27 follow-up. *Chest* 1988;93(1):119-22.
28
29 .18Korayem MM, Witmans M, MacLean J, Heo G, El-Hakim H, Flores-Mir C, et al. Craniofacial
30 morphology in pediatric patients with persistent obstructive sleep apnea with or without
31 positive airway pressure therapy: a cross-sectional cephalometric comparison with controls.
32 *American journal of orthodontics and dentofacial orthopedics : official publication of the*
33 *American Association of Orthodontists, its constituent societies, and the American Board of*
34 *Orthodontics* 2013;144(1):78-85.
35
36 .19Katyal V, Pamula Y, Martin AJ, Daynes CN, Kennedy JD, Sampson WJ. Craniofacial and
37 upper airway morphology in pediatric sleep-disordered breathing: Systematic review and
38 meta-analysis. *American journal of orthodontics and dentofacial orthopedics : official*
39 *publication of the American Association of Orthodontists, its constituent societies, and the*
40 *American Board of Orthodontics* 2013;143(1):20-30 e3.
41
42 .20Flores-Mir C, Korayem M, Heo G, Witmans M, Major MP, Major PW. Craniofacial
43 morphological characteristics in children with obstructive sleep apnea syndrome: a
44 systematic review and meta-analysis. *Journal of the American Dental Association*
45 2013;144(3):269-77.
46
47 .21Carvalho FR, Lentini-Oliveira D, Machado MA, Prado GF, Prado LB, Saconato H. Oral
48 appliances and functional orthopaedic appliances for obstructive sleep apnoea in children.
49 *The Cochrane database of systematic reviews* 2007(2):CD005520.
50
51
52
53
54
55
56
57
58
59
60

- 1
2
3 .22 Villa MP, Bernkopf E, Pagani J, Broia V, Montesano M, Ronchetti R. Randomized controlled
4 study of an oral jaw-positioning appliance for the treatment of obstructive sleep apnea in
5 children with malocclusion. *American journal of respiratory and critical care medicine*
6 2002;165(1):123-7.
7
- 8 .23 Schutz TC, Dominguez GC, Hallinan MP, Cunha TC, Tufik S. Class II correction improves
9 nocturnal breathing in adolescents. *The Angle orthodontist* 2011;81(2):222-8.
10
- 11 .24 Cozza P, Polimeni A, Ballanti F. A modified monobloc for the treatment of obstructive sleep
12 apnoea in paediatric patients. *Eur J Orthod* 2004;26(5):523-30.
13
- 14 .25 Zhang C, He H, Ngan P. Effects of twin block appliance on obstructive sleep apnea in children:
15 a preliminary study. *Sleep & breathing = Schlaf & Atmung* 2013;17(4):1309-14.
16
- 17 .26 Pliska BT, Almeida F. Effectiveness and outcome of oral appliance therapy. *Dental clinics of*
18 *North America* 2012;56(2):433-44.
19
- 20 .27 Hiyama S, Suda N, Ishii-Suzuki M, Tsuiki S, Ogawa M, Suzuki S, et al. Effects of maxillary
21 protraction on craniofacial structures and upper-airway dimension. *The Angle orthodontist*
22 2002;72(1):43-7.
23
- 24 .28 Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway
25 passage. *The Angle orthodontist* 2008;78(2):209-14.
26
- 27 .29 Kilinc AS, Arslan SG, Kama JD, Ozer T, Dari O. Effects on the sagittal pharyngeal dimensions
28 of protraction and rapid palatal expansion in Class III malocclusion subjects. *Eur J Orthod*
29 2008;30(1):61-6.
30
- 31 .30 Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot
32 study. *Eur J Orthod* 2006;28(2):184-9.
33
- 34 .31 Kaygisiz E, Tuncer BB, Yuksel S, Tuncer C, Yildiz C. Effects of maxillary protraction and
35 fixed appliance therapy on the pharyngeal airway. *The Angle orthodontist* 2009;79(4):660-7.
36
- 37 .32 Lee JW, Park KH, Kim SH, Park YG, Kim SJ. Correlation between skeletal changes by
38 maxillary protraction and upper airway dimensions. *The Angle orthodontist* 2011;81(3):426-
39 32.
40
- 41 .33 Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive
42 sleep apnea: a systematic review and meta-analysis. *Sleep medicine reviews* 2010;14(5):287-
43 97.
44
- 45 .34 World Health Organization (2012) WHO Handbook for guideline development. Available:
46 http://www.who.int/hiv/topics/mtct/grc_handbook_mar2010_1.pdf. Accessed 10 Sept 2013.
47
- 48 .35 Rowe G, Wright G. The Delphi technique as a forecasting tool: issues and analysis. *Int J*
49 *Forecasting* 1999;15(4):353-75.
50
51
52
53
54
55
56
57
58
59
60

- 1
2
3 .36Dalkey N, Helmer O. An Experimental Application of the Delphi Method to the use of experts.
4 *Management Science* 1963;9(3):458-67.
5
6 .37Richmond S, Shaw WC, O'Brien KD, Buchanan IB, Jones R, Stephens CD, et al. The
7 development of the PAR Index (Peer Assessment Rating): reliability and validity. *Eur J*
8 *Orthod* 1992;14(2):125-39.
9
10 .38Richmond S, Shaw WC, Roberts CT, Andrews M. The PAR Index (Peer Assessment Rating):
11 methods to determine outcome of orthodontic treatment in terms of improvement and
12 standards. *Eur J Orthod* 1992;14(3):180-7.
13
14 .39Daniels C, Richmond S. The development of the index of complexity, outcome and need
15 (ICON). *J Orthod* 2000;27(2):149-62.
16
17 .40Brook PH, Shaw WC. The development of an index of orthodontic treatment priority. *Eur J*
18 *Orthod* 1989;11(3):309-20.
19
20 .41Mars M, Batra P, Worrell E. Complete unilateral cleft lip and palate: validity of the five-year
21 index and the Goslon yardstick in predicting long-term dental arch relationships. *Cleft*
22 *Palate Craniofac J* 2006;43(5):557-62.
23
24 .42Mars M, Plint DA, Houston WJ, Bergland O, Semb G. The Goslon Yardstick: a new system of
25 assessing dental arch relationships in children with unilateral clefts of the lip and palate.
26 *Cleft Palate J* 1987;24(4):314-22.
27
28 .43Johnson N, Sandy J. An aesthetic index for evaluation of cleft repair. *Eur J Orthod*
29 2003;25(3):243-9.
30
31 .44Mossey PA, Clark JD, Gray D. Preliminary investigation of a modified Huddart/Bodenham
32 scoring system for assessment of maxillary arch constriction in unilateral cleft lip and palate
33 subjects. *Eur J Orthod* 2003;25(3):251-7.
34
35 .45Tothill C, Mossey PA. Assessment of arch constriction in patients with bilateral cleft lip and
36 palate and isolated cleft palate: a pilot study. *Eur J Orthod* 2007;29(2):193-7.
37
38 .46Huddart AG, Bodenham RS. The evaluation of arch form and occlusion in unilateral cleft palate
39 subjects. *Cleft Palate J* 1972;9:194-209.
40
41 .47Summers CJ. The occlusal index: a system for identifying and scoring occlusal disorders. *Am J*
42 *Orthod* 1971;59(6):552-67.
43
44 .48Streiner DL, Norman GR. *Health measurement scales: a practical guide to their development*
45 *and use. Oxford: Oxford Medical Publications, 1995.*
46
47 .49Terwee CB, Bot SDM, de Boer MR, van der Windt DAWM, Knol DL, Dekker J, et al. Quality
48 criteria were proposed for measurement properties of health status questionnaires. *J Clin*
49 *Epidemiol* 2007;60(1):34-42.
50
51
52
53
54
55
56
57
58
59
60

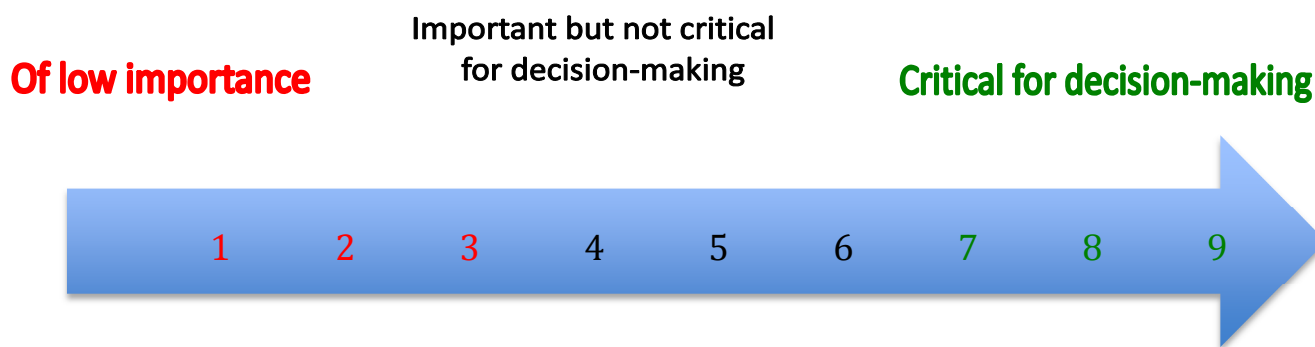
1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

For peer review only

1 Assigned Number _____
23 Factor Being Evaluated: _____
4
5
67
8 Important but not critical
9 Of low importance for decision-making Critical for decision-making
1017
18
19 Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.
20

- 21
22 1- In your opinion, is this factor commonly observed in Pediatric Obstructive Sleep Apnea patients?
23 Yes 1 2 3 4 5 6 7 8 9
24 No
- 25
26 2- In your opinion, does this factor contribute to Pediatric Obstructive Sleep Apnea symptoms?
27 Yes 1 2 3 4 5 6 7 8 9
28 No
- 29
30 3- In your opinion, would correcting this factor help diminish the symptoms of Pediatric Obstructive
31 Sleep Apnea?
32 Yes 1 2 3 4 5 6 7 8 9
33 No
- 34
35 4- In your opinion, does this factor contribute to orthodontic treatment need?
36 Yes 1 2 3 4 5 6 7 8 9
37 No
- 38
39 5- In your opinion, is the 5-point scale appropriate for this factor?
40 Yes 1 2 3 4 5 6 7 8 9
41 No; How would you correct it?
42 _____
- 43
44 6- Are the points on the scale attributed correctly for this factor?
45 Yes 1 2 3 4 5 6 7 8 9
46 No; How would you correct it?
47 _____
- 48
49 7- In your opinion, are the pictures appropriate for this factor?
50 Yes 1 2 3 4 5 6 7 8 9
51 No; How would you correct it?
52 _____
53
54
55
56
57
58
59
60

Assigned Number _____



Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.

1. Do you think that this index is useful in fulfilling the goals of having an easy to use index that helps non-dentally trained professionals assess the orthodontic treatment need to help the symptoms in pediatric patients with OSA?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

2. Do you believe that the organizing committee is developing this index in the appropriate way? (i.e. Reviewing the recommendations in the literature, seeking expert opinions, gaining end-user approval and testing its reliability)

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

3. In your opinion were all the factors, identified in this meeting, important for this index?

Yes 1 2 3 4 5 6 7 8 9

No; Which factors were not important?

4. In your opinion, have all the important areas concerning orthodontic treatment need to help symptoms of OSA been identified?

Yes 1 2 3 4 5 6 7 8 9

No; Which ones were missed?

5. Is the layout of the index easy to navigate?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Please provide any other feedback below that may help improve the index:

For peer review only

BMJ Open

Developing an Index for the Orthodontic Treatment Need in Pediatric Patients with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool between Physicians and Orthodontists

Journal:	<i>BMJ Open</i>
Manuscript ID:	bmjopen-2014-005680.R2
Article Type:	Protocol
Date Submitted by the Author:	14-Aug-2014
Complete List of Authors:	Altalibi, Mostafa; University of Alberta, School of Dentistry, Orthodontic Graduate Program Saltaji, Humam; University of Alberta, School of Dentistry, Orthodontic Graduate Program Roduta Roberts, Mary; University of Alberta, Department of Occupational Therapy, Faculty of Rehabilitation Medicine Major, Michael; University of Alberta, Inter-disciplinary Airway Research Clinic (I-ARC) Macleon, Joanna; University of Alberta, Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital Major, Paul; University of Alberta, School of Dentistry, Orthodontic Graduate Program
Primary Subject Heading:	Dentistry and oral medicine
Secondary Subject Heading:	Paediatrics, Ear, nose and throat/otolaryngology, Health services research
Keywords:	PAEDIATRICS, HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Otolaryngology < SURGERY

SCHOLARONE™
Manuscripts

1
2
3 **Developing an Index for the Orthodontic Treatment Need in Pediatric Patients**
4 **with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool**
5
6 **between Physicians and Orthodontists**
7
8

9
10
11 **Mostafa Altalibi^a; Humam Saltaji^b; Mary Roduta Roberts^c; Michael Major^d; Joanna**
12 **Maclean^e, Paul W Major^f**
13

14
15
16 ^a MSc Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta,
17 Edmonton, Alberta, Canada.

18
19 ^b PhD Candidate and Resident, Orthodontic Graduate Program, School of Dentistry, University of
20 Alberta, Edmonton, Alberta, Canada.

21
22 ^c Assistant Professor Department of Occupational Therapy, Faculty of Rehabilitation Medicine,
23 University of Alberta, Edmonton, Alberta, Canada.

24
25 ^d Director, Inter-disciplinary Airway Research Clinic (I-ARC), Assistant Clinical Professor,
26 Orthodontics, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

27
28 ^e Medical Director at Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital;
29 Assistant Professor, School of Medicine, University of Alberta, Edmonton, Alberta, Canada.

30
31 ^f Professor and Chair of Dentistry; Senior Associate Dean, Faculty of Medicine and Dentistry,
32 University of Alberta, Edmonton, Alberta, Canada.
33
34
35
36

37 **Corresponding Author**

38 Mostafa Altalibi DMD, MSc (Ortho)
39 Orthodontic Graduate Program
40 School of Dentistry
41 5-476, Edmonton Clinic Health Academy (ECHA)
42 University of Alberta
43 11405-87 Ave, Edmonton, Canada T6G 1C9
44 Phone: 780 885-7457
45 E-mail: altalibi@ualberta.ca
46
47
48
49
50

51
52 **Word Count:** 2807

53
54 **Key Words:** Obstructive Sleep Apnea; Pediatric; Index; Orthodontic Treatment Need; Visual
55 Analogue Scale; Development; Validation; Reliability.
56
57
58
59
60

ABSTRACT

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

INTRODUCTION

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.¹ More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.²⁻⁸ Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.^{9 10 11-13} Other physiologic processes that can be affected include stunted growth,¹⁴ high blood pressure,¹⁵ damage to the heart: ventricular hypertrophy,¹⁶ and cor pulmonale¹⁷. In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla¹⁸ or mandible, a long lower face height and restriction in the space of the upper airway^{19 20}. Furthermore, when evaluating the polysomnography of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances²¹⁻²⁶ can be of benefit at reducing the severity of OSA. Reverse pull headgear²⁷⁻³², and maxillary & mandibular advancement surgery³³ have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, ect) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment. Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

METHODS AND ANALYSIS

This research is classified as an applied interdisciplinary medical research. The overall study design involves components of qualitative analysis and quantitative analysis. The qualitative components

comprise focus groups and committee meetings. The quantitative components comprise reliability tests and a cross sectional validity test.

A) Initial Development

In accordance with the World Health Organization's recommendations on developing an index,³⁴ development of the index will be achieved through the following objectives:

- 1- *Establishing a Steering Committee*
- 2- *Scoping the index*
- 3- *Reviewing the literature*
- 4- *Drafting the index*
- 5- *Organizing an Index Development Group*
- 6- *Organizing an External Review Group*

Steering Committee

The Steering Committee will be established apriori, and will be identified by the principal investigator based on the required expertise in the involved fields and based on availability and access. The Steering Committee will be responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index. In these meetings unanimous consensus through discussion must be reached on all decisions before proceeding to the next step.

Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in

1
2
3 question will lead to an improvement in the OSA symptoms. The literature search will not be a
4 systematic review, and will include results from PubMed, Medline, EMBASE, Web of Science,
5 Scopus, the Cochrane Library and grey literature. The search strategy including: inclusion criteria,
6 exclusion criteria and key words, will be devised through the advice of the steering committee. The
7 searches themselves will be done in duplicate, using 2 different assessors. Of the relevant articles,
8 hand searches will be performed on the bibliography lists. The results will be discussed and
9 consensus will be reached between both assessors for the results of the search. Priority will be given
10 to the results of well-conducted and well-reported systematic reviews and randomized control trials.
11 Each of the factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will
12 be searched for the craniofacial morphology of pediatric OSA patients. All of this information will
13 be brought back to the Steering Committee for discussion of inclusion / elimination of factors in the
14 index.
15
16
17
18
19
20
21
22
23

24 **Drafting the Index**

25
26
27 Since this index will have visual-rating scales for each of the craniofacial and occlusal
28 factors, the steering committee will devise an outline suitable for displaying each of the factors
29 representing the index. The factors in the drafted index will also have a number of levels that will
30 divide the factor into categories of severity. The number of levels for a given factor will also be
31 determined by the Steering Committee. Details describing the nature of the illustrations, the amount
32 of levels that each of the factors will have, and the general layout of the index will be agreed upon
33 by the steering committee, and the graphic artist will then design a preliminary version of the index
34 based on the relevant factors and the feedback from the steering committee.
35
36
37
38
39
40

41 **The Index Development Meeting**

42
43
44 The Index Development Group (IDG) is formed by a group of external multidisciplinary
45 experts who provide evidence-based recommendations, on the content, layout and development
46 process of the index. This eclectic group will be chosen by the Steering committee based on
47 experience in the field, expertise and ability to contribute, availability and access. The group should
48 be small enough to be able to have effective discussions, while large enough to ensure the
49 appropriate representation from all the stakeholders. In this study we plan on including a broad
50 spectrum of medical specialists, family physicians, orthodontists, and methodologists. The goals of
51 this meeting are to gain additional information to strengthen the index, as well as gaining the
52 professional credibility of a broad range of experts on the topic of pediatric OSA.
53
54
55
56
57
58
59
60

1
2
3 Once the members have been chosen by the Steering Committee and accept the invitation to
4 participate, the IDG members will be emailed a document 4 days prior to the meeting, explaining:
5 the purpose of the index and meeting, how the meeting will proceed, what to expect in the meeting,
6 and a brief literature review of the orthodontic techniques currently available to help with the
7 symptoms of OSA. Moreover, every member will be randomly assigned a number in order to
8 maintain anonymity of the responses.
9

14 Procedure of the IDG Meeting

16 The meeting will commence with a brief introduction summarizing the literature review, the
17 purpose of the index and its relevance. The meeting will then proceed to collect feedback (
18 DIX 1) on the aforementioned chosen factors using a modified Delphi technique^{35 36}, which
19 is a communication technique that structures the meeting and minimizes bias in responses.
20 Participants' responses will be collected through a web-based response portal. Each factor will be
21 explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based on
22 its importance for decision-making. Once all the feedback is received on a particular factor, the
23 summary of the results will be displayed for everyone to see. Discussion will ensue for a maximum
24 of 5 minutes, with each person talking no more than 1 minute. Then everyone will be asked to re-
25 enter their feedback on the website for the same factor in light of the discussion. This cycle will
26 continue until a consensus of greater than 80% of the members is reached. Once consensus is
27 reached, then we will move on to the next factor and the participants will be asked to give feedback
28 on this new factor in a similar manner.
29
30
31
32
33
34
35
36
37
38

39 In addition, two negative control factors will be used to calibrate the responses. The first
40 negative control will be of orthodontic relevance but have no effect on the amelioration of the
41 symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of
42 relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck
43 circumference).
44
45
46
47
48

49 Once all the factors have been discussed and consensus reached, we will get feedback
50 through the website on the: alignment - importance of the index, and if the development process is
51 appropriate; relevance – content analysis, and whether all the factors identified are important;
52 representation – if there is anything that needs to be added to the index (APPENDIX 2). Finally,
53 additional written feedback will be accepted at the very end of the session.
54
55
56
57
58
59
60

1
2
3 All the feedback from the IDM will be summarized and presented to the Steering
4 Committee, and decisions will be made to remove factors, modify factors, and /or modify the
5 outline and layout of the index. These modifications will subsequently be presented to the graphic
6 artist and a second draft will be procured.
7
8
9

10 **The External Review Group Meeting**

11
12 The External Review Group (ERG) is composed of end-users and interested parties. The
13 group will also be identified based on the advice of the steering committee based on the ability of
14 the group to assess the index and contribute to its development, availability and access. This group
15 is not responsible for any content analysis, instead it will be responsible for reviewing the layout,
16 simplicity and ease of use. It will also be responsible to assess usefulness of the index in the
17 healthcare setting and give feedback on the feasibility of implementing the index in practice. The
18 goal of this meeting is to gain the end-user approval for the ease and feasibility of administering the
19 index. This group should be large enough to be a representative sample of the population, yet small
20 enough to allow for ease of explanation and healthy discussion. It is not as structured as the IDM,
21 and allows for the participants to freely express their opinions in an open forum.
22
23
24
25
26
27
28
29
30

31 In this meeting we will explain the theory behind the index by briefly reviewing the
32 literature and then explain the purpose of the index. Subsequently we will show the group a pilot
33 version of the index and a paper will be distributed to receive for their feedback based on the
34 following questions:
35
36
37

- 38 1- Do you understand the purpose of the index?
- 39 2- Do you understand what each factor is assessing?
 - 40 a. If not which one(s) do you not understand and why?
- 41 3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
- 42 4- Would you use this index in your practice?
- 43 5- Other recommendations:
 - 44
 - 45
 - 46
 - 47
 - 48

49 By compiling all the comments and scores onto a table, the results will display all the
50 feedback from the ERG meeting. All these results will be presented to the Steering Committee, and
51 decisions will be made to modify the outline and layout of the index. These modifications will
52 subsequently be presented to the graphic artist and a third draft will be procured.
53
54
55
56

57 **B) Reliability**

1
2
3 Reliability of the index will be tested within a group that represents the typical end-user
4 population. This includes physicians of family medicine, pediatricians, pediatric ENT, or pediatric
5 sleep physicians. Reliability will also be assessed within a group of orthodontists, who by their
6 training are experts at assessing malocclusion and craniofacial morphology. Therefore 10-20
7
8
9
10 physicians and orthodontists will be recruited as examiners in this study.

11
12 A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-
13 disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and their
14 intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief
15 explanation about the use and application of the index, the physicians as well as the orthodontists
16 will apply the developed index to the sample patient pool's pictures once. In order to diminish
17 recall bias, application of the index will be repeated 2 weeks later.

18
19 Intra-rater reliability and inter-rater reliability between the physicians, between the
20 orthodontists and between the physicians and orthodontists will be compared. The reliability will be
21 assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.

22 23 24 25 26 27 28 29 **C) Validation**

30
31 The Index will be validated using dichotomous responses from orthodontists on whether
32 specific patients would require orthodontic treatment to help their obstructive sleep apnea
33 symptoms and comparing it to the score that the index gave those same patients. This will be
34 achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric
35 OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral
36 pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic diagnosed
37 with OSA, and the orthodontists will be asked to rate these patients, using a “yes” or “no” response,
38 whether they would benefit from orthodontic treatment for their OSA symptoms. The index will
39 then be applied on the same patients by the principal investigator and the score of each patient will
40 be recorded. Using a stepwise multiple-logistic regression each of the identified factors will be
41 given a weight; this will represent the relative importance of the factor. Once analyzed if the
42 correlation is high between the expert scores and the cluster groups, then the clusters are
43 meaningful and valid. Furthermore, a cutoff for most efficient score above which to refer will be
44 chosen using a graph and observing the value that optimizes the sensitivity, specificity and overall
45 accuracy of the index. Finally, four grades of treatment-need will be determined using the twenty-
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60 five percentile ranges. The grades will be:

- 1- Minimal Need
- 2- Mild Need
- 3- Moderate Need
- 4- Severe Need

DISCUSSION

The development plan of this index has been conceived through a modification of the WHO Handbook for guideline development,³⁴ as well as reviewing the orthodontic literature for ways indices have been previously developed. The WHO provided an excellent starting point, from there modifications were made to cater to the specifics of this study, given that there are differences between developing a guideline and an index for orthodontic treatment need. The literature was useful, and among the index development protocols reviewed, certain assessed the orthodontic treatment need within the entire population,³⁷⁻⁴⁰ while others assessed it for a given subpopulation⁴¹⁻⁴⁶; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a protocol for our particular needs from the available literature and using the experience and expertise of the authors. Through this protocol we aim to develop an index that fulfills all of the following criteria:⁴⁷

1. Gradient of Numeric Values: The severity of the orthodontic treatment need within the pediatric OSA patients should be defined within a numerical scheme that demonstrates a finite and progressive gradient from low need to high need.
2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
3. Clinical Importance: The numerical scale should correspond with the clinically appraised orthodontic treatment need of pediatric OSA patients.
4. Statistical Ease: should be amendable to statistical analysis.
5. Reliability: Should have a high intra- and inter-rater reliability.
6. Practical: The instruments required to score the index should be practical to the setting in which it will be administered.
7. Minimal Judgment: Applying the index should require minimal judgment.
8. Simple: The index shouldn't have a high financial or time cost, therefore should be simple enough to administer to many patients.
9. Detect Change: The index should be able to detect changes in orthodontic treatment need in pediatric OSA patients.

1
2
3 10. Validity: should be valid over time.
4

5 Validity can be characterized into different types: Face Validity, Content Validity, Construct
6 Validity, and Criterion Validity.⁴⁸ In this study, we will examine these kinds of validity at different
7 stages of development. The first draft of the index will focus on establishing face validity.
8 Feedback from the steering committee and IDG will assist in establishing content and construct
9 validity. Assuming that the “gold standard” in assessing the orthodontic treatment-need in pediatric
10 patients with OSA is an orthodontist with experience in dealing with pediatric OSA patients, then
11 the subsequent modification of the index based on the reliability tests and the dichotomous
12 responses from the orthodontists provide the index with the necessary criterion-related validity
13 evidence through statistical means.⁴⁹
14
15
16
17
18
19
20

21 **Significance**

22
23
24 This index will help physicians and other medical professionals identify which craniofacial
25 phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA
26 management. Furthermore, due to the diverse medical effects of sleep deprivation, there will be a
27 trend to make sleep apnea into a centralized service, where the main focus is for a highly trained
28 multidisciplinary team to treat a high volume of patients to a standardized protocol, where
29 meticulous documentation is exercised. This index is part of that documentation process. It will
30 allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will
31 also allow long-term follow up and audit in order to enter into comparisons with other centers.
32
33
34
35
36
37

38 **Dissemination Plan**

39
40 The dissemination of this index will be done through a variety of ways in order to maximize
41 its reach. Primarily, it will be published in a peer-reviewed journal, which will allow its
42 introduction to the scientific literature. The journal should be a respected medical journal with
43 broad reach, in order to allow the greatest number of physicians to be exposed to the index.
44 Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian
45 and American population of medical professionals. Moreover, the index will be presented at
46 national and international conferences to increase the awareness of the index among the scientific
47 community. Finally, the index will be used at the University of Alberta’s Interdisciplinary Airway
48 Research Center, and more research, so that future research in this center will incorporate it. It will
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3 also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website
4 under the physician section, to further educate the doctors who visit the site on the index.
5
6
7
8
9

10 11 **Funding**

12
13
14 No funding has been provided for this project.
15

16 **Authors' Contributions**

17
18 MA HS MRR MM JM PM conceived and designed the study; MA drafted the manuscript and
19 integrated critical feedback from all of the other authors. All of the authors approved the final
20 version of the manuscript.
21
22
23

24 **Ethics approval**

25
26 The proposed research has received ethical approval numbered Pro00045067 from the University of
27 Alberta Ethics Board.
28

29 **Competing Interests**

30
31 The authors declare no conflicts of interests.
32
33
34
35

36 **REFERENCES**

- 37
38
39 .1 O'Brien LM, Mervis CB, Holbrook CR, et al. Neurobehavioral implications of habitual snoring
40 in children. *Pediatrics* 2004;114(1):44-9.
41
42 .2 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the*
43 *American Thoracic Society* 2008;5(2):242-52.
44
45 .3 Gislason T, Benediksdottir B. Snoring, apneic episodes, and nocturnal hypoxemia among
46 children 6 months to 6 years old. An epidemiologic study of lower limit of prevalence.
47 *Chest* 1995;107(4):963.6-
48
49 .4 Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds.
50 *Archives of disease in childhood* 1993;68(3):360-6.
51
52 .5 Redline S, Tishler PV, Schluchter M, et al. Risk factors for sleep-disordered breathing in
53 children. Associations with obesity, race, and respiratory problems. *American journal of*
54 *respiratory and critical care medicine* 1999;159(5 Pt 1):1527-32.
55
56
57
58
59
60

- 1
2
3 .6Bixler EO, Vgontzas AN, Lin HM, et al. Sleep disordered breathing in children in a general
4 population sample: prevalence and risk factors. *Sleep* 2009;32(6):731-6.
5
6 .7Li AM, So HK, Au CT, et al. Epidemiology of obstructive sleep apnoea syndrome in Chinese
7 children: a two-phase community study. *Thorax* 2010;65(11):991-7.
8
9 .8O'Brien LM, Holbrook CR, Mervis CB, et al. Sleep and neurobehavioral characteristics of 5- to
10 7-year-old children with parentally reported symptoms of attention-deficit/hyperactivity
11 disorder. *Pediatrics* 2003;111(3):554-63.
12
13
14 .9Huynh NT, Morton PD, Rompre PH, et al. Associations between sleep-disordered breathing
15 symptoms and facial and dental morphometry, assessed with screening examinations.
16 *American journal of orthodontics and dentofacial orthopedics : official publication of the*
17 *American Association of Orthodontists, its constituent societies, and the American Board of*
18 *Orthodontics* 2011;140(6):762-70.
19
20
21 .10Beebe DW. Neural and neurobehavioral dysfunction in children with obstructive sleep apnea.
22 *PLoS medicine* 2006;3(8):e323.
23
24 .11Chervin RD, Archbold KH, Dillon JE, et al. Inattention, hyperactivity, and symptoms of sleep-
25 disordered breathing. *Pediatrics* 2002;109(3):449-56.
26
27
28 .12Crabtree VM, Varni JW, Gozal D. Health-related quality of life and depressive symptoms in
29 children with suspected sleep-disordered breathing. *Sleep* 2004;27(6):1131-8.
30
31 .13Halbower AC, Degaonkar M, Barker PB, et al. Childhood obstructive sleep apnea associates
32 with neuropsychological deficits and neuronal brain injury. *PLoS medicine* 2006;3(8):e301.
33
34 .14Bar A, Tarasiuk A, Segev Y, et al. The effect of adenotonsillectomy on serum insulin-like
35 growth factor-I and growth in children with obstructive sleep apnea syndrome. *The Journal*
36 *of pediatrics* 1999;135(1):76-80.
37
38
39 .15Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea.
40 *American journal of respiratory and critical care medicine* 1998;157(4 Pt 1):103-109.
41
42 .16Amin RS, Kimball TR, Bean JA, et al. Left ventricular hypertrophy and abnormal ventricular
43 geometry in children and adolescents with obstructive sleep apnea. *American journal of*
44 *respiratory and critical care medicine* 2002;165(10):1395-9.
45
46
47 .17Sofer S, Weinhouse E, Tal A, et al. Cor pulmonale due to adenoidal or tonsillar hypertrophy or
48 both in children. Noninvasive diagnosis and follow-up. *Chest* 1988;93(1):119-22.
49
50 .18Korayem MM, Witmans M, MacLean J, et al. Craniofacial morphology in pediatric patients
51 with persistent obstructive sleep apnea with or without positive airway pressure therapy: a
52 cross-sectional cephalometric comparison with controls. *American journal of orthodontics*
53 *and dentofacial orthopedics : official publication of the American Association of*
54 *Orthodontists, its constituent societies, and the American Board of Orthodontics*
55 *2013;144(1):78-85.*
56
57
58
59
60

- 1
2
3 .19Katyal V, Pamula Y, Martin AJ, et al. Craniofacial and upper airway morphology in pediatric
4 sleep-disordered breathing: Systematic review and meta-analysis. *American journal of*
5 *orthodontics and dentofacial orthopedics : official publication of the American Association*
6 *of Orthodontists, its constituent societies, and the American Board of Orthodontics*
7 2013;143(1):20-30 e3.
8
9
10 .20Flores-Mir C, Korayem M, Heo G, et al. Craniofacial morphological characteristics in children
11 with obstructive sleep apnea syndrome: a systematic review and meta-analysis. *Journal of*
12 *the American Dental Association* 2013;144(3):269-77.
13
14 .21Carvalho FR, Lentini-Oliveira D, Machado MA, et al. Oral appliances and functional
15 orthopaedic appliances for obstructive sleep apnoea in children. *The Cochrane database of*
16 *systematic reviews* 2007(2):CD005520.
17
18
19 .22Villa MP, Bernkopf E, Pagani J, Broia V, et al. Randomized controlled study of an oral jaw-
20 positioning appliance for the treatment of obstructive sleep apnea in children with
21 malocclusion. *American journal of respiratory and critical care medicine* 2002;165(1):123-
22 7.
23
24 .23Schutz TC, Dominguez GC, Hallinan MP, et al. Class II correction improves nocturnal
25 breathing in adolescents. *The Angle orthodontist* 2011;81(2):222-8.
26
27
28 .24Cozza P, Polimeni A, Ballanti F. A modified monobloc for the treatment of obstructive sleep
29 apnoea in paediatric patients. *Eur J Orthod* 2004;26(5):523-30.
30
31 .25Zhang C, He H, Ngan P. Effects of twin block appliance on obstructive sleep apnea in children:
32 a preliminary study. *Sleep & breathing = Schlaf & Atmung* 2013;17(4):1309-14.
33
34 .26Pliska BT, Almeida F. Effectiveness and outcome of oral appliance therapy. *Dental clinics of*
35 *North America* 2012;56(2):433-44.
36
37
38 .27Hiyama S, Suda N, Ishii-Suzuki M, et al. Effects of maxillary protraction on craniofacial
39 structures and upper-airway dimension. *The Angle orthodontist* 2002;72(1):43-7.
40
41 .28Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway
42 passage. *The Angle orthodontist* 2008;78(2):209-14.
43
44 .29Kilinc AS, Arslan SG, Kama JD, et al. Effects on the sagittal pharyngeal dimensions of
45 protraction and rapid palatal expansion in Class III malocclusion subjects. *Eur J Orthod*
46 2008;30(1):61-6.
47
48
49 .30Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot
50 study. *Eur J Orthod* 2006;28(2):184-9.
51
52
53 .31Kaygisiz E, Tuncer BB, Yuksel S, Tuncer C, Yildiz C. Effects of maxillary protraction and
54 fixed appliance therapy on the pharyngeal airway. *The Angle orthodontist* 2009;79(4):660-7.
55
56 .32Lee JW, Park KH, Kim SH, et al. Correlation between skeletal changes by maxillary protraction
57 and upper airway dimensions. *The Angle orthodontist* 2011;81(3):426-32.
58
59
60

- 1
2
3 .33Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive
4 sleep apnea: a systematic review and meta-analysis. *Sleep medicine reviews* 2010;14(5):287-
5 97.
6
7 .34World Health Organization (2012) WHO Handbook for guideline development. Available:
8 http://www.who.int/hiv/topics/mtct/grc_handbook_mar2010_1.pdf. Accessed 10 Sept 2013.
9
10 .35Rowe G, Wright G. The Delphi technique as a forecasting tool: issues and analysis. *Int J*
11 *Forecasting* 1999;15(4):353-75.
12
13 .36Dalkey N, Helmer O. An Experimental Application of the Delphi Method to the use of experts.
14 *Management Science* 1963;9(3):458-67.
15
16
17 .37Richmond S, Shaw WC, O'Brien KD, Buchanan IB, Jones R, Stephens CD, et al. The
18 development of the PAR Index (Peer Assessment Rating): reliability and validity. *Eur J*
19 *Orthod* 1992;14(2):125-39.
20
21 .38Richmond S, Shaw WC, Roberts CT, et al. The PAR Index (Peer Assessment Rating): methods
22 to determine outcome of orthodontic treatment in terms of improvement and standards. *Eur*
23 *J Orthod* 1992;14(3):180-7.
24
25 .39Daniels C, Richmond S. The development of the index of complexity, outcome and need
26 (ICON). *J Orthod* 2000;27(2):149-62.
27
28 .40Brook PH, Shaw WC. The development of an index of orthodontic treatment priority. *Eur J*
29 *Orthod* 1989;11(3):309-20.
30
31 .41Mars M, Batra P, Worrell E. Complete unilateral cleft lip and palate: validity of the five-year
32 index and the Goslon yardstick in predicting long-term dental arch relationships. *Cleft*
33 *Palate Craniofac J* 2006;43(5):557-62.
34
35 .42Mars M, Plint DA, Houston WJ, et al. The Goslon Yardstick: a new system of assessing dental
36 arch relationships in children with unilateral clefts of the lip and palate. *Cleft Palate J*
37 1987;24(4):314-22.
38
39 .43Johnson N, Sandy J. An aesthetic index for evaluation of cleft repair. *Eur J Orthod*
40 2003;25(3):243-9.
41
42 .44Mossey PA, Clark JD, Gray D. Preliminary investigation of a modified Huddart/Bodenham
43 scoring system for assessment of maxillary arch constriction in unilateral cleft lip and palate
44 subjects. *Eur J Orthod* 2003;25(3):251-7.
45
46 .45Tohill C, Mossey PA. Assessment of arch constriction in patients with bilateral cleft lip and
47 palate and isolated cleft palate: a pilot study. *Eur J Orthod* 2007;29(2):193-7.
48
49 .46Huddart AG, Bodenham RS. The evaluation of arch form and occlusion in unilateral cleft palate
50 subjects. *Cleft Palate J* 1972;9:194-209.
51
52
53
54
55
56
57
58
59
60

- 1
2
3 .47Summers CJ. The occlusal index: a system for identifying and scoring occlusal disorders. *Am J*
4 *Orthod* 1971;59(6):552-67.
5
6 .48Streiner DL, Norman GR. *Health measurement scales: a practical guide to their development*
7 *and use*. Oxford: Oxford Medical Publications, 1995.
8
9 .49Terwee CB, Bot SDM, de Boer MR, et al. Quality criteria were proposed for measurement
10 properties of health status questionnaires. *J Clin Epidemiol* 2007;60(1):34-42.
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

For peer review only

1
2
3 **Developing an Index for the Orthodontic Treatment Need in Pediatric Patients**
4 **with Obstructive Sleep Apnea: A Protocol for a Novel Communication Tool**
5
6 **between Physicians and Orthodontists**
7
8

9
10
11 **Mostafa Altalibi^a; Humam Saltaji^b; Mary Roduta Roberts^c; Michael Major^d; Joanna**
12 **Maclean^e, Paul W Major^f**
13

14
15
16 ^a MSc Resident, Orthodontic Graduate Program, School of Dentistry, University of Alberta,
17 Edmonton, Alberta, Canada.

18
19 ^b PhD Candidate and Resident, Orthodontic Graduate Program, School of Dentistry, University of
20 Alberta, Edmonton, Alberta, Canada.

21
22 ^c Assistant Professor **Department of** Occupational Therapy, Faculty of Rehabilitation Medicine,
23 University of Alberta, Edmonton, Alberta, Canada.

24
25 ^d Director, Inter-disciplinary Airway Research Clinic (I-ARC), Assistant Clinical Professor,
26 Orthodontics, School of Dentistry, University of Alberta, Edmonton, Alberta, Canada.

27
28 ^e Medical Director at Northern Alberta Pediatric Sleep Laboratory, Stollery Children's Hospital;
29 Assistant Professor, School of Medicine, University of Alberta, Edmonton, Alberta, Canada.

30
31 ^f Professor and Chair of Dentistry; Senior Associate Dean, Faculty of Medicine and Dentistry,
32 University of Alberta, Edmonton, Alberta, Canada.
33
34
35
36

37 **Corresponding Author**

38 Mostafa Altalibi DMD, MSc (Ortho)
39 Orthodontic Graduate Program
40 School of Dentistry
41 5-476, Edmonton Clinic Health Academy (ECHA)
42 University of Alberta
43 11405-87 Ave, Edmonton, Canada T6G 1C9
44 Phone: 780 885-7457
45 E-mail: altalibi@ualberta.ca
46
47
48
49
50

51
52 **Word Count:** 2807

53
54 **Key Words:** Obstructive Sleep Apnea; Pediatric; Index; Orthodontic Treatment Need; Visual
55 Analogue Scale; Development; Validation; Reliability.
56
57
58
59
60

ABSTRACT

Introduction: Sleep disordered breathing in the pediatric population can manifest as an array of different systemic symptoms; among them is a distinct malocclusion and craniofacial phenotype. Emerging research suggests that the treatment of this malocclusion and/or craniofacial phenotype through orthodontic intervention may help with the symptoms of these patients. Selecting the patients that would benefit from orthodontic treatment can be a difficult task for the physician with minimal dental training. Therefore the aim of this study is to develop a simple index to be used by medical professionals, to identify those pediatric patients with orthodontic treatment needs that may benefit their obstructive sleep apnea (OSA) symptoms.

Methods and analysis: The methodology in this project has been devised through the World Health Organization's (WHO) recommendations on developing an index, with modifications based on the specific needs of this study. Based on the available literature, a draft index will be produced, and subjected to multiple iterative revisions based on the feedback from: the Index Development Group, a group of multidisciplinary and internationally acclaimed experts in the field; the External Review Group, a group of potential end-users and interested parties; and the Steering Committee. Once the index has been formalized, it will be subjected to a pair of reliability tests using physicians and orthodontists scored 2-weeks apart. Subsequently the index will be validated using dichotomous responses from orthodontists on whether they would treat a patient for OSA symptoms, and comparing the responses to the score of the index on the same patient.

Ethics and dissemination: The index will be translated into French, and will be presented in orthodontic and medical conferences, workshops, seminars, round table discussions and free copies for download will be made available on the website of the University of Alberta Interdisciplinary Airway Research Clinic (iarc.ualberta.ca). Furthermore, the index will be published in a peer-reviewed medical journal to further increase the exposure of the index.

Article Summary

Article focus

- To develop a simple index for medical professionals to identify children and adolescents with obstructive sleep apnea who may experience functional airway benefit from orthodontic treatment.

Strength and limitations of this study

- This index will help physicians and other medical professionals understand and identify which obstructive sleep apnea patients with malocclusions and craniofacial phenotypes are likely to benefit from targeted orthodontic treatment, and will allow them to refer these patients accordingly.
- This index development acknowledges the multi-factorial nature of SDB and the need for multi-disciplinary care. The ideal end result of this index is to facilitate and enhance effective collaboration between invested dental and medical specialties.
- Development of a validated index will facilitate future epidemiology studies, allow for quality assurance, and guide funding allocation. It will also allow long-term follow up and audit in order to enter into comparisons with other centers.

INTRODUCTION

Snoring, although ubiquitous in the adult population, is considered abnormal in children and adolescents.¹ More importantly, it may serve as an indicator of a more severe respiratory problem that presents as a continuum, from primary snoring to obstructive sleep apnea (OSA). Reports vary on the prevalence of obstructive sleep apnea ranging from 0.7% to 5% of the population under 18 years old.²⁻⁸ Moreover, breathing induced sleep disorders have been proven to have a profound effect on the child's behavior, growth and development; the myriad of symptoms include: morning tension-type headaches, excessive morning thirst, excessive fatigue and sleepiness, abnormal shyness, withdrawn and depressive presentation, pattern of attention-deficit/hyperactivity disorder (ADHD), memory impairments, aggressiveness, irritability, among many others.^{9 10 11-13} Other physiologic processes that can be affected include stunted growth,¹⁴ high blood pressure,¹⁵ damage to the heart: ventricular hypertrophy,¹⁶ and cor pulmonale¹⁷. In addition to behavioral and systemic health consequences, craniofacial development is also affected. These patients generally have a craniofacial component contributing to their OSA, which would manifest as a retrognathic maxilla¹⁸ or mandible, a long lower face height and restriction in the space of the upper airway^{19 20}. Furthermore, when evaluating the polysomnography of these patients, the evidence suggests that palatal expansion, and mandibular advancement appliances²¹⁻²⁶ can be of benefit at reducing the severity of OSA. Reverse pull headgear²⁷⁻³², and maxillary & mandibular advancement surgery³³ have also been shown to have great promise at helping this group of patients. Since orthodontic treatment of the OSA craniofacial phenotype is an integral component to multidisciplinary care, it is essential for medical professionals (physicians, nurses, ect) to recognize the phenotype that would benefit from orthodontic treatment. Unfortunately, there are no guidelines for non-dental trained practitioners to help identify which children with SDB would benefit from orthodontic treatment. Therefore this study aims to develop an index that can summarize the need for orthodontic treatment, in select cases of children with OSA, to physicians, and adjunct medical professionals. Once the index is developed, it will be assessed for reliability and will be validated. Upon completion, this index will equip medical professionals with a simple way to assess which patients have a malocclusion that contribute to their OSA and may benefit from orthodontic treatment.

METHODS AND ANALYSIS

This research is classified as an applied interdisciplinary medical research. The overall study design involves components of qualitative analysis and quantitative analysis. The qualitative components

comprise focus groups and committee meetings. The quantitative components comprise reliability tests and a cross sectional validity test.

A) Initial Development

In accordance with the World Health Organization's recommendations on developing an index,³⁴ development of the index will be achieved through the following objectives:

- 1- *Establishing a Steering Committee*
- 2- *Scoping the index*
- 3- *Reviewing the literature*
- 4- *Drafting the index*
- 5- *Organizing an Index Development Group*
- 6- *Organizing an External Review Group*

Steering Committee

The Steering Committee will be established apriori, and will be identified by the principal investigator based on the required expertise in the involved fields and based on availability and access. The Steering Committee will be responsible for overseeing every aspect of the study. It will be composed of a representative group of 3 experts in orthodontics, pediatric sleep medicine, and a methodologist specializing in psychometric property analysis. Their responsibilities include: scoping the index, overseeing evidence retrieval, drafting the index, selecting members of the index development group and external review group, finalizing the index. In these meetings unanimous consensus through discussion must be reached on all decisions before proceeding to the next step.

Scoping the Index

Scoping is the process of defining what factors will be investigated in the literature for inclusion in the index. Scoping will be achieved through the combined experience and expertise of the Steering Committee, and each factor that is suggested will be further investigated in the literature to establish an evidence-based approach to the development of the index.

Reviewing the Literature

For each of the scoped factors the literature will be reviewed to establish relevance. Specifically, the evidence must demonstrate that appropriate treatment of the craniofacial factor in

1
2
3 question will lead to an improvement in the OSA symptoms. The literature search will not be a
4 systematic review, and will include results from PubMed, Medline, EMBASE, Web of Science,
5 Scopus, the Cochrane Library and grey literature. The search strategy including: inclusion criteria,
6 exclusion criteria and key words, will be devised through the advice of the steering committee. The
7 searches themselves will be done in duplicate, using 2 different assessors. Of the relevant articles,
8 hand searches will be performed on the bibliography lists. The results will be discussed and
9 consensus will be reached between both assessors for the results of the search. Priority will be given
10 to the results of well-conducted and well-reported systematic reviews and randomized control trials.
11 Each of the factors will be assessed on its effect on pediatric OSA. Furthermore, the literature will
12 be searched for the craniofacial morphology of pediatric OSA patients. All of this information will
13 be brought back to the Steering Committee for discussion of inclusion / elimination of factors in the
14 index.
15
16

17 **Drafting the Index**

18
19 Since this index will have visual-rating scales for each of the craniofacial and occlusal
20 factors, the steering committee will devise an outline suitable for displaying each of the factors
21 representing the index. The factors in the drafted index will also have a number of levels that will
22 divide the factor into categories of severity. The number of levels for a given factor will also be
23 determined by the Steering Committee. Details describing the nature of the illustrations, the amount
24 of levels that each of the factors will have, and the general layout of the index will be agreed upon
25 by the steering committee, and the graphic artist will then design a preliminary version of the index
26 based on the relevant factors and the feedback from the steering committee.
27
28

29 **The Index Development Meeting**

30
31 The Index Development Group (IDG) is formed by a group of external multidisciplinary
32 experts who provide evidence-based recommendations, on the content, layout and development
33 process of the index. This eclectic group will be chosen by the Steering committee based on
34 experience in the field, expertise and ability to contribute, availability and access. The group should
35 be small enough to be able to have effective discussions, while large enough to ensure the
36 appropriate representation from all the stakeholders. In this study we plan on including a broad
37 spectrum of medical specialists, family physicians, orthodontists, and methodologists. The goals of
38 this meeting are to gain additional information to strengthen the index, as well as gaining the
39 professional credibility of a broad range of experts on the topic of pediatric OSA.
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3 Once the members have been chosen by the Steering Committee and accept the invitation to
4 participate, the IDG members will be emailed a document 4 days prior to the meeting, explaining:
5 the purpose of the index and meeting, how the meeting will proceed, what to expect in the meeting,
6 and a brief literature review of the orthodontic techniques currently available to help with the
7 symptoms of OSA. Moreover, every member will be randomly assigned a number in order to
8 maintain anonymity of the responses.
9

14 **Procedure of the IDG Meeting**

16 The meeting will commence with a brief introduction summarizing the literature review, the
17 purpose of the index and its relevance. The meeting will then proceed to collect feedback
18 (APPENDIX 1) on the aforementioned chosen factors using a modified Delphi technique^{35 36},
19 which is a communication technique that structures the meeting and minimizes bias in responses.
20 Participants' responses will be collected through a web-based response portal. Each factor will be
21 explored through yes or no questions, and yes responses followed-up with a scale of 1-9 based on
22 its importance for decision-making. Once all the feedback is received on a particular factor, the
23 summary of the results will be displayed for everyone to see. Discussion will ensue for a maximum
24 of 5 minutes, with each person talking no more than 1 minute. Then everyone will be asked to re-
25 enter their feedback on the website for the same factor in light of the discussion. This cycle will
26 continue until a consensus of greater than 80% of the members is reached. Once consensus is
27 reached, then we will move on to the next factor and the participants will be asked to give feedback
28 on this new factor in a similar manner.
29
30
31
32
33
34
35
36
37
38

39 In addition, two negative control factors will be used to calibrate the responses. The first
40 negative control will be of orthodontic relevance but have no effect on the amelioration of the
41 symptoms of OSA (ie. crowding, or impacted canine). The second negative control will be of
42 relevance to OSA symptoms, but cannot be changed by orthodontic treatment (ie. BMI or neck
43 circumference).
44
45
46
47
48

49 Once all the factors have been discussed and consensus reached, we will get feedback
50 through the website on the: alignment - importance of the index, and if the development process is
51 appropriate; relevance – content analysis, and whether all the factors identified are important;
52 representation – if there is anything that needs to be added to the index (APPENDIX 2). Finally,
53 additional written feedback will be accepted at the very end of the session.
54
55
56
57
58
59
60

1
2
3 All the feedback from the IDM will be summarized and presented to the Steering
4 Committee, and decisions will be made to remove factors, modify factors, and /or modify the
5 outline and layout of the index. These modifications will subsequently be presented to the graphic
6 artist and a second draft will be procured.
7
8
9

10 **The External Review Group Meeting**

11
12 The External Review Group (ERG) is composed of end-users and interested parties. **The**
13 **group will also be identified based on the advice of the steering committee based on the ability of**
14 **the group to assess the index and contribute to its development, availability and access.** This group
15 is not responsible for any content analysis, instead it will be responsible for reviewing the layout,
16 simplicity and ease of use. It will also be responsible to assess usefulness of the index in the
17 healthcare setting and give feedback on the feasibility of implementing the index in practice. The
18 goal of this meeting is to gain the end-user approval for the ease and feasibility of administering the
19 index. This group should be large enough to be a representative sample of the population, yet small
20 enough to allow for ease of explanation and healthy discussion. It is not as structured as the IDM,
21 and allows for the participants to freely express their opinions in an open forum.
22
23
24
25
26
27
28
29
30

31 In this meeting we will explain the theory behind the index by briefly reviewing the
32 literature and then explain the purpose of the index. Subsequently we will show the group a pilot
33 version of the index and a paper will be distributed to receive for their feedback based on the
34 following questions:
35
36
37

- 38 1- Do you understand the purpose of the index?
- 39 2- Do you understand what each factor is assessing?
 - 40 a. If not which one(s) do you not understand and why?
- 41 3- On a scale of 1 to 10, how simple would you rate this index to understand and use?
- 42 4- Would you use this index in your practice?
- 43 5- Other recommendations:
 - 44
 - 45
 - 46
 - 47
 - 48

49 **By compiling all the comments and scores onto a table, the results will display all the**
50 **feedback from the ERG meeting.** All these results will be presented to the Steering Committee, and
51 decisions will be made to modify the outline and layout of the index. These modifications will
52 subsequently be presented to the graphic artist and a third draft will be procured.
53
54
55
56

57 **B) Reliability**

1
2
3 Reliability of the index will be tested within a group that represents the typical end-user
4 population. This includes physicians of family medicine, pediatricians, pediatric ENT, or pediatric
5 sleep physicians. Reliability will also be assessed within a group of orthodontists, who by their
6 training are experts at assessing malocclusion and craniofacial morphology. Therefore 10-20
7
8
9
10 physicians and orthodontists will be recruited as examiners in this study.

11
12 A pool of 15-40 randomly selected patient charts from the University of Alberta Inter-
13 disciplinary Airway Research Clinic (I-ARC) will be recruited as reliability test subjects, and their
14 intra-oral and extra-oral photographs will be used in the reliability assessment. After a brief
15 explanation about the use and application of the index, the physicians as well as the orthodontists
16 will apply the developed index to the sample patient pool's pictures once. In order to diminish
17 recall bias, application of the index will be repeated 2 weeks later.

18
19 Intra-rater reliability and inter-rater reliability between the physicians, between the
20 orthodontists and between the physicians and orthodontists will be compared. The reliability will be
21 assessed using Interclass Correlation coefficients (ICC) and Bland –Altman Plots.

22 23 24 25 26 27 28 29 **C) Validation**

30
31 The Index will be validated using dichotomous responses from orthodontists on whether
32 specific patients would require orthodontic treatment to help their obstructive sleep apnea
33 symptoms and comparing it to the score that the index gave those same patients. This will be
34 achieved by setting up a website where 30 orthodontists with experience in dealing with pediatric
35 OSA will be recruited to take the assessment. The website will contain extra-oral and intra-oral
36 pictures of patients randomly selected from the Interdisciplinary Airway Research Clinic diagnosed
37 with OSA, and the orthodontists will be asked to rate these patients, using a “yes” or “no” response,
38 whether they would benefit from orthodontic treatment for their OSA symptoms. The index will
39 then be applied on the same patients by the principal investigator and the score of each patient will
40 be recorded. Using a stepwise multiple-logistic regression each of the identified factors will be
41 given a weight; this will represent the relative importance of the factor. Once analyzed if the
42 correlation is high between the expert scores and the cluster groups, then the clusters are
43 meaningful and valid. Furthermore, a cutoff for most efficient score above which to refer will be
44 chosen using a graph and observing the value that optimizes the sensitivity, specificity and overall
45 accuracy of the index. Finally, four grades of treatment-need will be determined using the twenty-
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60 five percentile ranges. The grades will be:

- 1- Minimal Need
- 2- Mild Need
- 3- Moderate Need
- 4- Severe Need

DISCUSSION

The development plan of this index has been conceived through a modification of the WHO Handbook for guideline development,³⁴ as well as reviewing the orthodontic literature for ways indices have been previously developed. The WHO provided an excellent starting point, from there modifications were made to cater to the specifics of this study, given that there are differences between developing a guideline and an index for orthodontic treatment need. The literature was useful, and among the index development protocols reviewed, certain assessed the orthodontic treatment need within the entire population,³⁷⁻⁴⁰ while others assessed it for a given subpopulation⁴¹⁻⁴⁶; each had strengths and weaknesses, and thus we further modified our methods, synthesizing a protocol for our particular needs from the available literature and using the experience and expertise of the authors. Through this protocol we aim to develop an index that fulfills all of the following criteria:⁴⁷

1. Gradient of Numeric Values: The severity of the orthodontic treatment need within the pediatric OSA patients should be defined within a numerical scheme that demonstrates a finite and progressive gradient from low need to high need.
2. Equal Sensitivity: should demonstrate equal sensitivity throughout the scale.
3. Clinical Importance: The numerical scale should correspond with the clinically appraised orthodontic treatment need of pediatric OSA patients.
4. Statistical Ease: should be amendable to statistical analysis.
5. Reliability: Should have a high intra- and inter-rater reliability.
6. Practical: The instruments required to score the index should be practical to the setting in which it will be administered.
7. Minimal Judgment: Applying the index should require minimal judgment.
8. Simple: The index shouldn't have a high financial or time cost, therefore should be simple enough to administer to many patients.
9. Detect Change: The index should be able to detect changes in orthodontic treatment need in pediatric OSA patients.

1
2
3 10. Validity: should be valid over time.
4

5 Validity can be characterized into different types: Face Validity, Content Validity, Construct
6 Validity, and Criterion Validity.⁴⁸ In this study, we will examine these kinds of validity at different
7 stages of development. The first draft of the index will focus on establishing face validity.
8 Feedback from the steering committee and IDG will assist in establishing content and construct
9 validity. Assuming that the “gold standard” in assessing the orthodontic treatment-need in pediatric
10 patients with OSA is an orthodontist with experience in dealing with pediatric OSA patients, then
11 the subsequent modification of the index based on the reliability tests and the dichotomous
12 responses from the orthodontists provide the index with the necessary criterion-related validity
13 evidence through statistical means.⁴⁹
14
15
16
17
18
19
20

21 **Significance**

22
23
24 This index will help physicians and other medical professionals identify which craniofacial
25 phenotypes may benefit from orthodontic treatment as part of their multi-disciplinary OSA
26 management. Furthermore, due to the diverse medical effects of sleep deprivation, there will be a
27 trend to make sleep apnea into a centralized service, where the main focus is for a highly trained
28 multidisciplinary team to treat a high volume of patients to a standardized protocol, where
29 meticulous documentation is exercised. This index is part of that documentation process. It will
30 allow for quality assurance, funding allocation and epidemiologic studies to be performed. It will
31 also allow long-term follow up and audit in order to enter into comparisons with other centers.
32
33
34
35
36
37

38 **Dissemination Plan**

39
40 The dissemination of this index will be done through a variety of ways in order to maximize
41 its reach. Primarily, it will be published in a peer-reviewed journal, which will allow its
42 introduction to the scientific literature. The journal should be a respected medical journal with
43 broad reach, in order to allow the greatest number of physicians to be exposed to the index.
44 Subsequently it will be translated to French, in order for it to be accessible to the entire Canadian
45 and American population of medical professionals. Moreover, the index will be presented at
46 national and international conferences to increase the awareness of the index among the scientific
47 community. Finally, the index will be used at the University of Alberta’s Interdisciplinary Airway
48 Research Center, and more research, so that future research in this center will incorporate it. It will
49
50
51
52
53
54
55
56
57
58
59
60

also be placed on the University of Alberta's Interdisciplinary Airway Research Center's website under the physician section, to further educate the doctors who visit the site on the index.

Funding

No funding has been provided for this project.

Authors' Contributions

MA HS MRR MM JM PM conceived and designed the study; MA drafted the manuscript and integrated critical feedback from all of the other authors. All of the authors approved the final version of the manuscript.

Ethics approval

The proposed research has received ethical approval numbered Pro00045067 from the University of Alberta Ethics Board.

Competing Interests

The authors declare no conflicts of interests.

REFERENCES

- .1 O'Brien LM, Mervis CB, Holbrook CR, Bruner JL, Klaus CJ, Rutherford J, et al. Neurobehavioral implications of habitual snoring in children. *Pediatrics* 2004;114(1):44-9.
- .2 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proceedings of the American Thoracic Society* 2008;5(2):242-52.
- .3 Gislason T, Benediktsdottir B. Snoring, apneic episodes, and nocturnal hypoxemia among children 6 months to 6 years old. An epidemiologic study of lower limit of prevalence. *Chest* 1995;107(4):963-6.
- .4 Ali NJ, Pitson DJ, Stradling JR. Snoring, sleep disturbance, and behaviour in 4-5 year olds. *Archives of disease in childhood* 1993;68(3):360-6.
- .5 Redline S, Tishler PV, Schluchter M, Aylor J, Clark K, Graham G. Risk factors for sleep-disordered breathing in children. Associations with obesity, race, and respiratory problems. *American journal of respiratory and critical care medicine* 1999;159(5 Pt 1):1527-32.
- .6 Bixler EO, Vgontzas AN, Lin HM, Liao D, Calhoun S, Vela-Bueno A, et al. Sleep disordered breathing in children in a general population sample: prevalence and risk factors. *Sleep* 2009;32(6):731-6.

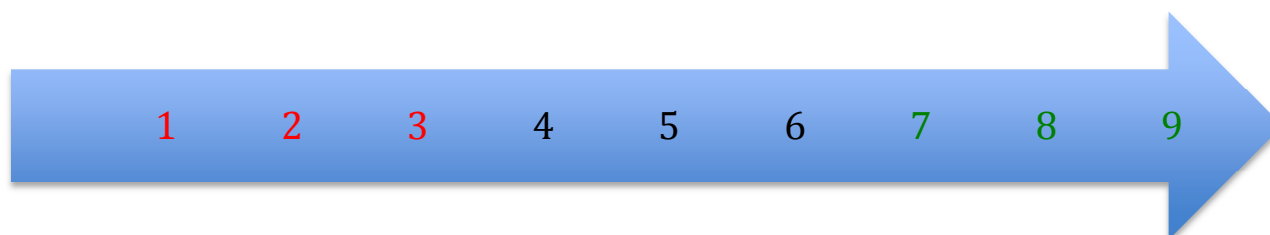
- 1
2
3 .7Li AM, So HK, Au CT, Ho C, Lau J, Ng SK, et al. Epidemiology of obstructive sleep apnoea
4 syndrome in Chinese children: a two-phase community study. *Thorax* 2010;65(11):991-7.
5
6 .8O'Brien LM, Holbrook CR, Mervis CB, Klaus CJ, Bruner JL, Raffield TJ, et al. Sleep and
7 neurobehavioral characteristics of 5- to 7-year-old children with parentally reported
8 symptoms of attention-deficit/hyperactivity disorder. *Pediatrics* 2003;111(3):554-63.
9
10 .9Huynh NT, Morton PD, Rompre PH, Papadakis A, Remise C. Associations between sleep-
11 disordered breathing symptoms and facial and dental morphometry, assessed with screening
12 examinations. *American journal of orthodontics and dentofacial orthopedics : official
13 publication of the American Association of Orthodontists, its constituent societies, and the
14 American Board of Orthodontics* 2011;140(6):762-70.
15
16
17 .10Beebe DW. Neural and neurobehavioral dysfunction in children with obstructive sleep apnea.
18 *PLoS medicine* 2006;3(8):e323.
19
20
21 .11Chervin RD, Archbold KH, Dillon JE, Panahi P, Pituch KJ, Dahl RE, et al. Inattention,
22 hyperactivity, and symptoms of sleep-disordered breathing. *Pediatrics* 2002;109(3):449-56.
23
24 .12Crabtree VM, Varni JW, Gozal D. Health-related quality of life and depressive symptoms in
25 children with suspected sleep-disordered breathing. *Sleep* 2004;27(6):1131-8.
26
27
28 .13Halbower AC, Degaonkar M, Barker PB, Earley CJ, Marcus CL, Smith PL, et al. Childhood
29 obstructive sleep apnea associates with neuropsychological deficits and neuronal brain
30 injury. *PLoS medicine* 2006;3(8):e301.
31
32 .14Bar A, Tarasiuk A, Segev Y, Phillip M, Tal A. The effect of adenotonsillectomy on serum
33 insulin-like growth factor-I and growth in children with obstructive sleep apnea syndrome.
34 *The Journal of pediatrics* 1999;135(1):76-80.
35
36
37 .15Marcus CL, Greene MG, Carroll JL. Blood pressure in children with obstructive sleep apnea.
38 *American journal of respiratory and critical care medicine* 1998;157(4 Pt 1.103-1098:(
39
40 .16Amin RS, Kimball TR, Bean JA, Jeffries JL, Willging JP, Cotton RT, et al. Left ventricular
41 hypertrophy and abnormal ventricular geometry in children and adolescents with obstructive
42 sleep apnea. *American journal of respiratory and critical care medicine* 2002;165(10):1395-
43 9.
44
45
46 .17Sofer S, Weinhouse E, Tal A, Wanderman KL, Margulis G, Leiberman A, et al. Cor pulmonale
47 due to adenoidal or tonsillar hypertrophy or both in children. Noninvasive diagnosis and
48 follow-up. *Chest* 1988;93(1):119-22.
49
50
51 .18Korayem MM, Witmans M, MacLean J, Heo G, El-Hakim H, Flores-Mir C, et al. Craniofacial
52 morphology in pediatric patients with persistent obstructive sleep apnea with or without
53 positive airway pressure therapy: a cross-sectional cephalometric comparison with controls.
54 *American journal of orthodontics and dentofacial orthopedics : official publication of the
55 American Association of Orthodontists, its constituent societies, and the American Board of
56 Orthodontics* 2013;144(1):78-85.
57
58
59
60

- 1
2
3 .19Katyal V, Pamula Y, Martin AJ, Daynes CN, Kennedy JD, Sampson WJ. Craniofacial and
4 upper airway morphology in pediatric sleep-disordered breathing: Systematic review and
5 meta-analysis. *American journal of orthodontics and dentofacial orthopedics : official*
6 *publication of the American Association of Orthodontists, its constituent societies, and the*
7 *American Board of Orthodontics* 2013;143(1):20-30 e3.
8
9
10 .20Flores-Mir C, Korayem M, Heo G, Witmans M, Major MP, Major PW. Craniofacial
11 morphological characteristics in children with obstructive sleep apnea syndrome: a
12 systematic review and meta-analysis. *Journal of the American Dental Association*
13 2013;144(3):269-77.
14
15 .21Carvalho FR, Lentini-Oliveira D, Machado MA, Prado GF, Prado LB, Saconato H. Oral
16 appliances and functional orthopaedic appliances for obstructive sleep apnoea in children.
17 *The Cochrane database of systematic reviews* 2007(2):CD005520.
18
19
20 .22Villa MP, Bernkopf E, Pagani J, Broia V, Montesano M, Ronchetti R. Randomized controlled
21 study of an oral jaw-positioning appliance for the treatment of obstructive sleep apnea in
22 children with malocclusion. *American journal of respiratory and critical care medicine*
23 2002;165(1):123-7.
24
25
26 .23Schutz TC, Dominguez GC, Hallinan MP, Cunha TC, Tufik S. Class II correction improves
27 nocturnal breathing in adolescents. *The Angle orthodontist* 2011;81(2):222-8.
28
29
30 .24Cozza P, Polimeni A, Ballanti F. A modified monobloc for the treatment of obstructive sleep
31 apnoea in paediatric patients. *Eur J Orthod* 2004;26(5):523-30.
32
33 .25Zhang C, He H, Ngan P. Effects of twin block appliance on obstructive sleep apnea in children:
34 a preliminary study. *Sleep & breathing = Schlaf & Atmung* 2013;17(4):1309-14.
35
36 .26Pliska BT, Almeida F. Effectiveness and outcome of oral appliance therapy. *Dental clinics of*
37 *North America* 2012;56(2):433-44.
38
39 .27Hiyama S, Suda N, Ishii-Suzuki M, Tsuiki S, Ogawa M, Suzuki S, et al. Effects of maxillary
40 protraction on craniofacial structures and upper-airway dimension. *The Angle orthodontist*
41 2002;72(1):43-7.
42
43
44 .28Oktay H, Ulukaya E. Maxillary protraction appliance effect on the size of the upper airway
45 passage. *The Angle orthodontist* 2008;78(2):209-14.
46
47 .29Kilinc AS, Arslan SG, Kama JD, Ozer T, Dari O. Effects on the sagittal pharyngeal dimensions
48 of protraction and rapid palatal expansion in Class III malocclusion subjects. *Eur J Orthod*
49 2008;30(1):61-6.
50
51
52 .30Sayinsu K, Isik F, Arun T. Sagittal airway dimensions following maxillary protraction: a pilot
53 study. *Eur J Orthod* 2006;28(2):184-9.
54
55
56 .31Kaygisiz E, Tuncer BB, Yuksel S, Tuncer C, Yildiz C. Effects of maxillary protraction and
57 fixed appliance therapy on the pharyngeal airway. *The Angle orthodontist* 2009;79(4):660-7.
58
59
60

- 1
2
3 .32Lee JW, Park KH, Kim SH, Park YG, Kim SJ. Correlation between skeletal changes by
4 maxillary protraction and upper airway dimensions. *The Angle orthodontist* 2011;81(3):426-
5 32.
6
7 .33Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive
8 sleep apnea: a systematic review and meta-analysis. *Sleep medicine reviews* 2010;14(5):287-
9 97.
10
11 .34World Health Organization (2012) WHO Handbook for guideline development. Available:
12 http://www.who.int/hiv/topics/mtct/grc_handbook_mar2010_1.pdf. Accessed 10 Sept 2013.
13
14
15 .35Rowe G, Wright G. The Delphi technique as a forecasting tool: issues and analysis. *Int J*
16 *Forecasting* 1999;15(4):353-75.
17
18 .36Dalkey N, Helmer O. An Experimental Application of the Delphi Method to the use of experts.
19 *Management Science* 1963;9(3):458-67.
20
21 .37Richmond S, Shaw WC, O'Brien KD, Buchanan IB, Jones R, Stephens CD, et al. The
22 development of the PAR Index (Peer Assessment Rating): reliability and validity. *Eur J*
23 *Orthod* 1992;14(2):125-39.
24
25 .38Richmond S, Shaw WC, Roberts CT, Andrews M. The PAR Index (Peer Assessment Rating):
26 methods to determine outcome of orthodontic treatment in terms of improvement and
27 standards. *Eur J Orthod* 1992;14(3):180-7.
28
29 .39Daniels C, Richmond S. The development of the index of complexity, outcome and need
30 (ICON). *J Orthod* 2000;27(2):149-62.
31
32 .40Brook PH, Shaw WC. The development of an index of orthodontic treatment priority. *Eur J*
33 *Orthod* 1989;11(3):309-20.
34
35 .41Mars M, Batra P, Worrell E. Complete unilateral cleft lip and palate: validity of the five-year
36 index and the Goslon yardstick in predicting long-term dental arch relationships. *Cleft*
37 *Palate Craniofac J* 2006;43(5):557-62.
38
39 .42Mars M, Plint DA, Houston WJ, Bergland O, Semb G. The Goslon Yardstick: a new system of
40 assessing dental arch relationships in children with unilateral clefts of the lip and palate.
41 *Cleft Palate J* 1987;24(4):314-22.
42
43 .43Johnson N, Sandy J. An aesthetic index for evaluation of cleft repair. *Eur J Orthod*
44 2003;25(3):243-9.
45
46 .44Mossey PA, Clark JD, Gray D. Preliminary investigation of a modified Huddart/Bodenham
47 scoring system for assessment of maxillary arch constriction in unilateral cleft lip and palate
48 subjects. *Eur J Orthod* 2003;25(3):251-7.
49
50 .45Tohill C, Mossey PA. Assessment of arch constriction in patients with bilateral cleft lip and
51 palate and isolated cleft palate: a pilot study. *Eur J Orthod* 2007;29(2):193-7.
52
53
54
55
56
57
58
59
60

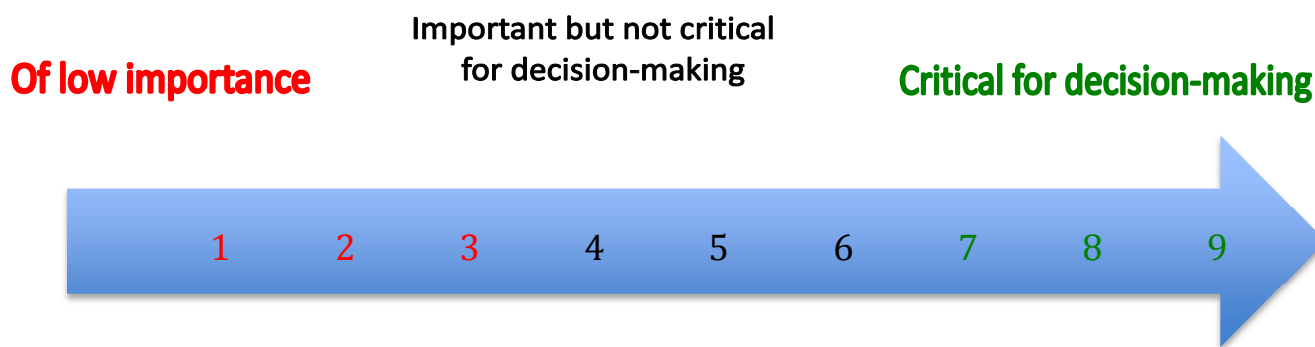
- 1
2
3 .46Huddart AG, Bodenham RS. The evaluation of arch form and occlusion in unilateral cleft palate
4 subjects. *Cleft Palate J* 1972;9:194-209.
5
6 .47Summers CJ. The occlusal index: a system for identifying and scoring occlusal disorders. *Am J*
7 *Orthod* 1971;59(6):552-67.
8
9 .48Streiner DL, Norman GR. *Health measurement scales: a practical guide to their development*
10 *and use*. Oxford: Oxford Medical Publications, 1995.
11
12 .49Terwee CB, Bot SDM, de Boer MR, van der Windt DAWM, Knol DL, Dekker J, et al. Quality
13 criteria were proposed for measurement properties of health status questionnaires. *J Clin*
14 *Epidemiol* 2007;60(1):34-42.
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

For peer review only

1 Assigned Number _____
23 Factor Being Evaluated: _____
4
5
67
8 Important but not critical
9 Of low importance for decision-making Critical for decision-making
1011
12
13
14
15
16
17
18
19 Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.
20

- 21
22 1- In your opinion, is this factor commonly observed in Pediatric Obstructive Sleep Apnea patients?
23 Yes 1 2 3 4 5 6 7 8 9
24 No
25
- 26 2- In your opinion, does this factor contribute to Pediatric Obstructive Sleep Apnea symptoms?
27 Yes 1 2 3 4 5 6 7 8 9
28 No
29
- 30 3- In your opinion, would correcting this factor help diminish the symptoms of Pediatric Obstructive
31 Sleep Apnea?
32 Yes 1 2 3 4 5 6 7 8 9
33 No
34
- 35 4- In your opinion, does this factor contribute to orthodontic treatment need?
36 Yes 1 2 3 4 5 6 7 8 9
37 No
38
- 39 5- In your opinion, is the 5-point scale appropriate for this factor?
40 Yes 1 2 3 4 5 6 7 8 9
41 No; How would you correct it?
42
43
44
45
46
-
- 47 6- Are the points on the scale attributed correctly for this factor?
48 Yes 1 2 3 4 5 6 7 8 9
49 No; How would you correct it?
50
51
52
53
54
-
- 55 7- In your opinion, are the pictures appropriate for this factor?
56 Yes 1 2 3 4 5 6 7 8 9
57 No; How would you correct it?
58
59
60

Assigned Number _____



Circle the "Yes" or "No" answers. If "Yes" rate the importance from 1-9 by circling the appropriate number.

1. Do you think that this index is useful in fulfilling the goals of having an easy to use index that helps non-dentally trained professionals assess the orthodontic treatment need to help the symptoms in pediatric patients with OSA?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

2. Do you believe that the organizing committee is developing this index in the appropriate way? (i.e. Reviewing the recommendations in the literature, seeking expert opinions, gaining end-user approval and testing its reliability)

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

3. In your opinion were all the factors, identified in this meeting, important for this index?

Yes 1 2 3 4 5 6 7 8 9

No; Which factors were not important?

4. In your opinion, have all the important areas concerning orthodontic treatment need to help symptoms of OSA been identified?

Yes 1 2 3 4 5 6 7 8 9

No; Which ones were missed?

5. Is the layout of the index easy to navigate?

Yes 1 2 3 4 5 6 7 8 9

No; How would you improve it?

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Please provide any other feedback below that may help improve the index:

For peer review only