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Review

Changing practice: Trends in skeletal surgery for obstructive sleep apnea

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ABSTRACT

Objectives/Hypothesis: The objective of this study was to systematically review the English literature for articles that have described skeletal surgeries in the treatment of obstructive sleep apnea in both adults and children. From these articles trends and patterns in the treatment of OSA with skeletal procedures are described.
Study design: Three databases including MEDLINE, Google Scholar and the Cochrane Library were searched through May 1, 2018.
Methods: The systematic and independent literature reviews were performed and the determination of included studies was made by consensus. Relevant studies were examined based on six categories of skeletal surgery: 1) Hyoid Advancement 2) Genioplasty/Genioglossus Advancement 3) Maxillary Expansion 4) Maxillomandibular Advancement 5) Mandibular Distraction and 6) Maxillomandibular Expansion.
Results: 1875 studies were analyzed for inclusion of which 414 were ultimately included in our analysis. A steady increase in the publication of articles pertaining to maxillary expansion and maxillomandibular advancement was identified. Research interest in hyoid advancement and genioplasty/genioglossus advancement has declined in the past decade.
Conclusions: Changing trends in skeletal surgery for OSA offer exciting and efficacious therapeutic surgical modalities. MMA is the most widely studied and efficacious multi-level surgery for OSA today. Newer modalities such as adult maxillary expansion offer encouraging early results with minimal complication rates, and further study should be directed in this area.

1. Introduction

Since the early 1980s a number of surgical approaches have been used in the treatment of obstructive sleep apnea (OSA). These include: tracheostomy, soft tissue surgeries, nasal surgery and skeletal surgeries including maxillomandibular advancement (MMA) (Camacho et al., 2013). The first described surgical treatment for OSA was by Valero and Alroy who described a case report of a 55 year-old man presenting with excessive daytime sleepiness and micrognathia treated with tracheostomy (Valero and Alroy, 1965). Particularly in the discussion of skeletal surgery, mandibular advancement alone was originally described in 1975 with the first descriptions of genioglossus advancement, hyoid suspension, and maxillomandibular advancement following shortly thereafter in the mid 1980s (Camacho et al., 2013).

OSA is undoubtedly a heterogenous disorder comprised of a complex and multi-faceted pathophysiology. A number of factors including impaired anatomy, low arousal threshold, ventilatory control instability, and reduced upper airway muscle tone amongst others have been identified as potential pathological correlates (Eckert et al., 2013). These anatomical targets have been exploited by

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sleep surgeons as potential therapeutic targets for novel surgical therapies, particularly in relation to skeletal surgery. The first of these targets is hyoid bone position, which has often been described to be abnormally low in patients with OSA and may in fact depress during sleep resulting in worsening hypopharyngeal obstruction. Given that airflow resistance is proportional to airway length, decreasing airway length can provide a decrease in resistance (Camacho et al., 2013). Anterior movement of the hyoid complex can improve patency of the hypopharyngeal airway by decreasing obstruction at the level of the tongue base. This underlies the therapeutic basis of hyoid surgery, which was first described by Riley et al. using the technique of hyoid myotomy and suspension, with the use of suspension of the hyoid to the mandible using fascia lata (Riley et al., 1984). Today, hyoid surgery is comprised of four main techniques including: hyoid myotomy and mandibular suspension, hyoidthyroidpexy, expansion hyoidplasty, and hyoid sectioning.

Maxillary transverse deficiency, which is defined as narrowing of the dental arch in the transverse dimension, has been previously described as a potential pathophysiological correlate in the development of OSA (Cistulli et al., 1998). Maxillary transverse deficiency is believed to increase nasal airflow resistance and displace the posterior tongue leading to pharyngeal collapse (Cistulli et al., 1998; Zeng et al., 2008). It is hypothesized that the mechanism of action of maxillary expansion is widening of the nasal floor with improved tongue position. Some authors have stipulated that widening of the maxilla increases muscle tension on the palatal muscles which insert onto the maxilla, thus reducing palate collapsibility during sleep (Abdullatif et al., 2016).

Thirdly, movement of the genial tubercle at the anterior-inferior aspect of the mandible with its associated soft tissue is another bony skeletal surgery option for OSA. First described by Riley et al., in 1984, the sliding genioplasty or genioglossus advancement has taken on many forms, all of which attempt to stabilize the tongue base and decrease hypopharyngeal narrowing. This surgery is often performed in combination with uvulopalatopharyngoplasty or hyoid advancement, but can significantly improve OSA as a standalone procedure (Song et al., 2017).

The most involved of the described skeletal procedures is MMA. This procedure is typically reserved for patients demonstrating lateral pharyngeal wall collapse who are refractory to medical treatment modalities. By focusing on advancement and rotation of both the maxilla and mandible, maximal effects can be achieved (Moher et al., 2009). Most notably, this procedure significantly increases airway caliber in a multi-level fashion while simultaneously increasing tension on the lateral pharyngeal soft tissue (Liu et al., 2015; Zaghi et al., 2016).

Our review of the literature sought to examine trends and patterns in research interest in skeletal surgeries for the treatment of obstructive sleep apnea since 1980. These skeletal surgeries comprised the following six main categories for analysis: 1) Hyoid Advancement 2) Genioplasty/Genioglossus Advancement 3) Maxillary Expansion 4) Maxillomandibular Advancement 5) Mandibular Distraction and 6) Maxillomandibular Expansion.

2. Materials and methods

Two of the authors (M.A. and C.G.) performed an independent literature search in each of the following categories of skeletal surgery: 1) Hyoid Advancement, 2) Genioplasty/Genioglossus Advancement, 3) Maxillary Expansion, 4) Maxillomandibular Advancement, 5) Mandibular Distraction, 6) Maxillomandibular Expansion to identify studies with potential relevance via search of MEDLINE, Google Scholar, and the Cochrane Library. After consensus was agreed upon by the two independent reviewers, the remaining authors (S.Z., M.C., S.L.) reviewed the appropriate database to ensure all included studies met the criteria for inclusion and exclusion. The time range reviewed spanned January 1, 1980 – May 1, 2018. Throughout the duration of the study, the PRISMA statement was followed (Moher et al., 2009).

2.1. Study selection

Eligible study designs included in the search database for this systematic review included published abstracts, case reports or series, case controls, cohort, randomized and non-randomized trials as well as meta analyses or systematic reviews. For each of the above-defined skeletal surgery categories, the studies included are those which reported the following: 1) Pediatric and adult patients (age: any) who were diagnosed with OSA and for whom one of the pre-defined skeletal surgeries was used either alone or in conjunction with another surgical modality. Exclusion criteria included studies where one of the pre-defined skeletal studies was used in conjunction with two or more surgical modalities in a single stage and studies which were outside of the scope of the English literature.

2.2. Statistics

Trends in the published literature pertaining to each surgical procedure were assessed using descriptive statistics to report the number of publications published in 5-year intervals spanning from 1995 to 2017. The total number of articles published for each category by level of evidence is reported. Graphical representation using box-whisker plot is provided to portray the strength of the level of evidence and changes over time with respect to the year published.

3. Results

3.1. Hyoid advancement

536 articles were reviewed for relevancy of which 64 were included for analysis. Of these 64 studies, 18 were of Level V evidence, animal or cadaveric studies. 41 studies were Level IV, 2 were Level III, 2 were Level II, and 1 study was of Level IB evidence (Table 1 and Fig. 1).

Table 1
Number of studies published by procedure type, arranged by 5-year interval.

5 Year Interval	Number of Studies Published By Procedure Type						
	HA (n = 64)	GP/GGA (n = 64)	ME (P) (n = 36)	ME (A) (n = 10)	MMA (n = 125)	MD (n = 113)	MME (n = 4)
2017–2012	14	16	26	7	67	42	2
2011–2007	14	17	5	1	25	35	1
2006–2001	18	23	3	1	12	29	1
2000–1995	9	5	1	1	14	7	0
1995–1989	9	3	1	0	7	0	0

HA – Hyoid Advancement, GP/GGA – Genioplasty/Genioglossus Advancement, ME (P) – Maxillary Expansion (Pediatric), ME (A) – Maxillary Expansion (Adult), MMA – Maxillomandibular Advancement, MD – Mandibular Distraction, MME – Maxillomandibular Expansion.

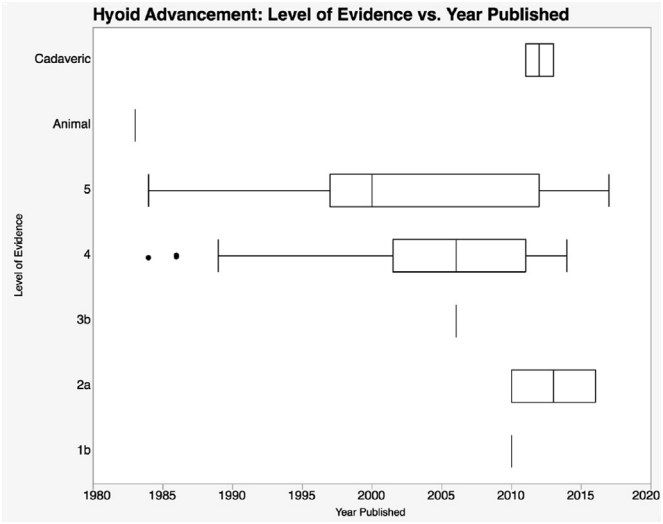

Fig. 1. Number of studies published on hyoid advancement, by level of evidence.

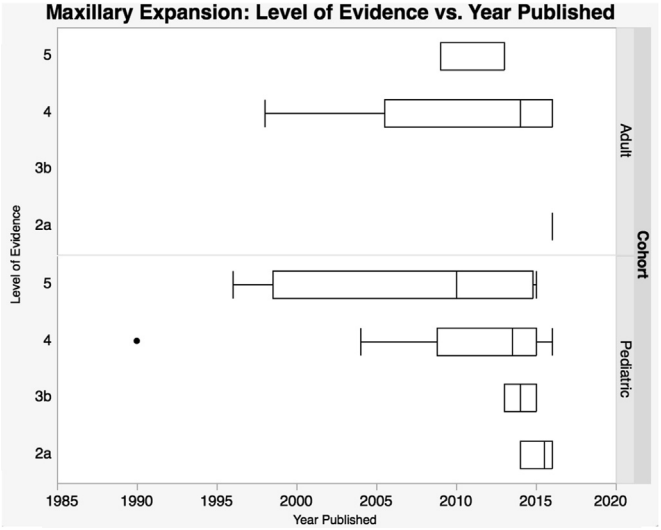

Fig. 3. Number of studies published on maxillary expansion, by level of evidence.

3.2. Genioplasty/Genioglossus advancement

586 articles were screened for relevancy, of which 64 articles were included for analysis. Level V studies comprised 20 of those included. 35 studies were Level IV evidence, 1 was Level III, 7 were Level II. No Level I studies were identified (Fig. 2).

3.3. Maxillary expansion

For the search on Maxillary Expansion, 69 articles were found, of which 44 were included for analysis. Results are divided into adult (age > 18) and pediatric (age < 18) (Fig. 3).

3.3.1. Pediatric

36 studies using maxillary expansion as a single modality or in combination with adenotonsillectomy (n = 2) in children with OSA were included for analysis. The earliest published literature was identified in 1990, with 4 of the analyzed studies identified as Level V evidence, 22 as Level IV, 4 as Level III, and 6 as Level IIA. No Level I studies were identified.

3.3.2. Adult

10 adult studies using maxillary expansion as a single modality for the treatment of Sleep-Disordered Breathing (SDB) or OSA were identified. Of these, 4 studies were Level V, 5 were Level IV, and 1 was Level II evidence. As in the pediatric sub-group, no Level I Studies were identified.

3.4. Maxillomandibular advancement

195 studies were identified in our literature search, of which 125 were included for analysis. Of these 125 studies, 3 were of Level V evidence, 96 were Level IV evidence, 4 were Level III evidence, 18 were Level II evidence, and 3 studies were of Level I evidence (Fig. 4).

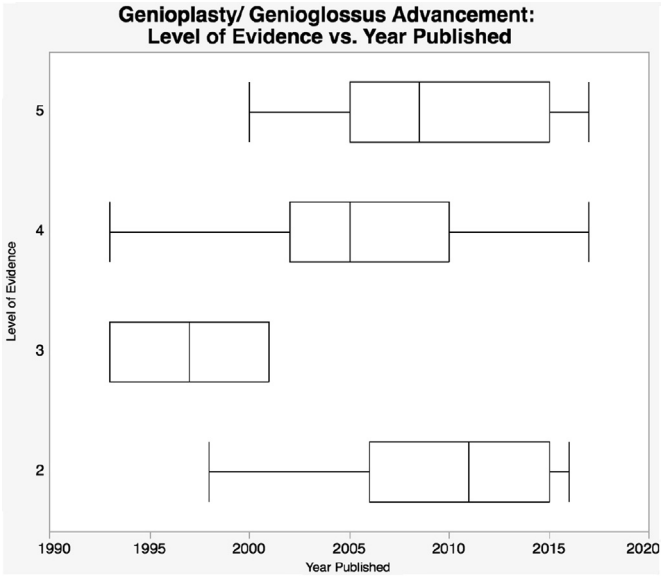

Fig. 2. Number of studies published on genioplasty/genioglossus advancement, by level of evidence.

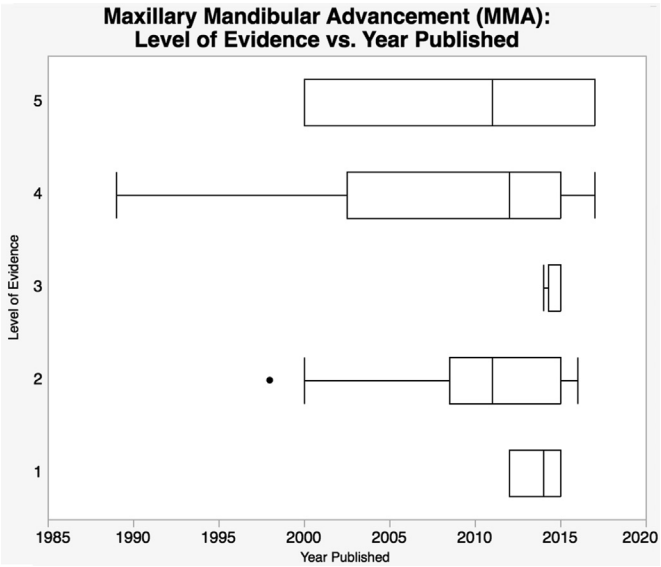

Fig. 4. Number of studies published on MMA, by level of evidence.

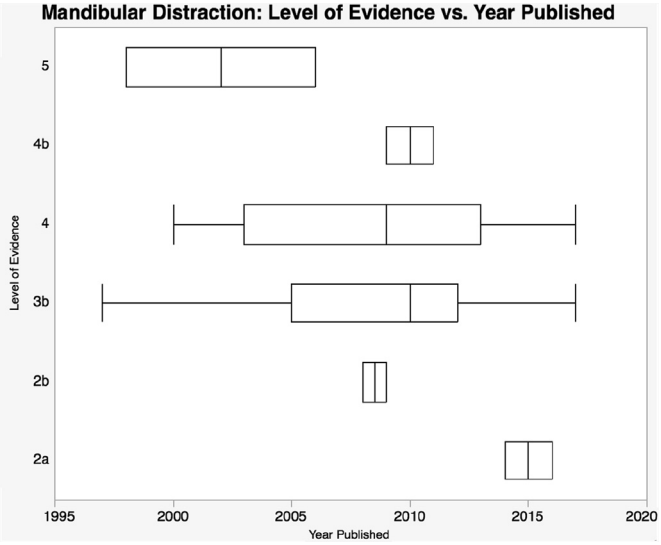

Fig. 5. Number of studies published on mandibular distraction, by level of evidence.

3.5. Mandibular distraction

425 studies were screened for relevancy, of which 113 were included in the final analysis. The earliest published literature was identified in 1997; 3 studies identified were of Level V evidence, 25 were of Level IV evidence, 81 were of Level III evidence, 4 were Level II evidence, and 0 were Level I (Fig. 5).

3.6. Maxillomandibular expansion

64 studies were screened for relevancy, of which 4 were included for analysis. The first published literature was identified in 2004. One study was identified as Level 2A evidence, one study was identified as Level 4 evidence, and the remaining two studies were case reports (Fig. 6).

4. Discussion

In the United States, approximately 15,000 skeletal surgeries are performed per year (Kezirian et al., 2010). Despite the gamut of available skeletal procedures used to treat OSA today and their widespread use, there has been no large-scale review identifying current research trends in this discipline. The goal of our review was to identify current targets for research interest and identify which skeletal procedures have seen waning popularity since the original description of the Stanford Protocol in 1986.

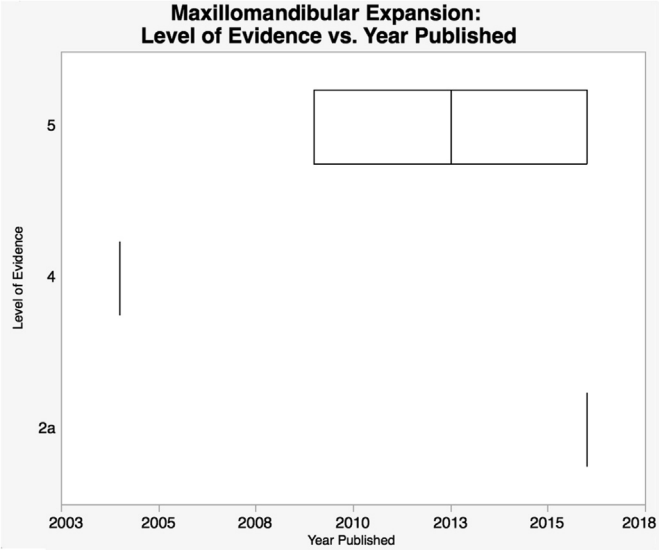

Fig. 6. Number of studies published on maxillomandibular expansion, by level of evidence.

With respect to hyoid advancement, the vast majority of published literature spans the early 2000s to present date, with the entirety of level I and II evidence published within the last 6 years. Overall, isolated hyoid advancement has been shown to significantly reduce AHI in adult patients with OSA with two main techniques. Hyoid myotomy with suspension and hyoidthyroidpexy demonstrate predominance in the published literature over the last decade. Only three studies examined expansion hyoidplasty (Hamans et al., 2013; Patton et al., 1983; Toh et al., 2013). One of these studies reported pre-operative and post-operative AHI results demonstrating non-favorable results (Hamans et al., 2013). This review of the literature, although somewhat lacking in higher level evidence, demonstrates hyoid surgery alone can reduce OSA severity in adults with additional study required to elucidate its' role as a single-use modality.

The use of genioplasty and genioglossus advancement was first advocated in the publication by Riley et al. describing the Stanford Protocol in 1986 (Riley et al., 1986). Since this time the vast majority of publications on the topic have been composed of either small case-series (level IV) or case reports (level V) of surgical modifications, as well as combinations with other surgical procedures, namely hyoid suspension. A recent meta-analysis of genioplasty and advancement techniques confirmed these procedures are effective in improving OSA outcomes such as AHI and lowest oxygen saturation. However there remains a tremendous amount of heterogeneity in the published literature and a need for more standardized research on this procedure (Song et al., 2017).

In more recent literature maxillary expansion for OSA in adults has seen rising prominence. The procedure was first described in a case report published in 1998 by Cistulli (Cistulli et al., 1998). Our review demonstrated a growing research interest in its use in adults with 9/10 included studies published between 2013 and 2017. Only one of these studies was of Level II evidence or higher, a systematic review and meta analysis published by our group (Abdullatif et al., 2016). In this review, six studies (with a cumulative total of 36 patients) reported pre and post-operative with a mean decrease in AHI of 59.3%. These results are encouraging, demonstrating minimal complication rates, and this area of active research holds significant promise for adults with OSA (Bach et al., 2013; Vinha et al., 2016a, 2016b).

While maxillary expansion in adults is an area of rising interest, Rapid Maxillary Expansion (RME) in children has been well described in the literature with increasing publication interest in the past five years. According to a large-scale systematic review and meta-analysis by Camacho et al. RME showed a mean 70% improvement in AHI in children with OSA. No large-scale randomized trials have been performed on its efficacy and long-term results have not been elucidated. Of great importance, only one study identified in our review reported on complications of RME in children (none were reported) as well as long term follow-up (>3 years) (Pirelli et al., 2015). This is an area in need of further study to confirm procedural safety and risk of recurrence. It is encouraging that four of the six available Level II studies have been published in the past two years indicating an emerging trend towards higher level evidence in this area.

Finally, the cornerstone of skeletal surgery in OSA: MMA has endured as the most widely studied of the bony skeletal surgeries in our review. It remains the most effective multi-level surgical

option for refractory OSA, especially in those who have undergone prior surgical therapies. Beginning with early reports of its utility in refractory OSA cases in the late 1980s, it has remained a staple of the sleep surgery literature. In recent years there has been an increase in higher quality studies utilizing internal controls (CPAP or other surgical modifications) to evaluate the effects of MMA. This continues in the present time, with continued high volume MMA research on modifications to surgical technique as well as longer-term outcomes with impressive results (Zaghi et al., 2016).

5. Conclusion

There remains a large heterogeneity in the level and quality of evidence available in sleep surgery today. Our review indicates promising research trends in the area of skeletal surgery, particularly in the area of maxillary expansion, which is demonstrating encouraging results in this growing field. Maxillomandibular advancement remains the cornerstone of skeletal surgical therapy, with impressive results and emerging literature on long-term complications, outcomes, and surgical modifications.

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