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Aesthetic Outcome of Maxillomandibular Advancement Surgery for the Treatment of Obstructive Sleep Apnea

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Graduate Program in Orthodontics

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Abstract

Introduction: Maxillomandibular advancement (MMA) surgery has been shown to be a successful treatment for obstructive sleep apnea (OSA); however, concerns may exist regarding aesthetic outcomes due to the large advancements involved.

Objectives: To evaluate facial profile changes in patients who underwent MMA surgery for OSA utilizing panels of oral and maxillofacial surgeons, orthodontists and laypeople.

Methods: Pre- and post-treatment silhouettes of patients who underwent MMA surgery for OSA were generated from profile photographs. Silhouettes were shown to surgeons, orthodontists and laypeople, who assessed aesthetics via a visual analogue scale (VAS).

Results: Positive, or neutral, post-surgical change was found in 21 of 22 patients. A significant difference was found between ratings of surgeons and orthodontists as compared to laypeople for 12 of 22 patients (p < 0.05).

Conclusions: MMA surgery for OSA does not have a negative impact on facial aesthetics. Surgeons and orthodontists reported more positive post-surgical change than laypeople; however, all groups of evaluators felt that post-surgical aesthetic changes were positive in the majority of patients.

Keywords

Obstructive Sleep Apnea (OSA), Sleep Disordered Breathing, Maxillomandibular Advancement (MMA), Orthognathic Surgery, Orthodontics, Visual Analogue Scale (VAS), Aesthetics, Profile Analysis
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Rick, my amazing husband, you have always been there for me every step of the way. Thank you for your patience, understanding, and encouragement. You are my best friend, and I love you with all my heart.

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“What if I fall?”
Oh but my darling,
What if you fly?
~Erin Hanson
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List of Abbreviations

AAO: American Association of Orthodontists
AHI: Apnea-Hypopnea Index
AS: Aesthetic Score
BMI: Body Mass Index
BSSO: Bilateral Sagittal Split Osteotomy
CAOMS: Canadian Association of Oral and Maxillofacial Surgeons
CPAP: Continuous Positive Airway Pressure
ESS: Epworth Sleepiness Scale
FOSQ: Functional Outcomes of Sleep Questionnaire
ICC: Intraclass Correlation Coefficient
IRR: Intra-rater Reliability
LAY: Laypeople
MMA: Maxillomandibular Advancement
MSLT: Multiple Sleep Latency Test
OMFS: Oral and Maxillofacial Surgeons
ORTHO: Orthodontists
OSA: Obstructive Sleep Apnea
OSAS: Obstructive Sleep Apnea Syndrome
PPG: Photoplethysmography
PSG: Polysomnography
QOL: Quality of Life
RDI: Respiratory Disturbance Index
REB: Research Ethics Board
REM: Rapid Eye Movement
RERAs: Respiratory Effort-Related Arousals
UPPP: Uvulopalatopharyngoplasty
VAS: Visual Analogue Scale
Chapter 1
Literature Review

1.1 Introduction

Obstructive sleep apnea (OSA) is a common, but often unrecognized, sleep disorder that may have serious consequences on quality of life and overall health. It is characterized by repeated occurrences of complete or partial airway obstruction during sleep due to pharyngeal collapse. This results in airflow cessation (apnea), or reduction (hypopnea), respectively. An apneic event is described as the absolute cessation of airflow, despite an effort to breathe, for a minimum of 10 seconds. In comparison, a hypopneic event is identified by (i) at least a 30 percent decrease in airflow (as compared to baseline) in excess of 10 seconds with a 4 percent or more oxygen desaturation, or (ii) at least a 50 percent decrease in airflow in excess of 10 seconds with at least a 3 percent oxygen desaturation or an arousal. A detailed medical history and various clinical examinations are used in order to arrive at an accurate diagnosis of OSA and to quantify the severity of the disorder. Controlled sleep studies in conjunction with diurnal and nocturnal symptoms as reported by the patient are used in diagnosis. From this information, appropriate methods for treatment may be selected.

1.2 Epidemiology of OSA

Overall, the prevalence of OSA is high in the general population; however, there have been inconsistencies in methodology for the study of OSA that
have resulted in a wide variation in reported prevalence. A recent review\(^1\) has determined overall adult population prevalence of OSA to be 9 to 38 percent. In addition, the American Heart Association and the American College of Cardiology advised that up to 82 percent of men and 93 percent of women with clinically significant OSA have not been formally diagnosed.\(^2\)

It is well recognized that some groups have a much higher prevalence of OSA. The overall prevalence of OSA is greater in male patients, older patients, and patients with a higher body-mass index.\(^1\) Increasing age correlates with an increasing incidence of OSA, and some studies have reported this disorder to be as high as 90% in men and 78% of women in elderly age groups.\(^3\) It is thought that increasing age is potentially related to hormonal differences, comorbidities that increase the incidence of OSA, neurological decline, and general changes in sleep cycle that may influence the development of OSA.\(^4,5\)

One reason for the concern in growing incidence of OSA is the increase in population averages for body mass index (BMI). Obesity has been identified as a major risk factor for OSA, and more than one-third of the American population is clinically obese according to the Centers for Disease Control and Prevention.\(^6\) This is particularly significant as every 10 percent weight gain increases the risk of OSA by a factor of six.\(^7\) It has been postulated that an increase in weight may increase OSA risk by causing changes in the structure or function of the upper airway, reducing lung capacity, and/or producing a discrepancy between ventilatory drive and load.\(^8\) Men seem to be predisposed to OSA due to the androgenic pattern of body fat distribution, specifically in the trunk and neck areas. In addition,
neurologic control of upper airway ventilation and muscle function are influenced by sex hormones, which also may play a role in the predisposition of men toward OSA. Significantly, obesity and OSA present with various comorbidities that have drastic impacts on overall health, including cardiovascular disease, hypertension, stroke, metabolic syndrome, and type 2 diabetes mellitus.

1.3 Pathophysiology of OSA

The presentation of OSA during sleep is cyclical in nature. Pharyngeal airway collapse despite respiratory effort arises in a repetitive manner during the sleep cycle in patients with OSA. The resultant complete or partial airway reduction occurs from an anatomic pharyngeal narrowing and/or a decrease in muscle tonicity that allows the base of the tongue to lie against the posterior aspect of the pharyngeal wall. As ventilation effort increases in response to a decrease in airflow, an arousal episode occurs, which then restores muscle tone and airway patency. As this pattern repeats multiple times per hour of sleep, insufficient rapid eye movement (REM) sleep is obtained. Repeated hypoxemia and fragmented sleep episodes contribute to both psychological and physiological comorbidities.

Typical nocturnal presentation of OSA includes snoring, gasping, witnessed apnea events, nocturnal awakening, and nocturia. A disruption in sleep pattern manifests as daytime sleepiness and fatigue. Individuals with OSA have been shown to display hypersomnia, cognitive dysfunction, early morning headaches, depression, and irritability. Daytime fatigue can have debilitating and
dangerous consequences on daily life. It has been reported that the incidence of motor vehicle accidents increases at least two times, and possibly up to seven-fold, in individuals affected by OSA.\textsuperscript{19}

Some of the anatomic factors that predispose to OSA include obesity, nasal septum deviation, chronic nasal congestion, decreased nasopharyngeal volume, and craniofacial abnormalities.\textsuperscript{20,21} Factors such as elongation of the soft palate, macroglossia, and tonsil/adenoid hypertrophy may also have an anatomical effect on airway reduction.\textsuperscript{22,23} Maxillary retrognathia, mandibular retrognathia, increased lower face vertical dimension, and inferior position of the hyoid bone have been shown to have a strong correlation with the development of OSA.\textsuperscript{24} In patients showing these anatomical presentations, tongue position tends to be lower and more posterior than normal, which then results in pharyngeal obstruction during sleep.

Although a decrease in muscle tone occurs in the pharyngeal muscles in all individuals during sleep, in patients with other comorbidities leading to a predisposition of OSA, pharyngeal collapse may cause a cessation in respiration.\textsuperscript{25} The retropalatal and retroglossal areas of the oropharynx are the most common location of pharyngeal collapse; however, hypopharyngeal areas may also be affected in some individuals.\textsuperscript{26} The degree of obstruction may be partial or total, and can be associated with soft tissue factors such as pharyngeal tonsillar hypertrophy or fatty infiltration of the pharyngeal wall in obese patients.\textsuperscript{27} In OSA patients with concurrent obesity, primary collapse has been shown to occur in the velopharyngeal area; in non-obese patient with OSA and mandibular retrognathia,
pharyngeal collapse is most commonly observed in both the velopharyngeal and oropharyngeal regions.\textsuperscript{28}

With pharyngeal wall collapse, the result is a complete or partial airway obstruction.\textsuperscript{18} It is important to note that these “obstructive” events comprise obstructive sleep apnea. In comparison, the brain stem respiratory motor output may be reduced, which results in a cessation or reduction in respiration despite an unobstructed airway.\textsuperscript{29} This is diagnosed as “central” sleep apnea. Central sleep apnea may less commonly present in combination with an obstructive form, whereby cerebral mechanisms controlling pharyngeal muscles do not function properly, and allow pharyngeal collapse.\textsuperscript{30}

When pharyngeal collapse occurs and an attempt at inhalation is made, an increase in negative intrathoracic pressure results. In normal inhalation, the chest and abdomen expand, which results in negative intrathoracic pressure that becomes balanced through the inhalation of air.\textsuperscript{31} In the absence of inhalation, due to a partial or complete airway blockage, increased intrathoracic pressure occurs and the cardiovascular system is taxed in an attempt at blood circulation. Oxygen desaturation, increased heart rate, and increased blood pressure are noted.\textsuperscript{32} It has been suggested that this mechanism may be related to the comorbidities often seen with OSA, such as hypertension, atrial fibrillation, cardiovascular disease, congestive heart failure, and stroke.\textsuperscript{33}
1.4 Diagnosis of OSA

In order to diagnose a patient with sleep apnea, a variety of methods are employed, such as a detailed medical history, questionnaires, and various clinical tests. Questionnaires are used to inquire about sleeping habits, and both nocturnal and daytime symptoms. The Epworth Sleepiness Scale (ESS)\textsuperscript{34} is a self-report questionnaire and one of the most widely used tests in initial screening for OSA. It uses eight situations (such as “sitting and reading”, or “watching TV”) and inquires about the likelihood of dozing during each, in order to measure a patient's likelihood of falling asleep during routine daily activities. Objective measures of sleepiness may be obtained via a Multiple Sleep Latency Test (MSLT)\textsuperscript{35}, which measures the amount of time required for an individual to fall asleep during the day. As the cost of administering the ESS is significantly less than the MSLT, and the convenience is higher, the ESS is often used as the definitive screening tool for OSA.\textsuperscript{36}

Currently, polysomnography (PSG) is the gold-standard for diagnosis of OSA.\textsuperscript{18} A controlled sleep study is used to measure the number of apneic and hypopneic events per hour of sleep. In this manner, an apnea-hypopnea index (AHI)\textsuperscript{37} may be identified to provide a quantitative measure of the OSA severity. A diagnosis of OSA is reached if five or more episodes of apnea and/or hypopnea are observed per hour of sleep, and the patient reports associated symptoms. Likewise, a patient is also diagnosed with OSA if fifteen or more episodes of apnea and/or hypopnea are observed per hour of sleep, regardless of associated symptoms.\textsuperscript{37} The values of the AHI may be used to classify an individual as having
mild (AHI 5-14), moderate (AHI 15-29) or severe (AHI ≥30) OSA, which provides an objective categorization of disorder severity.\textsuperscript{38}

The respiratory disturbance index (RDI)\textsuperscript{39} may be used in conjunction with the AHI. Measured during a sleep study, usually concurrently with the AHI, the RDI assesses the number of apneic and hypopneic events that occur, as well as the respiratory effort-related arousals (RERAs). These arousals occur due to increased strain on respiratory activity, but do not qualify as actual apneas or hypopneas.\textsuperscript{40}

Due to the increasing prevalence of OSA, and the cost and inconvenience of PSG, reliable methods for at-home diagnosis are needed. Although a recent study found that measuring blood volume changes and oxygen saturation using photoplethysmography (PPG) has a significant positive correlation with the results obtained through traditional PSG, especially in individuals with moderate or severe OSA,\textsuperscript{41} most of the literature refutes this claim due to poor sensitivity and specificity.\textsuperscript{42} A recent meta-analysis\textsuperscript{43} evaluating efficacy of laboratory and home sleep studies found that RDI values on portable devices were, on average, 10% lower when compared to laboratory studies. For this reason, it was concluded that although similar diagnostic information was obtained from portable and laboratory devices, at-home sleep studies tended to underestimate the severity of OSA. Although it may be possible to diagnose a patient with severe OSA via a portable device, the sensitivity of at-home sleep studies is not yet able to identify those patients with mild forms of the disorder.
Intermittent oxygen desaturation resulting from apnea or hypopnea events is present in many patients suffering from OSA, and oxygen saturation levels are considered to be damaging when they drop below 90%.\textsuperscript{44} It has been suggested that the use of a pulse oximeter to evaluate blood oxygen saturation during sleep may be used as an at-home tool to monitor the degree of OSA and the response of a patient to treatment modalities.\textsuperscript{45} Oxygen desaturation levels are not a consistent marker of the degree of OSA in all patients, so care must be used when relying on this method for diagnosis and treatment. It remains important to have a laboratory diagnosis with baseline oxygen desaturation level prior to attempting to use this form of at-home monitoring. Current recommendations include the use of at-home sleep studies for patients that already have an established baseline from a laboratory sleep study, or in undiagnosed patients that have a high probability of having OSA based on an ESS greater than 12 and clinical symptoms without severe co-morbidities.\textsuperscript{46}

1.5 Treatment of OSA

It is critical to have a definitive diagnosis of OSA, and a classification of its severity via a PSG sleep study, prior to commencing treatment. After a diagnosis has been obtained, non-surgical and surgical approaches may be used to control, or eliminate, the signs and symptoms of OSA. Of the non-surgical methods, weight loss, pharmacological approaches, oral appliances, and/or continuous positive airway pressure (CPAP) devices may be used.
Often, the initial approach in the treatment of OSA is reduction of BMI. As obesity is the primary contributing factor to OSA,7 a reduction in fat deposits has been shown to reduce the severity of OSA. It has been shown that a reduction of 10.7 kilograms of weight results in a 40% reduction in AHI in individuals with mild forms of OSA.47 Likewise, the risk of developing moderate or severe OSA increases 5-fold when BMI is in excess of 28.48

Various medications have been suggested that specifically target an increase in pharyngeal dilator muscle tone, reduction in airway resistance, or improve pharyngeal surface tension. Decrease in nasal congestion using fluticasone nasal spray has not been shown to affect AHI or significantly improve daytime sleepiness.49 Other pharmacological approaches may be directed toward improving inspiratory drive.50 Serum testosterone has been shown to be lower in men with OSA51 and some studies recommend testosterone supplementation to alter ventilator response through central chemoreceptors.50,51 Currently, testosterone replacement therapy is advocated only in conjunction with other OSA treatments.52 Central nervous system stimulants, such as modafinil and armodafinil, have been shown to assist with residual daytime fatigue in patients using CPAP therapy, without affecting normal sleep architecture.53 Oxygen therapy has been recommended for patients who are unable to tolerate CPAP and who are not candidates for a surgical procedure. Although oxygen does not change the RDI, it has been shown to increase oxygen saturation and alleviate OSA-related symptoms.44 Overall, pharmacologic therapy may have use as an adjunctive
treatment; however, there is presently insufficient evidence to advise the use of medications as a part of the primary treatment recommendations for OSA.\textsuperscript{54}

Oral appliances function to move the mandible anteriorly and can be relatively successful in the treatment of mild or moderate OSA. In repositioning the mandible anteriorly, the tongue, hyoid bone and associated musculature are concurrently advanced, which increases airway patency. Although not as effective as continuous positive airway pressure (CPAP), which is discussed below, the advantages of oral appliance therapy include decreased cost, decreased noise, and increased patient comfort.\textsuperscript{55} However, as oral appliances only affect retroglossal airway obstruction, they are not as effective as CPAP at reducing patient symptoms or decreasing AHI below 5.\textsuperscript{56} In addition, temporomandibular joint (TMJ) discomfort, tooth movement, and downward mandibular rotation may be side-effects of oral appliance use.\textsuperscript{57}

Presently, the most effective non-surgical method of reducing or eliminating OSA signs and symptoms is CPAP.\textsuperscript{58} Continuous pressure is applied during the breathing cycle which prevents or reduces the pharyngeal airway collapse. Despite the fact that CPAP remains the gold standard for treatment of OSA,\textsuperscript{58} many patients remain dissatisfied with this form of treatment. Nasal, pharyngeal, and ocular dryness, nasal congestion, rhinorrhea, and skin irritation and dermatitis are among the most common complaints of CPAP. Gastric distension, anxiety, claustrophobia, nasal obstruction, and lack of perceived benefit are also cited as patient concerns. It has been found that acceptance rates vary
from 50-95 percent, and 12-15 percent of patients will stop CPAP treatment within three years. Of patients using CPAP regularly, adherence has been found to vary from 40-80%.

Surgical treatments for OSA may include significantly more invasive procedures (Table 1). Tracheostomy is highly effective in providing a cure for OSA, but due to the risks and long-term care required, it is primarily reserved for patients with life-threatening OSA who cannot tolerate CPAP and have failed other surgical methods. Techniques that modify the upper airway may be site specific and target the nasal septum, turbinates, tonsils/adenoids, tongue, and/or oropharynx in the form of uvulopalatalpharyngoplasty (UPPP). These methods are used to improve airflow and reduce soft tissue bulk that may result in airway obstruction.
Table 1. Surgical procedures for the treatment of obstructive sleep apnea by target site. (Adapted from Smith et al.62)

<table>
<thead>
<tr>
<th>Target Site</th>
<th>Surgical Procedure</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nasal</td>
<td>Adenoidectomy</td>
</tr>
<tr>
<td></td>
<td>Superior Turbinate Reduction</td>
</tr>
<tr>
<td></td>
<td>Rhinoplasty</td>
</tr>
<tr>
<td>Oral/Palatal</td>
<td>Septoplasty</td>
</tr>
<tr>
<td></td>
<td>Tonsilar Pillar Implants</td>
</tr>
<tr>
<td></td>
<td>Tonsillectomy</td>
</tr>
<tr>
<td></td>
<td>Uvulopalatopharyngoplasty</td>
</tr>
<tr>
<td>Hypopharyngeal</td>
<td>Genioglossus Advancement</td>
</tr>
<tr>
<td></td>
<td>Glossectomy</td>
</tr>
<tr>
<td></td>
<td>Hyoid Suspension</td>
</tr>
<tr>
<td></td>
<td>Lingual Tonsillectomy</td>
</tr>
<tr>
<td>Other</td>
<td>Hypoglossal Nerve Stimulation</td>
</tr>
<tr>
<td></td>
<td>Maxillomandibular Advancement</td>
</tr>
<tr>
<td></td>
<td>Tracheostomy</td>
</tr>
</tbody>
</table>

UPPP is the most common procedure to modify the upper airway and usually involves tonsillectomy, adenoidectomy, uvulectomy, and excision of excess pharyngeal and soft palate tissue.63 Velopharyngeal insufficiency is an established complication, which may cause nasal reflux, dysphagia, and a nasal quality to vocal tones.64 In addition, significant postoperative pain and extended recovery time has been reported.65 Success rate of UPPP for the treatment of OSA has been shown to be highly variable (16 - 83%)66, and some studies have shown worsening of AHI in up to 44 percent of patients.67 It is presently recognized that patient selection for this procedure is very important, and UPPP can be quite successful in carefully
selected cases. The Friedman Clinical Staging System for Sleep-Disordered Breathing\textsuperscript{68} was developed to assist in the determination of the likelihood for success of UPPP in OSA treatment. This Four-Stage categorization is based on tonsil size, tongue-palate position (similar to Mallampati scoring\textsuperscript{62}), and BMI. An 80 percent and 40 percent success rate was found for UPPP as a treatment for OSA in patients with Friedman Stage I and Stage II anatomy respectively.\textsuperscript{68} Based on this information, UPPP may be helpful in patients with retropalatal and retrolingual obstruction; hypopharyngeal and multilevel obstruction patients are generally not good candidates for UPPP surgery.\textsuperscript{66}

Permanent methods to treat OSA, in the form of surgery, are often sought by patients due to concerns with other treatment approaches. Of the surgical methods available for the treatment of OSA, maxillomandibular advancement (MMA) has been shown to be the most effective, with multiple studies showing MMA to provide highly successful treatment for OSA.\textsuperscript{69,70,71} Due to the overall low morbidity and high success rate of MMA, combined with the variable success rate of other surgical methods, MMA can be considered the gold standard for surgical treatment of OSA.\textsuperscript{69} MMA surgery is a “telegnathic” surgery, with the purpose of lengthening the jaws, in contrast to an “orthognathic” surgery, which is used to straighten the jaws. For this reason, although orthodontic treatment is a component of many MMA surgeries for OSA, it is not an absolute requirement. MMA surgery for OSA involves a Le Fort I maxillary osteotomy\textsuperscript{72} with a concurrent bilateral sagittal split mandibular osteotomy (BSSO).\textsuperscript{73} It is common for a genial advancement to be simultaneously performed. In this manner, both
jaws are advanced to maximum degrees, and rigidly fixed into position. This results in the soft palate, tongue, hyoid, and other attached soft tissue structures being concurrently brought forward. Physical expansion of the pharyngeal airway has been shown to be the ultimate result of MMA surgery, which acts to counteract the pathophysiology of OSA. The soft-tissue structures composing the pharynx, such as the suprathyroid, palatal, and lateral pharyngeal muscles, are tightened by MMA surgery, which reduces or eliminates pharyngeal collapse.

The goals of MMA surgery in the treatment of OSA are to increase the cross-sectional dimension of the pharyngeal airway and prevent pharyngeal collapse during sleep. These changes may be verified through lateral cephalograms and cone-beam computed tomography where linear and volumetric airway increases may be visualized (Figure 1).
Figure 1a

Figure 1b

Figure 1. Lateral cephalometric radiographs taken before (Figure 1a) and after (Figure 1b) maxillomandibular advancement surgery. An increased change is shown post-surgically in the most occluded portion of the Inferior Airway Space (IAS).

1.6 Outcomes of MMA Surgery for OSA

The objective outcomes of MMA surgery are often evaluated by assessment of pre- and post-surgical AHI, or RDI, values. Various interpretations of AHI reduction have been used to define the overall surgical success rate. In a study by Smatt et al., of 18 patients undergoing MMA surgery for OSA, a post-operative AHI of 15 or less, with overall reduction of at least 50% from initial AHI, was used as the criteria for success. From this, an overall success rate of 84% was
described. In another study by Varghese et al.,\textsuperscript{80} of 24 patients, 42 percent of patients reached an AHI $<5$, with 71 percent achieving an AHI $\leq 10$. The RDI was used in a study by Li et al.\textsuperscript{81} in order to objectively assess the success rate of MMA surgery for OSA. A post-operative RDI of $<20$ was found in 95% of patients who underwent MMA surgery following previous soft tissue resection surgery. Pirklbauer et al.\textsuperscript{82} conducted a systematic review on the success of MMA surgery for OSA and found that success rates varied from 65 to 100 percent depending on which criteria were used in defining success rate.

It is important to note that although mandibular retrognathia and a long lower face height are risk factors in the development of OSA, skeletal Class I patients may also benefit from MMA surgery for OSA treatment. In a previous study, it was found that only 30 percent of patients presented with a skeletal Class II, whereas 60 percent were initially skeletal Class I, and only 10% were skeletal Class III.\textsuperscript{83} Ronchi et al.\textsuperscript{27} found that following MMA surgery for the treatment of OSA, patients with initial presentation of skeletal Class I or Class II showed equal benefit from this treatment.

The \textit{subjective} success rates of MMA surgery for OSA are equally as important as the objective measures. Various methods may be used to assess a patient’s satisfaction with the surgical outcome. Patient questionnaires such as the ESS\textsuperscript{84} and patient-reported quality of life (QOL) surveys such as Functional Outcomes of Sleep Questionnaire (FOSQ)\textsuperscript{84} may be used in order to assist in determining patient satisfaction levels. It is important that assurances can be made to patients undergoing MMA surgery for OSA that improvements to quality
of life will be obtained. Pre- and post-operative daytime sleepiness was assessed using the ESS to subjectively measure the success of MMA surgery for OSA in a study by Goodday et al. ESS values showed that pre-operatively 40% of patients were very sleepy, 32% were sleepy, and 28% were not sleepy. These values decreased significantly following surgery, with <1% of patients remaining in the very sleepy category, 9% remaining sleepy, and 90% not sleepy.

In another study of 22 patients, not only did mean AHI decrease from 42 to 7, but sleep quality, daytime function, and overall health (measured by physical, mental, emotional and sexual scales) improved postoperatively when measured via a QOL questionnaire. A study of 15 patients by Lye et al. showed that 93 percent of patients reported high levels of satisfaction and increased QOL following MMA surgery for OSA.

1.7 Long-term Success of MMA for OSA

Surgical relapse is a known concern with any surgery involving the maxilla and mandible, especially with mandibular advancements exceeding 8.5 mm. Despite this, data presently available for evaluation of the long-term efficacy of MMA as treatment for OSA is promising. The systematic review by Holty et al. found an 86% success rate following a minimum two-year follow-up period. Forty patients were followed by Li et al. for 50.7 months and there was found to be a 90% success rate after this period. Boyd et al. found an AHI reduction of 76.9% after over 6 years following surgery, despite significant weight gain in some of the
study participants. Substantial and sustained reductions in AHI, ESS, and improved QOL, as measured with various questionnaires, were shown to occur.

A study by Jaspers et al.\textsuperscript{91} was the first to follow patients who underwent MMA surgery for the treatment of OSA over an 8-year period. Only one patient was found to have surgical relapse after 8 years and this patient continued to have an ESS that was low, despite having an increase in AHI from 2 to 43. All other patients had stable results for ESS and AHI over an 8-year period, although two patients had an AHI over 5, which is defined as OSA. The main criticism of this study is the small sample size (n= 6).

In addition, a recent study by Vigneron et al.\textsuperscript{92} included 88 patients and showed the long-term (mean follow-up 12.5 years) success rate to be 28% for the entire group. AHI was reduced post-operatively between 50 and 80% for all patients, except in one instance. It was noted, however, that the success rate was 100% for patients less than 45 years of age, with BMI <25, initial AHI <45, and who underwent pre-operative orthodontics. The authors concluded that certain groups of patients have a much higher success rate when having MMA surgery for treatment of OSA. It was also recommended that pre-operative orthodontics should be considered in order to maximize the aesthetic result.

1.8 Morbidity and Mortality of MMA Surgery

As with any surgical procedure, success rates depend on both the ability of the surgery to deliver a specific therapeutic result, but also the mitigation of any inherent risks with the procedure itself. A retrospective review of 655
consecutive orthognathic surgeries over a 13-year period was conducted by Panula et al.\textsuperscript{93} for surgical complications. Mild neurosensory deficit in the areas innervated by the inferior alveolar nerve was the most common complication, occurring in 32% of patients. Severe damage to the inferior alveolar nerve resulting in disturbing defects to the patient resulted in 3% of cases. Poor intra-operative splits of the mandible occurred in 12 patients, and a total of 10 teeth were damaged during the osteotomy procedure. Plates and screws loosened in a minority of patients, and only 16 required a second surgical procedure to remedy this. Serious complications were found to be exceedingly rare, with the most common of these including major bleeding from the maxillary artery necessitating blood transfusion (1 patient). No fatalities, no loss of bony segments, and no tooth loss resulted from any of the cases observed.

The safety of MMA surgery for OSA was supported in a study by Giarda et al.\textsuperscript{94} The rate of major surgical complication (ischaemic necrosis, cardiac complication) occurred at a rate of 1%. Minor complications (mandibular relapse, facial paraesthesia, and temporomandibular joint dysfunction) occurred in 3.1% of patients. Four patients out of 16 reported paraesthesia immediately after surgery, however, this resolved completely in all patients at follow-up.

Although the majority of patients undergoing MMA surgery for orthodontic and orthognathic reasons are young and healthy, patients undergoing MMA surgery for OSA are typically older, obese, have a higher American Society of Anesthesiologists classification, and are likely have other significant comorbidities. In a study by Passeri et al.\textsuperscript{95}, the absolute risk of a complication was 3.9 for
patients undergoing MMA surgery for OSA, compared to 1.3 for patients undergoing MMA surgery for orthognathic reasons. In both cases, however, overall mortality was 0 and overall risk was assessed to be relatively low.

A systematic review and meta-analysis was performed by Holty et al.\(^{89}\) to examine the morbidity and mortality of MMA surgery for OSA. Twenty-two studies were evaluated for rate of complications. It was found that major complications resulted in only 1 percent of cases, with minor complications including minor haemorrhage, local infections treated successfully with antibiotics, neurosensory deficits resolving within 12 months, malocclusion, and worsening of facial appearance occurring in a minority of patients.

1.9 Aesthetic Concerns With MMA Surgery for OSA

In order to maximize the therapeutic benefits of MMA surgery in the treatment of OSA, large advancements are often the surgical goal. The telegnathic nature of this type of MMA surgery involves the movement of the hard tissues and their soft tissue counterparts to great degrees. Advancements of 10-12 mm of the maxillomandibular complex are not uncommon.\(^{96}\) In this manner, advancement of the maxilla, mandible, and genial tuberosities allows for maximal expansion of the pharyngeal space. In addition, the soft tissues of the face will also be advanced along with the hard tissues of the maxillomandibular complex. It has been found that when alar base cinch and V-Y closure are used as surgical techniques, the soft tissues of the upper lip reflect maxillary hard tissue movements in a ratio of 0.9:1.\(^{97}\) Mandibular and genial advancement surgeries result in a hard-to-soft
tissue anterior movement ratio of 1:1.\(^9^8\) As the primary goal of the surgery is not esthetic, but rather to provide surgical movements to the maximum degrees while remaining within the framework of surgical stability, it is understandable that both patients and health care providers will have concerns about the final aesthetic result.

Some general concerns with MMA surgery and its effect on aesthetics have been identified. Excessive maxillomandibular protrusion, bulging upper lip, prominent upper incisors, widening of the alar base, and increased nasal supratip break have been identified as some of these aesthetic concerns.\(^9^9\) Despite these aesthetic concerns, significant improvements in patients’ profiles were noted when they underwent MMA for orthognathic reasons. In a study that evaluated pre- and post-surgical silhouettes of patients who had MMA surgery for orthognathic reasons, favourable aesthetic outcomes were noted.\(^1^0^0\) Interestingly, the only silhouette that was ranked unfavourably following surgery was a patient who had a 10 mm advancement of the mandible; this magnitude of advancement is more consistent with surgical movements that may be seen in OSA patients.

As stated previously, characteristics of patients undergoing MMA surgery for orthognathic reasons may be significantly different than those of patients undergoing MMA surgery for OSA. Orthognathic patients are typically young, female, and have normal BMI, whereas OSA patients are more likely to be older, male, and obese.\(^9^8\) Similarly, although mandibular and/or maxillary retrognathia are common pathophysiologica l findings in patients with OSA, these patients may not have an obviously evident dentofacial deformity. Few studies are available that
examine the resulting aesthetics of the large anterior movements of the maxillomandibular complex in OSA patients treated with MMA surgery.

Li et al.\textsuperscript{101} evaluated patient perceptions of post-surgical facial appearance following MMA surgery for the treatment of OSA. Despite the final surgical result showing significant radiographic maxillomandibular protrusion when compared to aesthetic norms, 90 percent of patients reported positive or neutral changes in their facial appearance. The authors noted that patients with the greatest risk of adverse facial changes have thin facial soft tissues and include younger, non-obese patients.

A study involving a Chinese population of patients by Liu et al.\textsuperscript{96} examined both patient and layperson assessment of facial aesthetic outcomes following MMA surgery for treatment of OSA. In this study, the success rate in treatment of OSA was determined to be 83 percent. All patients except one felt that the aesthetic changes in their post-operative profile were positive. When a panel of 100 laypersons was asked to rate pre- and post-operative photographs, it was found that post-surgical profiles were significantly more attractive in 11 of 12 patients, despite being outside of aesthetic norms. Some concerns with this study include small sample size, unequal numbers of male and female patients (11 male, 1 female), and non-standardized and unaltered photographs that may result in the introduction of confounding variables.

A study by Cohen-Levy et al.\textsuperscript{102} examined the aesthetics of pre- and post-surgical profiles in fifteen adult male patients who underwent MMA surgery to treat OSA. Success rate was defined as an AHI less than 15 with no surgical
complications, and was found to be 80%. All patients reported reduced symptoms of OSA, and all but one were satisfied with the aesthetic outcome. When assessed by three panels consisting of laypersons, art students, and orthodontists, 85 percent of post-operative profiles were thought to be more aesthetic than the pre-operative counterparts, with no differences between the three panels. The post-operative profiles that were rated lower than the corresponding pre-operative profile had more protrusive lips, acute naso-labial angle, and nose that appeared small post-surgically. These patients were also brachyfacial pre-treatment. It was noted that retrusive lips and an obtuse naso-labial angle pre-surgically might be predictors of more favourable surgical outcomes. Orthodontic preparation to retrace the incisors and upper lip may be indicated to adjust these factors in patients pre-surgically.

1.10 Rationale for the Study

MMA has been shown to be the most successful surgical treatment for OSA when objective measures are evaluated. In a study of 22 OSA patients treated with MMA surgery by a single surgeon at the University of Western Ontario, significant AHI and ESS reductions were achieved. In this study, 86% of patients experienced successful treatment, which was defined as a 50% reduction in pre-operative AHI, or AHI reduced below 15. The same patients were examined in a subsequent study to evaluate subjective outcomes of MMA surgery for OSA. From these data, 86% of patients indicated that their symptoms had improved
following surgery, and 86% of patients indicated an improvement in overall quality of life.

Aesthetic changes following MMA surgery have the ability to greatly impact the confidence and self-esteem of a patient. In traditional orthognathic surgery, surgical movements of the maxilla and mandible are often planned in order to provide the most balanced facial components possible. It is recognized that facial balance is a primary factor in assessing the attractiveness of a face. \(^{100}\) In most cases, patients undergoing orthognathic surgery are highly satisfied with the aesthetic changes that are achieved. \(^{105}\) In the case of patients being treated for OSA, the advancements of the maxilla and mandible are often maximized in order to provide a therapeutic outcome. In this manner, the primary goal of the surgery is to reduce or eliminate the signs and symptoms of a disorder, and aesthetics may be a secondary consideration. In order to maximize patient satisfaction, it is important to achieve a balance between the therapeutic goals and the aesthetic outcomes.

Some studies have assessed patient perception of the facial aesthetic outcomes following MMA surgery for OSA with positive results. Islam \textit{et al.} \(^{106}\) found that 69% of patients felt positive or neutral toward the facial changes that resulted after surgery. Another study \(^{101}\) reported 90% patient satisfaction in aesthetic outcome. In a study by Emanuele, \(^{104}\) only 9% of patients felt that their attractiveness had decreased following MMA surgery.

Although multiple studies exist evaluating self-reported aesthetic changes following MMA surgery for OSA, very few studies exist that evaluate the
perceptions of external sources when considering pre- and post-surgical aesthetics of these patients. For this reason, research is required in order to properly assess the aesthetic results of the large advancements that comprise MMA surgery for OSA.

1.11 Purpose of the Study

There is a definite need to assess the aesthetic outcomes of MMA surgery in the treatment of OSA. The objective of this study is to evaluate facial profile changes in patients treated for OSA with MMA surgery utilizing panels of oral and maxillofacial surgeons, orthodontists, and laypeople. In addition, any potential differences or similarities in the way these groups of evaluators view pre-surgical and post-surgical aesthetics will be examined.
Chapter 2
Materials and Methods

2.1 Ethics Approval

Ethics approval was obtained from the Health Sciences Research Ethics Board (HSREB) at the University of Western Ontario (approval number 108450, Appendix I). Approval was also obtained from the Lawson Institute at the London Health Sciences Centre (LHSC), which encompasses the London Health Sciences Centre Division of Oral and Maxillofacial Surgery at University Hospital, a teaching hospital affiliated with the University of Western Ontario.

2.2 Patient Data

This study was a retrospective cohort analysis. A chart review was conducted of consecutive patients undergoing MMA surgery for OSA by a single oral and maxillofacial surgeon at LHSC between the years 2002 and 2016. The following inclusion criteria were used:

1) A diagnosis of OSA through an overnight sleep-study (PSG) by a sleep physician. A pre-surgical AHI greater than 5 was required to be diagnosed with OSA.
2) Over 18 years of age.
3) Pre- and post-surgical PSG data available, including AHI.
4) Good quality pre- and post-surgical profile photographs whereby silhouettes could be fabricated.
From the charts obtained during this period, exclusions were made based on the following criteria:

1) Patients with incomplete chart data for pre- and post-surgical PSG, including AHI, and/or photographs.
2) Patients under the age of 18.
3) Patients with craniofacial abnormalities and syndromes.
4) Poor photograph quality (presence of facial hair, blurred image) that made the fabrication of an accurate silhouette not possible.

The chart review resulted in an initial sample size of 44 patients (17 females, 27 males), collected from the hospital records of one oral and maxillofacial surgeon (Dr. M. Shimizu) at the Department of Oral and Maxillofacial Surgery, London Health Sciences Centre. Of these charts, 4 were excluded due to a pre-surgical AHI <5, which precluded a diagnosis of OSA. Three of the initial patient pool were under 18 years of age, 5 patients had pre- and/or post-surgical photographs that we were unable to reliably convert to silhouette form, and 7 patients did not have post-surgical sleep-study data (AHI) available. In addition, 3 patients were currently undergoing treatment and therefore had incomplete data. After the application of the inclusion and exclusion criteria, 22 patients were included in this study (10 females, 12 males).
2.3 Surgical Technique

Prior to MMA surgery, pre-surgical lateral cephalograms were evaluated for each patient using the Delaire analysis\textsuperscript{107} in order to plan the surgery and establish the amount and direction of each surgical movement. The Delaire analysis determined the ideal position of the maxilla and mandible, and then a clinical judgement was used in order to assess the maximum advancement without causing significant negative aesthetic effects. In general, advancements of 10 mm are the surgical goal; however, this is not always attainable if the Delaire analysis or clinical evaluation caution against this degree of movement. A surgical splint was fabricated prior to surgery in order to allow precise surgical movements intraoperatively.

Maxillary surgery consisted of a Le Fort I advancement\textsuperscript{72} with or without an impaction (if required). Mandibular surgery consisted of advancement via bilateral sagittal split ramus osteotomy (BSSO).\textsuperscript{73} The mandibular condyles were left in a seated position (proximal segment) and the body of the mandible, including the alveolar bone and teeth, was advanced (distal segment). Rigid fixation was used in the form of bicortical screws and/or miniplates and screws, using the surgical splint as a guide. A genioplasty was performed where indicated to provide additional advancement of the genial tubercles and corresponding musculature (genioglossus, geniohyoid, mylohyoid, and digastric).
2.4 Photograph-to-Silhouette Conversion

Pre- and post-surgical photographs of patient profiles (right side) were used to create silhouettes. All photographs were taken with patients situated in natural head position in maximum intercuspation. Profile photographs were imported as high-resolution files into Photoshop CC 2017 (Adobe, California). In a few circumstances, patients were hyperextending their neck and when this was determined to be the case, profiles were rotated to provide consistency between pre- and post-surgical images; superimpositions of pre- and post-surgical images were prepared, using soft tissue glabella and nasion as a guide. Outlines were generated and profiles were filled in with black, leaving the background white (Figure 2, Appendix II). Silhouettes were cropped at soft tissue glabella in order to reduce the impact of inconsistencies due to variation in hairstyles, which resulted in discrepancies between some silhouettes.
2.5 Development of the Questionnaire

Qualtrics® (Qualtrics, 2017, Provo, UT) web based software was used to fabricate a questionnaire in which evaluators could provide an aesthetic rating for each silhouette. An informed consent information package was included for evaluators describing the study in detail (Appendix III). Initial questions asked about evaluator demographic information, including gender (female or male), age (18-29, 30-39, 40-49, 50+), and profession (oral and maxillofacial surgeon, orthodontist, other (layperson)) (Figure 3). A question about geographic location was included in order to confirm that only Canadian respondents were included.
Questionnaire slides were fabricated, with each silhouette presented on a separate slide. Evaluators were introduced to each silhouette by an initial sentence related to the patient’s gender (female or male) and description of his or her approximate age (Figure 4, Appendix IV). Age categories designated patients as being young adult (18 - 35 years), middle-aged adult (35 - 49 years), or older adult (50+ years). From this descriptive designation, there were silhouettes of 5 young adults, 9 middle-aged adults, and 8 older adults included.

**Figure 3.** Qualtrics® software was used to fabricate a survey for this study. Initial questions asked evaluators to provide information on gender, age, and profession. A question was included about geographic location to confirm that only Canadian responses were included.
Figure 4. Qualtrics® software was used to present 22 pairs of silhouettes plus 6 duplicate silhouettes to panels of oral and maxillofacial surgeons, orthodontists, and laypeople. Evaluators were asked to use the sliding visual-analogue scale in order to provide an evaluation regarding the facial balance (attractiveness) for each silhouette. Pre- and post-surgical silhouettes were presented in random order, with duplicate silhouettes also randomly inserted into the questionnaire.

Evaluators were asked to rate each silhouette based on attractiveness, paying particular attention to facial balance. A sliding visual analogue scale (VAS) was provided at the bottom of each silhouette in order for evaluators to indicate their preference as to the attractiveness of the silhouette. The VAS ranged from “0.0” to “10.0”. A score of “0.0” indicated that the evaluator thought that the
silhouette was “very unattractive”, whereas a score of “10.0” indicated a “very attractive” silhouette.

The 22 pre-treatment and 22 post-treatment patient silhouettes were placed in random order using a random number generator (random.org). In addition, 6 of the 44 silhouettes were randomly inserted into the questionnaire in duplicate to test intra-observer reliability. Evaluators were unaware that duplicate silhouettes were included. The total number of silhouettes rated by each evaluator was 50.

2.6 Content Validation

Prior to survey distribution, eight graduate students in the orthodontic residency program at the University of Western Ontario were asked to complete the survey and provide feedback. These results were used to assess survey design and to ensure that questionnaire data were being tracked accurately. Data collection methods were assessed for completeness and reliability. Any feedback that was offered regarding survey design was noted. The eight surveys provided for content validation were not included in the final results.

2.7 Recruitment of Evaluators

Laypeople were recruited from current patients and parents/guardians of patients who were over the age of 18 and presently undergoing orthodontic treatment at the University of Western Ontario. Consecutive sampling was used to obtain a sample size of 70 completed surveys. Survey access was provided to
laypeople via a tablet at the Graduate Orthodontic Clinic or by email if they preferred to receive the survey via a web-link.

Oral and maxillofacial surgeons were recruited through the email list of current members of the Canadian Association of Oral and Maxillofacial Surgeons (CAOMS). Three hundred and forty-six active and retired members of the CAOMS were sent an invitation to participate in the survey, along with a web link to access it, if desired. Orthodontists were recruited through the email list of the American Association of Orthodontists (AAO). The survey was distributed to a sample of five hundred and sixty-one active and retired Canadian members. Two reminder emails were sent one week apart after the initial invitation to members of the CAOMS and AAO. Survey collection was ongoing for the surgeon and orthodontist groups until the point of time in which no new surveys were obtained; this resulted in a collection period of 6 weeks.

Although the survey was done in an anonymous manner, IP addresses were collected and used in conjunction with demographic information in order to determine if duplicate results were obtained. This method was used in order to assist in controlling for any potential individuals repeating the survey.

2.8 Statistical Analysis

Data were downloaded from Qualtrics® software into Microsoft Excel® spreadsheet format in order to manipulate the randomized data into an orderly fashion. Once the data sets were ordered by patient number, they were transferred to SPSS statistical package version 23 (SPSS Inc., Chicago, IL) and statistical
analysis was performed. Data were evaluated for normalcy and any outliers were identified visually using a Box Plot. Most data sets were normally distributed. For any data sets with outliers, it was confirmed that the outliers did not have a significant effect on the data analysis. In addition, Levene's test for homogeneity of variance was performed in order to assess the data (p ≤ 0.05).

A paired two-tailed t-test was used in order to compare the mean pre-surgical with the mean post-surgical aesthetic scores for each patient (p ≤ 0.05). This was assessed for each of the panels of evaluators separately (surgeons, orthodontists, laypeople), as well as the evaluators combined. A One-Way ANOVA was used to compare the mean difference between pre- and post-surgical evaluator scores between each group (surgeons, orthodontists, laypeople) (p ≤ 0.05). Post-hoc Tukey analysis was used in order to assess differences between evaluator groups for those groups passing Levene's test. For any patients with heterogeneity of data variance, a Welch's ANOVA and Games-Howell post hoc analysis was performed.

Intra-rater reliability was assessed for six pairs of duplicate silhouettes. Intraclass Correlation Coefficient estimates and their 95% confidence intervals were calculated in order to assess correlations between the ratings given for silhouette “A” as compared to silhouette “B”. These differences were examined between group ratings given by surgeons, orthodontists, and laypeople.
Chapter 3
Results

3.1 Patient Demographics

A retrospective chart analysis of consecutive patients undergoing MMA surgery for OSA between 2002 and 2016 was performed. Twenty-two patients met the inclusion criteria previously described. Of the 22 patients included in this study, there were 10 females and 12 males (Table 2). The mean age at time of surgery was 44 years and 6 months, and ranged from 19 years and 2 months to 71 years and 5 months. The mean pre-surgical ESS was 11.7 (range 3 - 19), and mean post-surgical ESS was 7.3 (range 1-14). The mean pre-surgical AHI was 47.4 (range 9.0 - 120.2), and mean post-surgical AHI was 9.4 (range 0.2 - 37.8). Nineteen patients (86%) underwent concurrent orthodontic treatment. All patients underwent surgical procedures consisting of BSSO with mandibular advancement and Le Fort I with maxillary advancement. Of the 22 patients, 14 (64%) also had the genial tubercles advanced via a functional genioplasty during surgery.
Table 2. Patient demographic information including gender, age at time of surgery, pre- and post-surgical Epworth Sleepiness Scale (ESS), and pre- and post-surgical Apnea-Hypopnea Index (AHI) are shown. Type of surgical treatment (Bilateral Sagittal Split Osteotomy, Le Fort I, Genioplasty) and presence of concurrent orthodontic treatment are also indicated for each patient.

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<td>30.9</td>
<td>3.3</td>
<td>Y</td>
<td>Y</td>
<td>N</td>
<td>Y</td>
</tr>
<tr>
<td>22</td>
<td>M</td>
<td>42,11</td>
<td>4</td>
<td>N/A</td>
<td>46</td>
<td>1.6</td>
<td>Y</td>
<td>Y</td>
<td>N</td>
<td>Y</td>
</tr>
<tr>
<td>Mean</td>
<td>-</td>
<td>44.6</td>
<td>11.7</td>
<td>7.3</td>
<td>47.2</td>
<td>9.4</td>
<td>-</td>
<td>-</td>
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</tr>
</tbody>
</table>

The linear measurements for surgical movements were obtained from patient charts and are shown in Table 3 for each patient. A BSSO was performed on all patients, with a mean advancement of 10.5 mm (range 7 - 15 mm). All 22 patients also had a Le Fort I maxillary osteotomy with mean advancement of 5.7 mm (range 3 - 10 mm). For the 14 patients undergoing a functional genioplasty procedure, a mean advancement of 6.8 mm (range 3 - 9 mm) was performed.
Table 3. Orthognathic surgical movements, in millimeters, are shown for 22 patients. Surgery involved the advancement of the mandible (BSSO) and maxilla (Le Fort I) for all patients. Some patients underwent concurrent advancement of the genial tubercles (Genioplasty).

<table>
<thead>
<tr>
<th>Patient</th>
<th>BSSO</th>
<th>Le Fort I</th>
<th>Genioplasty</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>9</td>
<td>7</td>
<td>N/A</td>
</tr>
<tr>
<td>2</td>
<td>9</td>
<td>5</td>
<td>N/A</td>
</tr>
<tr>
<td>3</td>
<td>11</td>
<td>10</td>
<td>6</td>
</tr>
<tr>
<td>4</td>
<td>15</td>
<td>7</td>
<td>3</td>
</tr>
<tr>
<td>5</td>
<td>11</td>
<td>3</td>
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<td>15</td>
<td>6</td>
<td>N/A</td>
</tr>
<tr>
<td>9</td>
<td>10</td>
<td>4</td>
<td>N/A</td>
</tr>
<tr>
<td>10</td>
<td>8</td>
<td>8</td>
<td>6</td>
</tr>
<tr>
<td>11</td>
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<tr>
<td>12</td>
<td>9</td>
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<tr>
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<td>9</td>
<td>3</td>
<td>N/A</td>
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<tr>
<td>16</td>
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<td>10</td>
<td>7</td>
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<td>3</td>
<td>6</td>
</tr>
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<td>19</td>
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<td>5</td>
<td>6</td>
</tr>
<tr>
<td>21</td>
<td>10</td>
<td>3</td>
<td>N/A</td>
</tr>
<tr>
<td>22</td>
<td>11</td>
<td>8</td>
<td>N/A</td>
</tr>
<tr>
<td>Average</td>
<td>10.6</td>
<td>5.7</td>
<td>6.8</td>
</tr>
</tbody>
</table>

3.2 Evaluator Demographics

Surveys were distributed via current email lists to oral and maxillofacial surgeons and orthodontists via the CAOMS and AAO respectively. Only surveys that were returned with every question answered in full were included in the results. Of 346 active and retired Canadian surgeons that received an email invitation to participate in the survey, 57 surveys were returned...
(Table 4). Of the 57 surveys returned, only 48 fully completed surveys were obtained, giving a response rate of 13.9%. An email invitation was sent to 561 active and retired Canadian orthodontists. Of the 60 surveys that were returned, only 58 were completed in full and included in the results. The orthodontist response rate was 10.3%.

Table 4. Number of survey invitations distributed to oral and maxillofacial surgeons (OMFS) and orthodontists (ORTHO). Number of total responses and complete responses are shown. Any incomplete surveys were discarded from the final data.

<table>
<thead>
<tr>
<th>Group</th>
<th>Email Requests</th>
<th>Number of Responses</th>
<th>Complete Surveys</th>
<th>Response Rate</th>
</tr>
</thead>
<tbody>
<tr>
<td>OMFS</td>
<td>346</td>
<td>57</td>
<td>48</td>
<td>13.90%</td>
</tr>
<tr>
<td>ORTHO</td>
<td>561</td>
<td>60</td>
<td>58</td>
<td>10.30%</td>
</tr>
</tbody>
</table>

Laypeople over the age of 18 were surveyed from patients and parents/guardians in the University of Western Ontario Graduate Orthodontic Clinic. Consecutive eligible evaluators were invited to participate. The number of individuals declining to participate is unknown, but was very low. Overall, 87 laypeople agreed to participate in the survey; however, 17 surveys were discarded due to incomplete data acquisition.

Demographics for evaluator gender are shown in Figure 5. Completed surveys were returned by 4 female and 44 male surgeons, 13 female and 45 male orthodontists, and 41 female and 29 male laypeople.
Figure 5. Distribution of evaluator gender is shown for oral and maxillofacial surgeons (OMFS), orthodontists (ORTHO), and laypeople (LAY).

Distributions for evaluator age are shown in Figure 6. There were 0 surgeons, 3 orthodontists, and 9 laypeople in the 18 - 29 age category. Of evaluators between the ages of 30 - 39, there were 7 surgeons, 20 orthodontists, and 14 laypeople. Nine surgeons, 6 orthodontists, and 28 laypeople responded to the survey between the ages of 40 - 49. Of evaluators age 50 and above, there were 32 surgeons, 29 orthodontists, and 19 laypeople.
3.3 Evaluation of Pre- and Post-Surgical Aesthetic Scores

For each of the 22 surgical patients, pre-surgical aesthetic scores were compared to post-surgical aesthetic scores for all evaluators combined (Figure 7). Of the 22 patients, 19 had a statistically significant increase (p < 0.001) in post-surgical aesthetic score as compared to the pre-surgical evaluation. One patient had a statistically significant decrease in aesthetic score (patient 12, p < 0.001).
Figure 7. Mean pre-surgical and post-surgical aesthetic score with standard deviations is shown for 22 patients for all evaluators combined. Nineteen of 22 patients had a statistically significant increase in aesthetic score. Differences were not significant for patients 8 and 10 (NS indicates p > 0.05).

Surgeon evaluators rated 19 of 22 patients as having a statistically significant increase in post-surgical aesthetic scores (Figure 8, p < 0.001). One patient of 22 (patient 12) was given more negative post-surgical aesthetic scores that, when compared to the pre-surgical scores, was statistically significant (p < 0.001).
**Figure 8.** Mean surgeon evaluator pre-surgical and post-surgical aesthetic score with standard deviations for 22 patients. Nineteen of 22 patients had a statistically significant increase in aesthetic scores. Differences were not significant for patients 8 and 10 (NS indicates \( p > 0.05 \)).

Pre- and post-surgical aesthetic ratings for 22 patients as given by orthodontists were evaluated (Figure 9). Eighteen of 22 patients were given post-surgical scores that were statistically higher than the pre-surgical scores (\( p < 0.05 \)). Only two patients were evaluated as having more negative post-surgical aesthetic scores as compared to the pre-surgical rating. In both of these cases, the change in the aesthetic rating was statistically significant (patient 10, \( p = 0.009 \); patient 12, \( p = 0.019 \)).
Figure 9. Mean orthodontist evaluator pre-surgical and post-surgical aesthetic score with standard deviations for 22 patients. Eighteen of 22 patients had a statistically significant increase in aesthetic scores. Differences were not significant for patients 8 and 20 (NS indicates p > 0.05).

Pre-surgical and post-surgical aesthetic scores were examined for laypeople evaluators (Figure 10). Of 22 patients, 17 had a statistically significant increase in post-surgical aesthetic scores when compared to pre-surgical scores (p < 0.01). Two patients had a statistically significant decrease in post-surgical aesthetic scores (patient 8, p = 0.027; patient 12, p = 0.020).
There was a mean overall increase in post-surgical aesthetic scores of 1.9 (+1.9) for all evaluators combined. When evaluator groups were examined separately, surgeon and orthodontist groups were found to each have mean increases of 2.1 (+1.7). This compares to a mean post-surgical increase of 1.6 (+1.9) for laypeople evaluators. A significant difference was found between surgeon and laypeople (p < 0.001), and orthodontist and laypeople (p < 0.001) evaluators, but not between surgeon and orthodontist evaluators (p > 0.05).
3.4 Positive or Neutral Change in Aesthetic Scores

The number of evaluators rating each patient with a positive or neutral change for aesthetic scores was examined for the evaluators as a whole, and for each subgroup of evaluators (Table 5). The average standard deviation for all mean pre- and post-surgical differences was determined to be 1.9. A pre-surgical silhouette that was given a rating greater than 1.9 points higher than the post-surgical score was determined to be a negative aesthetic change, which is consistent with one standard deviation. Overall, 95 percent of evaluators felt that the post-surgical silhouette was a positive or neutral change as compared to the pre-surgical silhouette. Ninety-five percent of surgeon and orthodontist evaluators rated the post-surgical silhouette as positive or neutral. For laypeople evaluators, 94 percent felt that the post-surgical silhouette was positive or neutral when compared to the pre-surgical silhouette.
Table 5. Percentage of evaluators rating the post-surgical silhouette as neutral or higher than the pre-surgical silhouette is shown for 22 patients. The average standard deviation for all mean pre- and post-surgical differences was determined to be 1.9. A post-surgical silhouette was deemed to be negative as compared to the pre-surgical counterpart if the post-surgical silhouette was scored lower than the pre-surgical silhouette by ≥ 1.9 points.

<table>
<thead>
<tr>
<th>Patient</th>
<th>OMFS</th>
<th>ORTHO</th>
<th>LAY</th>
<th>OVERALL</th>
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</thead>
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<td>1</td>
<td>98</td>
<td>96</td>
<td>89</td>
<td>94</td>
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<tr>
<td>Total</td>
<td>95</td>
<td>95</td>
<td>94</td>
<td>95</td>
</tr>
</tbody>
</table>

3.5 Comparison of Surgeons, Orthodontists, and Laypeople Evaluations

Data were analyzed in order to determine the presence of any differences between mean aesthetic score changes given by oral and maxillofacial surgeon,
orthodontist and laypeople evaluators (Figure 11). Of 22 patients, 12 were shown
to be statistically different between groups (p < 0.05). A significant difference was
found for surgeon and laypeople evaluators between the mean aesthetic score
changes for patient 1 (p = 0.032), 3 (p = 0.007), 4 (p = 0.027), 7 (p = 0.004), 11
(p = 0.010), 14 (p = 0.048), 16 (p < 0.001), 17 (p = 0.013), and 21 (p = 0.008).
Orthodontists and laypeople ratings showed a significant difference for patient 3
(p = 0.002), 9 (p < 0.001), 10 (p = 0.010), 15 (p = 0.049), 16 (p < 0.001), 17
(p = 0.040), and 21 (p = 0.002). No significant differences were found between the
ratings given between surgeons and orthodontists for any of the patients
(p > 0.05). No significant difference (p > 0.05) was found between any of the
evaluator groups for patient 2, 5, 6, 8, 12, 13, 18, 19, 20, and 22.
Figure 11. Mean aesthetic score change is shown with standard deviations for oral and maxillofacial surgeons (OMFS), orthodontists (ORTHO), and laypeople (LAY). Significant differences between OMFS and LAY are indicated (X; p < 0.05). Significant differences between ORTHO and LAY are indicated (Y; p < 0.05). No significant differences were found between mean ratings of OMFS and ORTHO for any of the 22 patients (p > 0.05).
3.6 Intra-rater Reliability

Six duplicate silhouettes were included in the survey in order to evaluate intra-rater reliability (IRR). Intraclass correlation coefficients (ICC) and their 95% confidence intervals were calculated for each of the silhouette pairs, when comparing the rating for silhouette “A” to silhouette “B”. Values were assessed for each of the evaluator groups (Table 6). It was found that the overall intra-rater reliability was moderate, at 0.52, for the sample combined. The mean intra-rater reliability was 0.43, 0.55, and 0.59 for the surgeon, orthodontist, and laypeople groups respectively.

**Table 6.** Intra-rater reliability is shown for oral and maxillofacial surgeons (OMFS), orthodontists (ORTHO), and laypeople (LAY). Six pairs of duplicate silhouettes were included in the survey. Intraclass correlation coefficients and their 95% confidence intervals were calculated for each group of evaluators.

<table>
<thead>
<tr>
<th>Pair</th>
<th>OMFS</th>
<th>ORTHO</th>
<th>LAY</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>0.3 (0.02-0.54)</td>
<td>0.59 (0.40-0.74)</td>
<td>0.68 (0.53-0.79)</td>
</tr>
<tr>
<td>2</td>
<td>0.52 (0.28-0.70)</td>
<td>0.58 (0.38-0.73)</td>
<td>0.54 (0.35-0.69)</td>
</tr>
<tr>
<td>3</td>
<td>0.39 (0.12-0.60)</td>
<td>0.33 (0.08-0.55)</td>
<td>0.74 (0.61-0.83)</td>
</tr>
<tr>
<td>4</td>
<td>0.64 (0.44-0.78)</td>
<td>0.73 (0.58-0.83)</td>
<td>0.5 (0.31-0.66)</td>
</tr>
<tr>
<td>5</td>
<td>0.29 (0.01-0.53)</td>
<td>0.46 (0.23-0.64)</td>
<td>0.3 (0.07-0.5)</td>
</tr>
<tr>
<td>6</td>
<td>0.45 (0.20-0.65)</td>
<td>0.58 (0.38-0.73)</td>
<td>0.75 (0.62-0.84)</td>
</tr>
<tr>
<td>Mean</td>
<td>0.43</td>
<td>0.55</td>
<td>0.59</td>
</tr>
</tbody>
</table>
Obstructive sleep apnea (OSA) is a common sleep disorder that can have a significant detrimental effect on the quality of life of affected individuals. Although various therapies are available for the treatment of OSA, CPAP remains the gold-standard of non-surgical treatment. Successful surgical treatment for OSA has been achieved via maxillomandibular advancement (MMA) surgery, and remains a definitive treatment option for patients unable to tolerate CPAP therapy, or who do not want to be burdened with long-term wear. Due to the large advancements required for surgical correction of OSA through MMA surgery, concerns may be raised about the post-surgical aesthetic outcome. Despite the possibility of negative aesthetic sequelae as a result of MMA surgery, some studies have shown that patients are highly satisfied with their post-surgical aesthetic outcomes.

Limited data is presently available to evaluate aesthetic outcomes, especially as viewed by external evaluators, in patients following MMA surgery for OSA. This study was done to evaluate facial profile changes in patients treated for OSA with MMA surgery utilizing panels of oral and maxillofacial surgeons, orthodontists, and laypeople. A secondary purpose of this study was to determine if there was any difference in the way surgeons, orthodontists, and laypeople evaluated pre-surgical and post-surgical aesthetics.
4.1 Online Survey Design and Response Rate

Due to the relatively small numbers of surgeons and orthodontists, a Canada-wide survey was required. Previous studies have shown that mail-based surveys have a higher response rate (31.5%) than email-based surveys (20.7%). However, when time and financial constraints were accounted for, an online survey was determined to be the most efficient way to approach all Canadian active and retired surgeons and orthodontists. Of 346 surgeons who received an email invitation, 48 completed surveys were obtained, resulting in a response rate of 13.9%. An email was sent to 561 orthodontists inviting them to participate in the survey, and 58 completed surveys were returned, which corresponds to a response rate of 10.3%. These values are below expected response rates for an online survey, which have been reported as 20.7%.

Incomplete surveys were removed from the data evaluation. The majority of surveys that were removed included incomplete responses in the form of a low number of matched pairs. In this case, most incomplete surveys had either every-other silhouette skipped, or the second half of the survey left blank. The decision to remove these surveys was based on the fact that evaluators were thought to be poorly focused on providing accurate responses, or had difficulties in accessing and navigating the survey.

Previous studies have indicated that survey length is an important factor in motivating respondents to complete any given survey; up to 25% of respondents have indicated that excessive survey length is the main reason preventing them from completing a survey. This survey can be considered to be
of relatively longer length when compared to similar online surveys evaluating profile aesthetics.\textsuperscript{111,112} In examining our content validation, preliminary feedback from graduate orthodontic residents at the University of Western Ontario indicated that this was indeed a lengthy survey. Although it was identified as a survey of longer length, we maintained the same number of patient silhouettes. It was felt that all patients meeting our inclusion criteria should be evaluated in order to prevent the introduction of bias.

In addition to survey length, Sheehan \textit{et al.}\textsuperscript{113} noted a historical decline in survey response rates. Factors are generally related to respondents being busy or not realizing the benefit to participation. By approaching laypeople personally, and by allowing them to respond to the survey while waiting for their children to complete orthodontic treatment, a higher degree of participation was achieved in this evaluator category. When consideration is given to the removal of incomplete surveys and longer survey length of the present study, combined with a decrease in response interest to survey completion in general, the overall response rates for this survey are reasonable.

4.2 Validity of Silhouettes vs. Photographs

In order to satisfy concerns from the REB regarding patient privacy, patient photographs were converted to silhouettes for this study. Aesthetic benefits have been attributed to MMA surgery, and it has been noted that a “reverse face lift” effect can be achieved.\textsuperscript{114} Advancing the maxilla and mandible has been shown to provide a rejuvenation in soft tissue, increased soft tissue
support, improved soft tissue drape, and decrease in the appearance of wrinkles.\textsuperscript{114} For this reason, photographs would have perhaps provided visual cues that may have resulted in improved ratings for post-surgical results. In studies comparing the use of photographs and silhouettes in profile aesthetic evaluation for African American patients, it has been found that rater preferences are closer to established aesthetic norms for photographs, and flatter profiles with less lip projection were generally preferred in silhouettes.\textsuperscript{115}

Other studies have evaluated the use of photographs and silhouettes and found no difference in the way that they are rated, proving the applicability of both methods.\textsuperscript{116} It has been shown that silhouettes are processed like regular facial stimuli and that they allow for accurate estimations in both age and gender.\textsuperscript{117} Of importance, Tauk \textit{et al.}\textsuperscript{118} have shown that silhouettes of the entire face, and not just the lower third, must be used in order to provide an accurate assessment of attractiveness.

The main benefit in using silhouettes over photographs is that a standardized set of questions may be produced. Through the generation of silhouetted profiles, distractors such as differences in hair, makeup, and lighting may be eliminated. However, challenges of silhouettes include difficulties in tracing profiles when hairstyles and facial hair mask soft tissue contours, and for this reason, these specific patients were removed from this study.
4.3 Differences in Pre-surgical and Post-surgical Aesthetic Scores

Large advancements of the maxillomandibular complex are required in order to provide the most benefit for the treatment of OSA. For this reason, it is important to investigate any potential negative effects on facial aesthetics. For all evaluators combined, mean post-surgical aesthetic scores were significantly improved or neutral for 21 of 22 patients when compared to the mean pre-surgical aesthetic scores. This corresponds to a 95 percent positive or neutral change between pre-and post-surgical aesthetics. This is consistent with other studies that evaluated aesthetics of MMA surgery for OSA. A study by Cohen-Levy et al. found that post-surgical profiles were preferred by 85 percent of the combined panel of orthodontists, fine arts students, and laypeople. Likewise, a study by Liu et al. found that laypeople rated 11 of 12 patients’ post-operative aesthetic scores higher than pre-operative scores.

Patient number 12 was the only patient who had a significant decrease in post-surgical aesthetics. It is important to note that this patient’s mean pre-surgical aesthetic score (7.08) was the fifth highest total score of all 44 pre- and post-surgical scores combined. Since this patient was rated very high pre-surgically, it was perhaps difficult to improve on this score post-surgically. When evaluating this patient’s silhouette post-surgically, it was found that the genial advancement might have resulted in a profile that was assessed as having a chin projection that was too prominent. This finding is consistent with previous studies that determined excessively large changes in genial projection following MMA surgery lead to worsening in profile aesthetics. It has been found in previous
studies\textsuperscript{100,119} that a strong genial projection is especially not tolerated in female patients, and that convexity is seen as more attractive. As patient 12 was identified as female to the evaluators, they may have been less forgiving regarding this feature and the resultant straight profile.

When evaluator groups were assessed individually, it was found that surgeon, orthodontist, and laypeople evaluators felt that patient 12 had a significant decrease in profile aesthetics, as was noted in the combined scores. Patient 8 had a significant decrease in post-surgical aesthetic scores for laypeople only. The age of patient 8 was 71 years and 5 months, which was significantly higher than the next oldest patient (59 years, 11 months). Some evaluators may have found it difficult to provide an aesthetic score due to this patient being an extreme outlier when compared to the other patients in the survey. Differences exist in the aging face that may be perceived as unattractive when compared to some of the younger patients in this study.\textsuperscript{120} It has been noted that older faces show facial aesthetic units as more distinct components, whereas youthful faces have facial units that flow together and blend more evenly.\textsuperscript{121} This may especially account for the negative result for patient 8 in the laypeople group, as this population of evaluators may not have been cognizant to correct for these changes related to aging and not necessarily due to surgery.

Orthodontists evaluated patient 10 as having a significant negative change post-surgically. Again, patient 10 was given a very high pre-surgical aesthetic score by orthodontists (7.96). This was the highest score given to any of the 50
silhouettes presented to the orthodontic group. For this reason, an improvement in post-surgical aesthetics may have been difficult to obtain.

4.4 Self- versus Rater-Reported Aesthetic Evaluations

High patient satisfaction rates have been reported with facial aesthetics following MMA surgery for the treatment of OSA. Islam et al.\textsuperscript{106} determined that almost 70\% of patients felt that their facial aesthetics improved or stayed the same following MMA surgery. In a study by Li et al.\textsuperscript{101} in excess of 90\% of patients undergoing MMA surgery for OSA felt that the post-surgical facial changes were either neutral or positive, despite the significant maxillomandibular protrusion that resulted from the surgery. The current study involved many of the same patients who were previously evaluated for quality of life and subjective outcomes. In that study,\textsuperscript{104} almost 95\% of patients felt that there was a positive change, or no change, in their profile aesthetics.

Although patient satisfaction appears to be positive for post-operative aesthetics, limited information is available on the perception of aesthetic changes for external evaluator groups. Differences have been noted in the way that patients perceive their post-surgical aesthetics when compared to professionals and laypeople.\textsuperscript{122} In this study, significant differences existed between the mean aesthetic score changes of pre- and post-surgical silhouettes for professional and laypeople groups of evaluators. No differences were found between the ways that surgeons and orthodontists evaluated aesthetic score changes. However, when looking at specific patients, differences did exist between the ratings given by
laypeople and surgeons or orthodontists. Previous studies have examined the way that different groups evaluate pre- and post-surgical aesthetics with conflicting results. In some studies,\textsuperscript{123,124,125} no differences have been reported between professional and laypeople evaluator groups for aesthetic ratings of patients following MMA surgery for orthognathic reasons. Other studies have shown that differences do in fact exist in the way that professionals and laypeople evaluate facial aesthetics, with the greatest variability in perceived attractiveness within laypeople groups.\textsuperscript{100,126} Many studies have shown that specific training, such as dental or specialty programs, influences concepts of facial aesthetics.\textsuperscript{124,127}

The overall mean change in post-surgical aesthetic score was an increase of 1.9. When evaluator groups were examined separately, it was found that surgeon and orthodontist groups each reported a mean post-surgical increase of 2.1. This may be compared to a mean increase of 1.6 in laypeople. Only one study evaluating differences between professional groups and laypeople's assessment of MMA surgery aesthetic results within the context of OSA is available. Cohen-Levy et al.\textsuperscript{102} found that no significant difference existed between the aesthetic ratings given by orthodontists, fine arts students, and laypeople in the assessment of pre- and post-surgical profiles following MMA surgery for OSA.

Most of the previous studies evaluating MMA surgery aesthetics have reported agreement in the way that professional groups and laypeople evaluate pre- and post-surgical appearances following MMA surgery.\textsuperscript{100,128} However, it is important to note that these studies assessed MMA surgery for the achievement of ideal results, and did not involve advancements to the same degree as those used.
for OSA patients. Because the professional groups were not blinded to the objectives of the present study, it is possible that more protrusive outcomes were accepted, which were not viewed as positively by laypeople. For this reason, due to the large advancements involved in these types of surgeries, laypeople may not determine post-surgical results to be as positive as those seen by surgeons or orthodontists in the context of this study.

4.5 Clinical Recommendations for Informed Consent

Overall, multiple studies have shown that orthognathic surgery in general is both predictable and safe.\textsuperscript{89,93-95} When looking at MMA surgery in the treatment of OSA, positive outcomes are generally obtained with few incidences of complications.\textsuperscript{70,89} There are some differences in complication rates for patients undergoing MMA surgery for dentofacial deformities when compared to those patients undergoing MMA surgery for OSA. In the OSA population undergoing MMA surgery, major complication rates, most of which were cardiac in nature, were 1\% of all patients.\textsuperscript{89} In another study,\textsuperscript{95} infection was more common in OSA patients when compared to orthognathic patients. This is likely due to the fact that this patient group tends to be older, and have more comorbidities. It is important to note that although there was a higher incidence of surgical complications for cardiac reasons or infection in the OSA group, no long-term adverse outcomes resulted.\textsuperscript{95} Assessment of patients’ pre-surgical health and the risks of obesity, advanced age, and cardiovascular complications must be addressed along with potential surgical consequences.
Surgical relapse may be seen in patients undergoing mandibular advancement for orthognathic and OSA reasons. It has been found that ten to twenty percent surgical relapse may occur in up to fifteen percent of OSA patients after MMA surgery.\textsuperscript{89} It appears that this relapse, when it occurs, has no apparent effect on associated OSA symptoms and does not result in a worsening of the AHI.\textsuperscript{129} This is likely due to the overcorrection that is done at the time of surgery in anticipation of small amounts of relapse. Importantly, some studies have shown that surgical relapse is not associated with the degree of mandibular advancement.\textsuperscript{129,130} This is especially significant due to the fact that large surgical advancements are provided for maximal OSA treatment results.

The most common complication following MMA surgery for OSA or orthognathic reasons is paraesthesia involving the inferior alveolar nerve. It has been shown that there is a higher risk for paraesthesia in OSA patients,\textsuperscript{92} likely due to the large mandibular advancements that are performed. Fortunately, most cases of paraesthesia resolve in the first year following surgery.\textsuperscript{89}

Although aesthetic concerns may be raised by patients considering MMA surgery for the treatment of OSA, it does not appear that negative effects arise in the majority of patients. Excessive maxillomandibular protrusion, bulging upper lip, prominent upper incisors, widening of the alar base, and increased nasal supratip break have been identified as some of these aesthetic concerns.\textsuperscript{99} Fortunately, high post-surgical aesthetic satisfaction has been reported by patients.\textsuperscript{101,104,106} Li\textit{et al}.\textsuperscript{101} noted that potentially unfavourable outcomes should be discussed with patients who are young, non-obese, and present with pre-
surgical bimaxillary protrusion; these patients were shown to have the greatest risk of dissatisfaction with treatment when self-reported post-surgical aesthetic outcomes were assessed. When evaluating the aesthetic perception of MMA surgery for OSA patients, professional groups and laypeople report high degrees of positive post-surgical results. The present study confirms that post-surgical aesthetics are positive, even when advancing the maxillomandibular complex to great degrees in OSA patients.

4.6 Intra-rater Reliability

Six pairs of duplicate silhouettes were included in the survey in order to determine the reliability of aesthetic scores by evaluators. Some other studies, with similar design, provided additional silhouettes to familiarize the participants with the procedure. It was decided to start the questionnaire without any prompting on which type of silhouettes were considered “very attractive” or “very unattractive” in order to avoid biasing or confusing the evaluators.

Intraclass correlation coefficient estimates and their 95% confidence intervals were calculated to assess intra-rater reliability. It was found that the overall correlation was moderate (0.52), as compared to the scale provided by Rankin, and laypeople had the highest correlation (0.59) within ratings. Only a few studies report correlation coefficients, and, of the ones that do, a wide variety of results have been reported (range 0.46 – 0.78). In a study by Maple et al., it was found that laypeople had the highest correlation coefficients because
of the tendency for professionals to “overevaluate” profiles rather than providing an initial reaction.

Although correlation coefficients were moderate, it is felt that the similarity in trends between evaluator ratings shows a general consistency between these groups; in all three evaluator groups, patient 12 was shown to have a decrease in post-surgical aesthetic score. Other studies have shown that photographs rated by dental professionals can show statistically significant differences between first and second sets of ratings.\textsuperscript{133}

4.7 Differences Between Evaluator Genders and Ages

Certain demographic evaluator groups were underrepresented in this survey. The number of female surgeons and orthodontists practicing in Canada is significantly lower than the number of male counterparts, which is reflected in the number of completed surveys returned in these professional categories. Due to the small number of female evaluators in the surgeon and orthodontist categories, differences between the aesthetic score ratings by gender were not examined. In contrast, the number of female evaluators in the laypeople category was much higher than the number of male evaluators. This difference likely represents the higher representation of female caregivers bringing their children to appointments. Previous studies have shown that no significant differences exist between male and female evaluators when rating the attractiveness of facial profiles.\textsuperscript{111}
A similar concern with underrepresentation in specific age categories was realised. Most of the survey responses from surgeon and orthodontic evaluators were from respondents that identified in the 50+ age category. For this reason, statistical analysis was not used to evaluate differences between evaluator age groups as adequate statistical power would not be achieved. Differences in the way that aesthetics are evaluated, and treatment plans are formulated, have changed in the oral and maxillofacial surgery and orthodontic professions over the past decades. For this reason, it is possible that differences in aesthetic ratings may exist between younger and older professional evaluators when assessing surgical outcomes. When examining laypeople age categories, a higher number of evaluators were over the age of 40. Again, this is likely related to the demographic of caregivers seeking treatment for our orthodontic population at the clinic.

4.8 Strengths and Limitations of the Study

One of the main strengths of this study was that it had a good distribution of patient ages and genders. Most of the previous studies of similar design reported on older, male patients, which is consistent with the classical OSA patient. As it is now recognized that a target group for MMA surgery for the treatment of OSA is a younger population including both genders, data reporting for male and female patients of all ages is important.

The same difficulties existed for this study as with other online-based studies. For this type of data collection, evaluator participation is a problem. In
order to achieve a higher response rate, evaluator incentives may be considered in the future.

Variations existed in the methods used to recruit evaluators for participation in this study. Due to privacy regulations, direct contact with the professional evaluators was not permitted, and third-party members (CAOMS, AAO) implemented the email distribution. In contrast, laypeople, who were already familiar with the researchers, were approached through personal contact in the clinic at the University of Western Ontario. Although we do not feel that these differences in sampling methods introduced any significant bias, it may not be ruled out.

The variability in sampling methods does, however, highlight important differences in participant response rates. The numbers of laypeople declining to participate in the study was exceptionally low, whereas the response rate for surgeons and orthodontists was lower than expected for online surveys. It is thought that personally approaching potential evaluators played a significant role in response rate. Likewise, laypeople were approached while they were waiting for their children who were undergoing orthodontic treatment. As laypeople had time at that moment to complete the survey, and the professional groups may have received an email at an inconvenient time, this may have also affected response rates.

Survey results may require caution when interpreting layperson outcomes. Due to difficulties with sampling layperson populations, it was decided to consecutively sample individuals from the graduate orthodontic clinic at the
University of Western Ontario. As these layperson evaluators are involved in some facet of orthodontics prior to survey participation, innate bias may be present. These individuals may be predisposed to aesthetic ideals that may influence the results obtained in this survey.

Silhouettes were used to protect patient identity and eliminate distracting subjective variables; however, difficulties do exist with this approach. Since MMA surgery for the treatment of OSA has positive effects on soft tissue drape, photographs have a definite benefit. Evaluators may find a more positive post-surgical outcome following this type of surgery when photographs are available for assessment.

4.9 Future Research

Maxillomandibular advancement surgery has a number of benefits for patients that are not visible in a silhouetted profile. Especially for middle-aged patients, improved soft tissue drape and a diminished appearance of wrinkles are benefits that may be achieved following the surgery, and the surgery itself has been referred to as a “reverse face lift”. In the future, studies using photographs rather than silhouettes should be used in order to assess the post-surgical results obtained from this type of surgery, and the effect on aesthetics. Control of lighting, patient head position, hairstyles, and makeup application should be recognized.

From a cursory evaluation of the patient facial types in this study, it appears that pre-surgical Class II profiles that became less mandibular retrognathic following surgery were rated higher than initially straight or
prognathic profiles. For this reason, future research should be done to confirm these concepts. Larger numbers of patient groups could be used in order to divide pre-surgical profiles into categories by degree of convexity to assess surgical results by initial skeletal classification. Pre- and post-surgical cephalometric variables could also be correlated with aesthetic scores.

Patients of various genders and age categories could be assessed for post-operative aesthetics if a larger patient pool could be obtained. In this study, one patient, who was significantly older than the rest of the sample, consistently received post-operative scores that were lower than the pre-operative counterparts. It would be interesting to evaluate if this negative outcome was specifically related to this particular individual, or if a trend exists that may advise caution with large advancements in an aging population.

This study was limited by relatively small response rates for professional groups. In the future, it would be interesting to assess larger groups of evaluators such that differences in ratings by gender could be determined. Likewise, potential differences between the ways aesthetics are perceived by evaluators of different ages could be assessed.

4.10 Summary

Overall, it does not appear that MMA surgery for the treatment of OSA has a negative impact on facial profile aesthetics. Of the 22 patients evaluated, 21 had a significantly positive or neutral change in facial aesthetics when the data from all groups of evaluators was combined. The mean change in aesthetic scores
post-surgically was found to be +1.9 for all groups combined, and as high as +2.1 in the surgeon and orthodontist evaluator groups. There was a significant difference in the way that surgeons and orthodontists rated silhouettes as compared to laypeople for 12 of 22 patients. There were no significant differences between the way that surgeons and orthodontists rated the silhouettes.

It is important to assess the post-surgical aesthetic outcome of MMA surgery for the treatment of OSA due to the large surgical advancements that are produced. One of the considerations of this type of treatment should be to find a balance between therapeutic functional goals and ultimate aesthetics. It does not appear that the current surgical methods utilized for treating OSA with MMA surgery have a negative effect on post-surgical facial profile aesthetics.
Chapter 5
Conclusions

Research in the area of post-surgical aesthetics for patients undergoing maxillomandibular advancement (MMA) surgery for the treatment of obstructive sleep apnea (OSA) has been lacking. There is a definite need to assess the aesthetic outcomes for these patients. The objective of this study was to evaluate facial profile aesthetic changes in patients treated for OSA with MMA surgery utilizing panels of oral and maxillofacial surgeons, orthodontists, and laypeople. In addition, differences in the way these groups of evaluators view pre-surgical and post-surgical aesthetics was examined. The following conclusions were established:

1) In general, MMA surgery for the treatment of OSA does not have a negative impact on facial profile aesthetics.

2) All groups of evaluators (surgeons, orthodontists, and laypeople) felt that post-surgical aesthetic changes were positive in the majority of patients undergoing MMA surgery for the treatment of OSA.

3) Surgeons and orthodontists reported a higher positive post-surgical change than laypeople.
References


3. Franklin KA, Lindberg E. Obstructive sleep apnea is a common disorder in the population - a review on the epidemiology of sleep apnea. J Thorac Dis. 7:1311-1322.


Appendix I

Western University HSREB Approval

Western University Health Science Research Ethics Board
HSREB Amendment Approval Notice

Principal Investigator: Dr. Ali Tassi
Department & Institution: Schulich School of Medicine and Dentistry/Schulich School of Medicine & Dentistry, Western University

Review Type: Delegated
HSREB File Number: 108450
Study Title: Aesthetic Outcome of Maxillomandibular Advancement Surgery for the Treatment of Obstructive Sleep Apnea

HSREB Amendment Approval Date: July 12, 2017
HSREB Expiry Date: March 21, 2018

Documents Approved and/or Received for Information:

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<tr>
<th>Document Name</th>
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<tr>
<td>Revised Western University Protocol</td>
<td>Received June 20, 2017</td>
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The Western University Health Science Research Ethics Board (HSREB) has reviewed and approved the amendment to the above named study, as of the HSREB Initial Approval Date noted above.

HSREB approval for this study remains valid until the HSREB Expiry Date noted above, conditional to timely submission and acceptance of HSREB Continuing Ethics Review.

The Western University HSREB operates in compliance with the Tri-Council Policy Statement Ethical Conduct for Research Involving Humans (TCPS2), the International Conference on Harmonization of Technical Requirements for Registration of Pharmaceuticals for Human Use Guideline for Good Clinical Practice Practices (ICH E6 R1), the Ontario Personal Health Information Protection Act (PHIPA, 2004), Part 4 of the Natural Health Product Regulations, Health Canada Medical Device Regulations and Part C, Division 5, of the Food and Drug Regulations of Health Canada.

Members of the HSREB who are named as Investigators in research studies do not participate in discussions related to, nor vote on such studies when they are presented to the REB.

The HSREB is registered with the U.S. Department of Health & Human Services under the IRB registration number IRB 00000940.
Appendix II
Pre- and Post-Surgical Silhouettes for 22 Patients

Patient 1: Pre-Surgical

Patient 1: Post-Surgical

Patient 2: Pre-Surgical

Patient 2: Post-Surgical
Patient 3: Pre-Surgical

Patient 3: Post-Surgical

Patient 4: Pre-Surgical

Patient 4: Post-Surgical
Patient 9: Pre-Surgical

Patient 9: Post-Surgical

Patient 10: Pre-Surgical

Patient 10: Post-Surgical
Patient 11: Pre-Surgical

Patient 11: Post-Surgical

Patient 12: Pre-Surgical

Patient 12: Post-Surgical
Patient 13: Pre-Surgical

Patient 13: Post-Surgical

Patient 14: Pre-Surgical

Patient 14: Post-Surgical
Patient 15: Pre-Surgical

Patient 15: Post-Surgical

Patient 16: Pre-Surgical

Patient 16: Post-Surgical
Patient 17: Pre-Surgical

Patient 17: Post-Surgical

Patient 18: Pre-Surgical

Patient 18: Post-Surgical
Patient 19: Pre-Surgical

Patient 19: Post-Surgical

Patient 20: Pre-Surgical

Patient 20: Post-Surgical
Patient 21: Pre-Surgical

Patient 21: Post-Surgical

Patient 22: Pre-Surgical

Patient 22: Post-Surgical
Appendix III
Letter of Information and Consent

Aesthetic Outcomes of Maxillomandibular Advancement Surgery for the Treatment of Obstructive Sleep Apnea
Letter of Information/Consent

Principle Investigator
Dr. Ali Tassi
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Schulich School of Medicine and Dentistry
The University of Western Ontario
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Co-Investigators
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Schulich School of Medicine and Dentistry
The University of Western Ontario
University Hospital – Department of Dentistry

Dr. Jennifer Curran
Orthodontic Resident, Division of Graduate Orthodontics
Schulich School of Medicine and Dentistry
The University of Western Ontario
Introduction
You are being invited to participate in a research study directed by Dr. Michael Shimizu and Dr. Ali Tassi along with their resident, Dr. Jennifer Curran, to evaluate aesthetic outcomes in patients who have undergone maxillomandibular advancement surgery for treatment of obstructive sleep apnea syndrome (OSAS).
We are currently asking for participants 18 or over who are oral and maxillofacial surgeons, orthodontists, or individuals without a background in the dental/surgical field. You have met these criteria for participation in this study, if you wish. We have provided this consent form for you to read carefully, and will answer any questions you may have regarding the information it contains.

Purpose of Study
The purpose of this study is to evaluate the facial changes in patients treated for obstructive sleep apnea with maxillomandibular advancement (MMA) surgery. This information may help clinicians in selecting patients for MMA surgery and enable them to provide patients with information about expected clinical outcomes in regards to sleep, aesthetics and quality of life. Dr. Jennifer Curran, a resident in the Graduate Orthodontics Program at the University of Western Ontario, will administer the study. The study will consist of a series of silhouettes that you will be asked to assess for aesthetics, paying particular attention to the facial balance in each silhouette.

Procedures
The individuals who will be invited to complete the study are oral and maxillofacial surgeons, orthodontists, or individuals without a background in the dental/surgical field. Participation in the study is completely voluntary, and participants are able to withdraw their participation at any time. This letter of information and consent describes the study so you can make an informed decision on participating. Please feel free to ask questions if anything is unclear or if there are phrases or words you do not understand. If you agree to participate, you will be asked to complete an electronic document indicating the attractiveness of patient silhouettes. We will address any questions you have as needed.

Number of Participants
There are 100 potential individuals who may participate in this study.

Participant Inclusion and Exclusion Criteria
Participants will be included if they have completed a specialty degree in orthodontics or in oral and maxillofacial surgery. Other participants may be included if they do not have any background in dental or surgical fields, and if they are over the age of 18. Participants who are unable to give informed consent will be excluded.
Description of the Research
As a participant in the study, you will be asked to fill out an information package rating the esthetics of various silhouettes. This will take approximately 20-30 minutes. Below each silhouette, a scaled ruler is provided, and you are asked to rate each silhouette ranging from very unattractive (0) to very attractive (10) by placing a line directly on the ruler to indicate your esthetic preferences. This marking can be anywhere along the line, and does not have to be a whole number. In assessing “attractiveness”, we will ask you to consider how pleasing the patient’s silhouette is to you, paying attention to things like facial balance. Dr. Curran will examine and analyze the data collected to draw conclusions regarding patient esthetics following maxillomandibular advancement surgery for the treatment of obstructive sleep apnea. None of your personal data will be released from the study, other than your profession, gender, and age. After completing the survey, no follow up is required with respect to this research project specifically.

Time Requirements
The completion of the study should take approximately 20-30 minutes.

Risks
No risks are thought to be associated with the completion of this study.

Benefits
We hope to gather insight into the aesthetic outcome of patients undergoing maxillomandibular advancement surgery for obstructive sleep apnea. It is thought that this will aid in future treatment planning of surgery for patients, and provide potential patients with information about clinical outcomes.

Right to Refuse
Your participation in this study is voluntary. You may refuse to participate or you may withdraw from the study at any time with no effect on the results of your treatment. You do not waive any of your legal rights by signing the consent form.

Compensation for Participation
There is no compensation for the study.

Use of Data
Data collected via the questionnaire will be secured via encrypted, and password protected software and hard drives, and locked in appropriate University servers and storage facilities.

New Findings
If, during the course of this study, new information becomes available that may relate to your willingness to continue to participate, this information will be provided to you by the investigator.
Confidentiality
Your privacy will be respected. If the results of this study are published, your name will not be used and no information that discloses your identity will be collected or released. To monitor the conduct of research, the research team, authorized study personnel, Western University Health Science Research Ethics Board and the Lawson Health Research Institute may require access to your study-related records. Additionally, representatives of the Research Ethics Board may follow up with you directly for the same purpose. All participants will be given a study number. Only that number will be used on any study analysis related documents. By signing the consent form you allow Dr. Curran to review the questionnaire you will fill in.

Contacts
If you have any questions during the study, or wish to withdraw from the study at any time, you may contact Dr. Michael Shimizu, Dr. Ali Tassi or Dr. Jennifer Curran. If you have any questions or concerns about your rights as a research participant or the conduct of this study you may contact: Dr. David Hill, Scientific Director, Lawson Health Research Institute.

Consent
I have read and understand the consent form for this study. I have been given sufficient time to consider the above information and to seek advice, if so desired. I have had the opportunity to ask questions which have been answered to my satisfaction. I am voluntarily agreeing to participate in this study. I will be provided with a copy of this consent form for my own information, if I wish.

By signing below, I am agreeing to participate in this study.

Name: __________________________________________

Date: __________________________________________

Signature: ______________________________________

Consent obtained by:

Name: __________________________________________

Date: __________________________________________

Signature: ______________________________________
Appendix IV

Survey in Entirety

Consent Form

I have read, understood, and printed a copy of the above consent form and desire of my own free will to participate in this study.

- Yes
- No

On the following slides you will see profile silhouettes of patients who have undergone maxillomandibular advancement (jaw) surgery as treatment for obstructive sleep apnea. Some of these silhouettes represent the patients before surgery, and some silhouettes are following surgery. Please rate each silhouette on the sliding scale from 0 (very unattractive) to 10 (very attractive). The rating does not need to be a whole number, and may include a decimal. There are 50 silhouettes in total, and the survey should take approximately 10-15 minutes to complete.
What is your gender?
- Male
- Female

What is your age?
- 18-29
- 30-39
- 40-49
- 50+

What is your profession?
- Oral and Maxillofacial Surgeon
- Orthodontist
- Other

Which province (if in Canada) or country do you live in?

Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of an older adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive
1=Very Attractive

Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive
1=Very Attractive
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).
Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0 = Very Unattractive

10 = Very Attractive

Please rate the following silhouette of an older adult male for facial balance (attractiveness).

0 = Very Unattractive

10 = Very Attractive
Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive                      10=Very Attractive

Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive                      10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive


Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

![Silhouette of an older adult female for facial balance](image1)

0=Very Unattractive 10=Very Attractive

Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

![Silhouette of a middle-aged adult male for facial balance](image2)

0=Very Unattractive 10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive    10=Very Attractive

Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive    10=Very Attractive
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive

Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive
10=Very Attractive
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).
Please rate the following silhouette of a middle-aged adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

Please rate the following silhouette of an older adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

>>
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive
Please rate the following silhouette of a middle-aged adult female for facial balance (attractiveness).

0=Very Unattractive                      10=Very Attractive

Please rate the following silhouette of an older adult male for facial balance (attractiveness).

0=Very Unattractive                      10=Very Attractive
Please rate the following silhouette of an older adult male for facial balance (attractiveness).

0=Very Unattractive

Please rate the following silhouette of a young adult male for facial balance (attractiveness).

0=Very Unattractive
Please rate the following silhouette of an older adult female for facial balance (attractiveness).

0=Very Unattractive

10=Very Attractive

We thank you for your time spent taking this survey. Your response has been recorded.
Curriculum Vitae

Name: Jennifer L. Curran

Post-secondary Education: University of Western Ontario
London, Ontario, Canada
Honours Bachelor of Science 1997-2001

University of Western Ontario
London, Ontario, Canada
Doctor of Dental Surgery 2002-2006

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