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Review Article

Treatment of OSAHS associated with skeletal class II dentofacial anomaly through sagittal mandibular osteogenic distraction of the bilateral branch and box genioplasty: A case report and systematic review

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ABSTRACT

Introduction: Obstructive Sleep Apnea-Hypopnea Syndrome (OSAHS) is a significant public health issue affecting 2 %–4 % of adults globally due to its high prevalence and underdiagnosis. It is characterized by airway obstruction, particularly in individuals with craniofacial abnormalities. Surgical interventions, such as mandibular distraction osteogenic (MDO) and maxillomandibular advancement, can correct these abnormalities and improve the airway.

Objective: The purpose of this study is to present a combination of surgical techniques: MDO by bilateral sagittal split osteotomy (BSSO) plus box genioplasty of the mandibular symphysis to optimize the increase in the anteroposterior airway as an initial treatment, before orthognathic surgery or as a definitive treatment in patients with OSAHS.

Methods: A systematic review, following PRISMA guidelines, was performed using searches in the MEDLINE/PubMed, Cochrane and Web of Science databases. Various variables were considered and presented comprehensively in tables and figures alongside a case report.

Results: Postoperative analyses demonstrated airway improvements in the 3, 6 and 9 months, with a final relapse of 19.8 % in B point, 30 % in pogonion and 19.6 % in the minimum cross-sectional area, respectively. Evaluations showed enhancements in clinical parameters such as saturation, AHI and polysomnographic in the ninth month.

Conclusion: MDO by BSSO plus box genioplasty in adult patients with OSAHS and class II skeletal dentofacial abnormalities significantly increase airway without bone relapse, suggesting an effective therapeutic option for this condition.

1. Introduction

Obstructive sleep apnoea/hypopnea syndrome (OSAHS) is a disease characterized by intermittent and repeated episodes of total (apnoea) or partial (hypopnea) obstruction of the upper airways (UA) during sleep [1–3]. It can cause excessive daytime sleepiness, neurocognitive disorders, quality of life deterioration and an increased risk of accidents. It has also been associated with cardiovascular, metabolic and renal diseases [2,4–6].

Epidemiological studies demonstrate that OSAHS is quite common in the general population, with a prevalence of 34 % in men and 17 % in women aged 18–60 years, representing 2 %–5 % of the global adult population. Despite its frequency, OSAHS is an underdiagnosed condition [7–10]. Risk factors include male sex, age $\geq$ 65 years, body mass index (BMI) > 30, and African American or Latin American ancestry [11]. The diagnostic criteria for OSAHS are defined as sleepiness, alone or associated with at least two other factors (repeated micro-awakenings, non-restorative sleep and nocturia, among others), or polysomnography with an apnoea-hypopnea index (AHI) $\geq$ 5 events per hour [10].

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The severity of OSAHS is evaluated according to AHI and daytime sleepiness, both classified into three levels: mild (AHI 5–15; unwanted sleepiness with little impact on social or professional life), moderate (AHI 15–30; unwanted sleepiness with a moderate impact on social or professional life) or severe (AHI $\geq$30; unwanted sleepiness with a significant impact on social or professional life). The severity level is defined by the most severe component [10].

Anatomical factors that increase the risk of OSAHS are related to craniofacial and soft-tissue abnormalities [1,12]. Congenital syndromic and non-syndromic mandibular micrognathia are usually associated with a small and abnormally shaped upper airway, which increases the risk of collapse [13]. Additionally, increases in the volume of the lateral pharyngeal walls and tongue represent soft-tissue risk factors [14].

Imaging methods used for airway evaluation include cephalometric radiography, magnetic resonance imaging (MRI), computed tomography (CT), and cone beam computed tomography (CBCT) [15]. Fujita and Sher even recommend fibre-optic pharyngoscopy [4].

Several surgical procedures have been proposed to correct craniofacial abnormalities associated with OSAHS, such as mandibular distraction osteogenic (MDO), maxillomandibular advancement (MMA), genioplasties, orthognathic surgery or a combination of these [16].

In 1950, Ilizarov et al. [17] developed an external skeletal fixation system combined with biomechanical stimulation methods to form new bone tissue within the site widened by distraction osteotomy. They also described the 'tension-stress' effect, which occurs when bone and surrounding tissue expand under ideal circumstances [18]. These traction forces form a flat tissue parallel to the tension vector applied by the distractor [19].

In 1992, McCarthy et al. reported their results regarding the gradual lengthening of human mandibles. Two years later, Havilik and Barlett et al. [20] and Moore et al. [21] reported treatment for severe micrognathia using extraoral distractors [22]. Since then, osteogenic distraction (OD) has been increasingly applied in the craniofacial region [18].

The osteotomy is performed, and the OD device is rigidly fixed to the bone. Following a latency period of 0–7 days to allow positive regulation of bone metabolism, the device is activated at a rate of 1 mm/day, with possible variations in the distraction range [23].

Mandibular osteotomies are crucial for correcting dentofacial deformities [24]. The primary surgical technique employed for mandibular mobilization is bilateral sagittal split osteotomy (BSSO) [25].

The first design of the BSSO technique was implemented by Obwegeser and Trauner in 1954, through an extraoral approach involving a horizontal cut over the lingula, which was later re-angled by modifications from Kazanjian [26]. Dal Pont modified the Obwegeser method by introducing a retromolar osteotomy, reducing the displacement of the proximal segment caused by muscle activity and allowing its use for prognathism, retrognathism and open bite cases [25].

Hunsuck et al. suggested that the medial osteotomy should extend to the posterior region of the lingula without affecting the posterior edge of the ramus, and the lateral osteotomy should be performed at the junction of the ramus and the mandibular body distal to the second molar [25,27]. Epker described another modification of the Obwegeser and Dal Pont technique, minimizing complications such as excessive oedema, neurological complications related to the inferior alveolar nerve and hemorrhages [25].

BSSO is a fundamental surgical technique used to treat class II mandibular hypoplasia [24], facilitating osteotomies in orthognathic surgery and applying it in anteroposterior OD.

An initial intervention with mandibular advancement improves the patient's airway and facial appearance. However, some studies suggest that this correction is prone to failure, requiring re-interventions in some cases [16]. The surgical trend has shifted from mandibular surgery alone to the use of combined MMA [28,29].

Multiple surgical options target the manipulation of the tongue, hyoid, mandible and retrolingual pharynx. Advancement of the genioglossus muscle has proven to be an effective treatment [30]. This muscle is the primary dilator of the pharynx; besides protruding the tongue, its role has been implicated in the pathophysiology of OSAHS, where UA collapse occurs due to dilator muscle failure [31].

In 1942, Hofer et al. performed the first genioplasty with an extraoral approach involving osteotomy and advancement of the suprahyoid muscles, anterior digastric muscle, geniohyoid and platysma, achieving an increase of 1–1.5 cm. In 1957, Obwegeser and Trauner modified the technique through an intraoral approach, without releasing the platysma muscle, achieving more aesthetically favorable results [32]. Genioplasty involves stabilization of the hypopharyngeal airway through a sliding horizontal osteotomy that allows anterior movement of the genioglossus complex, adding tension to the tongue base and expanding the airways [31].

Surgical techniques for mandibular advancement through maxillary osteotomies have reported medium- and long-term postoperative relapses, necessitating complementary techniques to optimize airway advancement.

The purpose of this work is to present the combination of surgical techniques in the mandible: distraction through BSSO plus box genioplasty of the mandibular symphysis to optimize the increase in the anteroposterior airway diameter quantified in the volumetric (three-dimensional) analysis of a patient with OSAHS and skeletal class II dentofacial anomaly, along with a systematic review of OD in this type of treatment.

2. Methods

2.1. Case report

2.1.1. Patient information

A 41-year-old male patient was admitted to a private practice of Maxillofacial Surgery in December 2022. His reason for consultation was possible sleep apnea and retrognathia. As a morbid history, he had hypertension. He did not report any allergies or consumption habits such as alcohol, tobacco or drugs.

2.1.2. Diagnostic evaluation

After an evaluation by the specialist, imaging tests and polysomnography, OSAHS and skeletal dentofacial anomaly class II were diagnosed. For the CT scan of the patient undergoing this study, was used the SOMATOM Sensation 16/Cardiac, VB42 (Siemens AG Healthcare), a multi-detector computed tomography (MDCT) scanner. The protocol proposed by Whyte et al.[33] was followed, which included the preparation and positioning of the patient according to the following points: immobilize the head with the Frankfort plane at 90° to the table; tilt and rotation should be avoided; the teeth should be in occlusion for both the examination and the volumetric examination; swallowing and breath holding should be avoided during the examination; calm breathing is encouraged; tongue position on the palate. Likewise, for the measurement of condylar angulation, the protocol of Tabatabaei et al. was followed, which describes condylar angulation as the angle formed between the condylar axis (i.e., a line perpendicular to the longitudinal axis of the condylar process) and the sagittal plane. It should be noted that this parameter was measured in the axial projection, where the condylar process had the largest mediolateral diameter [34].

He started orthodontic and periodontal treatment after maxillofacial evaluation. An initial alveolar bone graft was performed to address bone support deficits at the roots of the upper and lower anterior teeth. After eight months of orthodontic treatment, a meeting was held with the treatment team because the patient's local bite conditions created a molar loading zone with section and rupture of the orthodontic arch, generating inadequate progress of orthodontic treatment. The interdisciplinary decision was made to start with OD through BSSO after a two- and three-dimensional (3D) analysis with VTO (virtual treatment objectives) design.

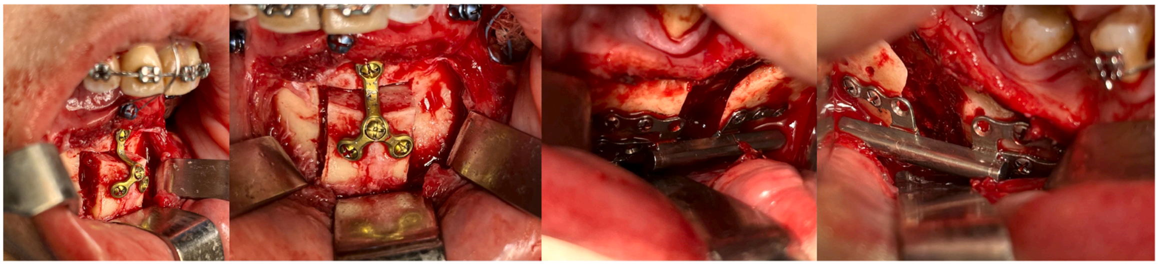

Fig. 1. Intraoperative photographs. Box advancement genioplasty of the mandibular symphysis lateral view (1.1) and frontal view (1.2). Extraoral in situ mandibular osteogenic distraction devices in bilateral sagittal split osteotomy (BSSO) (1.3 and 1.4).

Fig. 2. Airway measurements at different points. Anteroposterior and transverse airway measurements in axial view (2.1). Airway measurements performed in the projection of point B, pogonion point and MCA in axial view (2.2). Distance between the hyoid and the lower. border of the mandible in sagitario view (2.3).

2.1.3. Therapeutic intervention

Two X0101–20 mandibular osteogenic distractors of the Orthomax Cibey brand were adapted to stereolithographic models, developing a BSSO design that extends distal to the last molar and performing a guide for cutting and positioning the distractors in the position chosen by the surgeon.

In November 2023, the procedure was performed under general anesthesia. The anesthesiology team determines a difficult airway, achieving nasotracheal intubation with video laryngoscope without the need for vigilant intubation or more invasive maneuvers.

Local anesthetic 200 mg of 2 % lidocaine with 1:100,000 epinephrine, 5 tubes of 1.8 cc is infiltrated. Starting the procedure with the vestibular incision of the chin with cold scalpel blade number 15 for osteotomy box genioplasty, which consists of a box designed in the mandibular symphysis whose caudal edge is the entire thickness of the basilar edge on both sides of the symphysis (Fig. 1.1 and 1.2). The segment rises approximately 14 mm towards the cephalic and then joins at the midline. This dimension is calculated by reviewing the preoperative 3D image, the design of the osteotomy must be developed over the genioglossal and geniohyoid processes that have the insertion of the respective muscles. All osteotomy is performed with piezoelectric instruments to prevent blood vessel involvement in the floor of the mouth. The Biomet osteosynthesis plate is positioned in a Y-shape adapted with four 8 mm screws; the box is advanced to verify the contact of the inner cortical of the box with the external cortical of the native chin; in this case, an advance of 7 mm was achieved (Fig. 2).

Then, the mandibular vestibular approach is performed with electrocautery to access the BSSO with a cutting guide, using a reciprocating saw, without producing openings of the mandibular branches. Both distractors are positioned using locating guides, which are fixed with four screws each, which serve as guides so that after opening the branches, there is an imprint of the device, added to a marking with a graphite pencil of the contour of the distractors (Fig. 1.3 and 1.4).

A small retromandibular incision of 5 mm on each side and a blunt dissection are made, tunnelling both stems of the distractors (stems are understood as the active ends that allow the devices to rotate). Subsequently, the distractors are removed, and the mandibular branches are opened with chisels, using the progressive opening method, without using a hammer to avoid condylar loading. Once both mandibular branches are opened, the indemnity of the inferior alveolar nerve is verified. This anatomical element should be ideally positioned in the distal (dentate) mandibular segment; otherwise, the neurovascular package is released. Then, the dissection of the pterigomaseterine band is performed on both sides. All the previously mentioned maneuvers are aimed at decreasing the resistance of the soft tissues and preventing damage to the neurovascular bundle with gradual traction but with a planned magnitude greater than 15 mm.

Both distractors were positioned with their extraoral stems emerging. The turns of both distractors were verified, confirming a target advancement of 8 mm on each side. Both activators of the distractors were closed again to their initial positions. Profuse lavage was performed with physiological saline solution, hemostasis and intraoral sutures with muscle and mucous plane Vicryl 3.0. The surgical procedure ended without complications.

Due to box genioplasty, which allows the suprahyoid musculature to advance, the patient was removed in good general condition and hemodynamically stable without the need for intensive care or prolonged intubation. Following a three-day stay in medical-surgical care, he was granted a medical discharge in optimal general and local conditions.

2.1.4. MDO protocol

Distraction began after a 7-day latency period, activated twice a day at a distraction range of 0.4 mm, achieving 0.8 mm daily until the desired mandibular advancement was achieved. Weekly clinical and imaging controls with panoramic radiography were performed for a month to verify correct and symmetrical opening of the devices.

The distraction period was stopped after 18 mm of mandibular advancement was reached, completing 22 days of distraction. The treatment was halted as great resistance was observed in the distractor's active end in the last turns, considering the clinical and imaging results to be successful according to the preoperative plan. After the weekly controls, monthly evaluations were carried out; a CT was requested in the third, sixth and ninth months to verify the UA increase and facial changes.

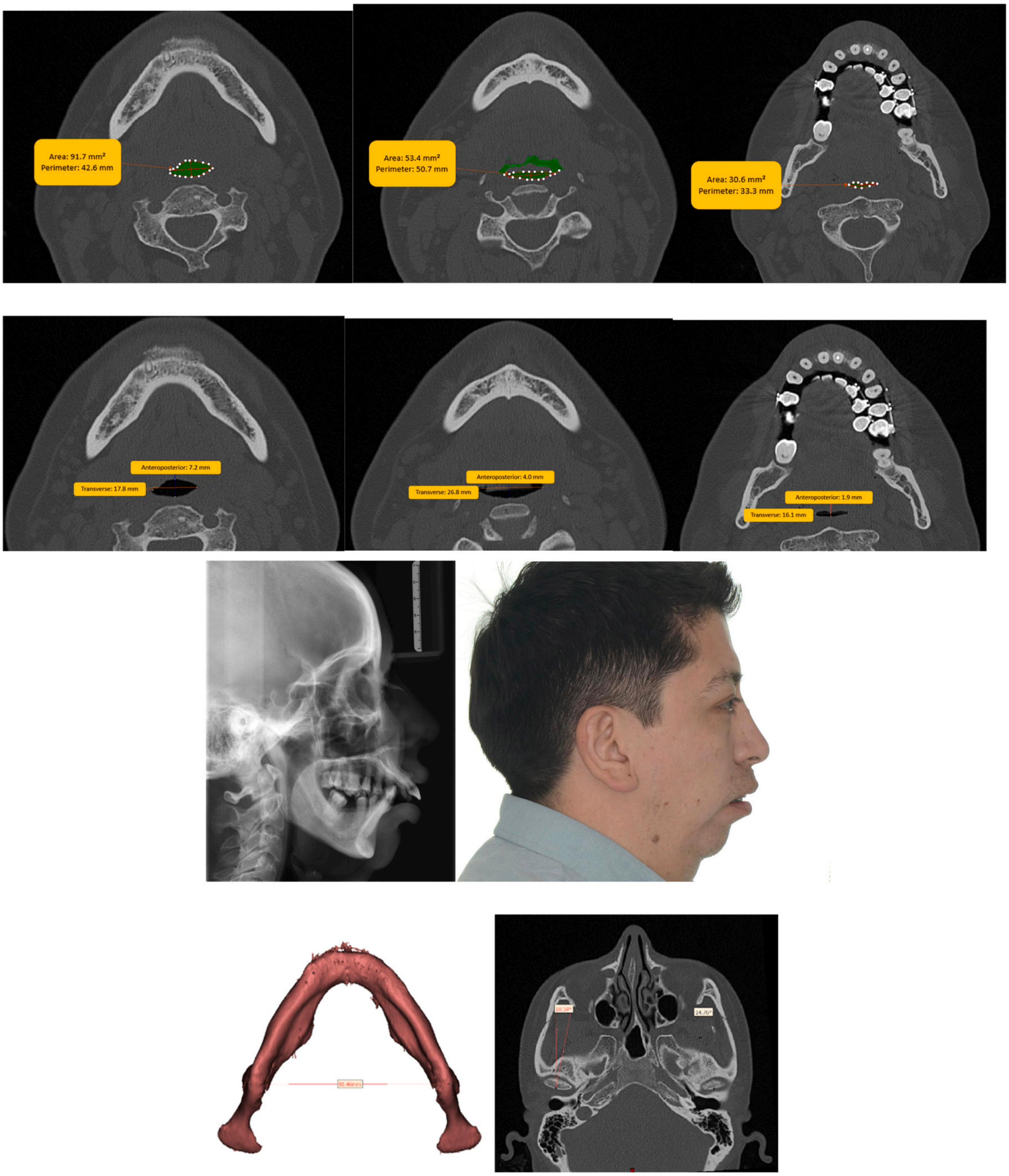

Fig. 3. Pre-surgical airway and physical results. Point B (3.1), pogonion (3.2) and minimum cross-sectional area (MCA) (3.3). Anterior-posterior results in Point B (C.4), pogonion (3.5) and MCA (3.6). Pre-surgical lateral profile teleradiography (3.7) and facial photograph (3.8). Condylar angulation in 3D reconstruction and axial CT section (3.9- 3.10).

For the three-dimensional UA analysis, point B and pogonion were selected, defined as the deepest point of the concavity of the lower alveolar bone and the most prominent or anterior point of the chin contour, respectively. These were used as vertical and sagittal mandibular references, projecting posteriorly into the oropharynx.

2.2. *Systematic review*

2.2.1. *Study design*

A systematic review of the literature was performed according to the PRISMA guidelines (Preferred Reporting Items for Systematic Reviews and Meta-Analyses), under the research question: What are the advantages and disadvantages of performing isolated MDO versus MDO with BSSO plus mandibular symphysis box advancement in patients older than 18 years, diagnosed with OSAHS to optimize the increase in the anteroposterior diameter of the airway?

2.2.2. *Eligibility criteria*

The criteria used in study selection were complete texts translated into English or Spanish, human patients, and adults older than 18 years diagnosed with OSAHS who underwent MDO, regardless of the distractor type.

Understood as OSAHS, according to the American Academy of Sleep Medicine Task Force, as OSAHS characterized by repeated episodes of UA obstruction during sleep, usually associated with sleep disruption and decreased oxyhemoglobin saturation.

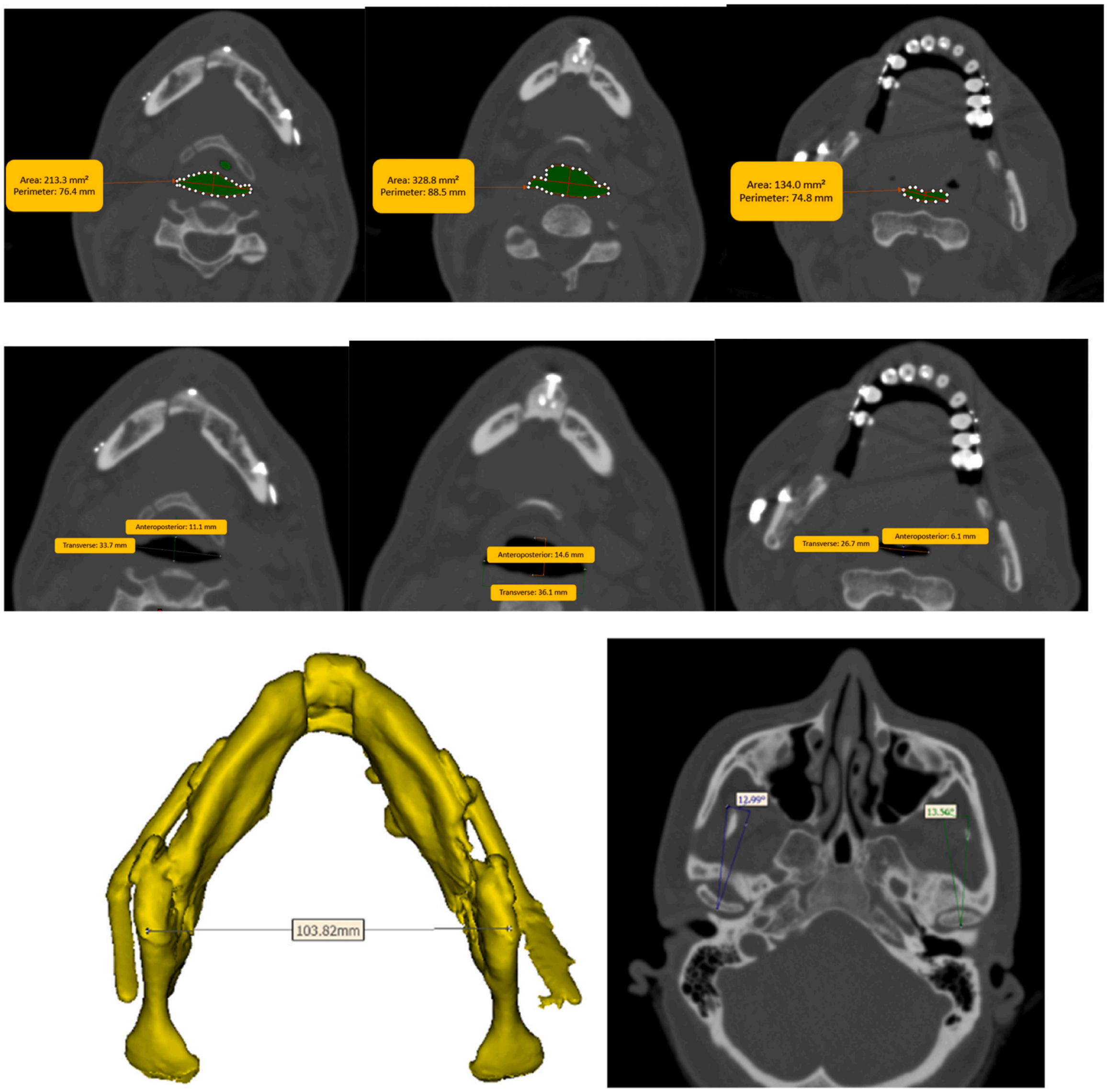

Fig. 4. Post-operative airway results at 3 months. Point B (4.1), pogonion (4.2) and minimum cross-sectional area (MCA) (4.3). Anterior-posterior results in Point B (4.4), pogonion (4.5) and MCA (4.6) at 3 months. Condylar angulation in 3D reconstruction and axial CT section (4.7- 4.8).

The diagnosis of OSAHS is confirmed by polysomnography, where adult apnea is considered a pause in breathing of 10 s or more, and hypopnea a 50 % reduction in airflow for a period equal to or greater than 10 s, associated with a decrease of more than 3 % in oxyhemoglobin saturation and/or a reduced waking state. Apnea is classified as a presentation of five or more respiratory events (apnea and/or hypopnea) per hour of sleep.

Cohort studies, clinical (randomized or non-randomized), prospective, comparative, retrospective, case series and case reports were included, with no restrictions on publication year and follow-up time. Patients and specific data that fit the inclusion criteria were included in cases where the study and the information provided were allowed, and those not within the same study were excluded.

Animal studies, narrative reviews, systematic reviews, and in vitro studies were excluded. In turn, publications that included intervention regions other than the mandible, such as the maxillary, were excluded. Neonatal and pediatric patients and syndromes such as Pierre Robin syndrome, hemifacial microsomia, cleft lip and palate, Cranial Synostosis, Treacher Collins, Crouzon Syndrome, and Alagille Syndrome, among others, were excluded.

2.2.3. Sources of information

To identify potentially relevant articles, the literature databases MEDLINE/PubMed, Cochrane Library and Web of Science were selected. Two authors conducted the research independently between March 25 and May 20, 2024.

2.2.4. Search strategy

According to the protocol described, an electronic search was conducted using the selected databases. The leading search was performed in PubMed, with the following medical subject heading (MeSH) keywords: “("Osteogenesis, Distraction"[Mesh])) AND “Sleep Apnea Syndromes"[Mesh]” (PUBMED, MeSH subject). Cochrane Library and Web of Science searched with the keywords used in PUBMED or their synonyms. Additionally, a search using free and manual terms was performed individually.

2.2.5. Selection of articles

Two reviewers, S.D.A and F.J.P., independently selected the articles. The primary data was exported to the Mendeley reference manager. The two reviewers independently reviewed the titles and abstracts, identifying articles eligible for full review. Disagreements were resolved by

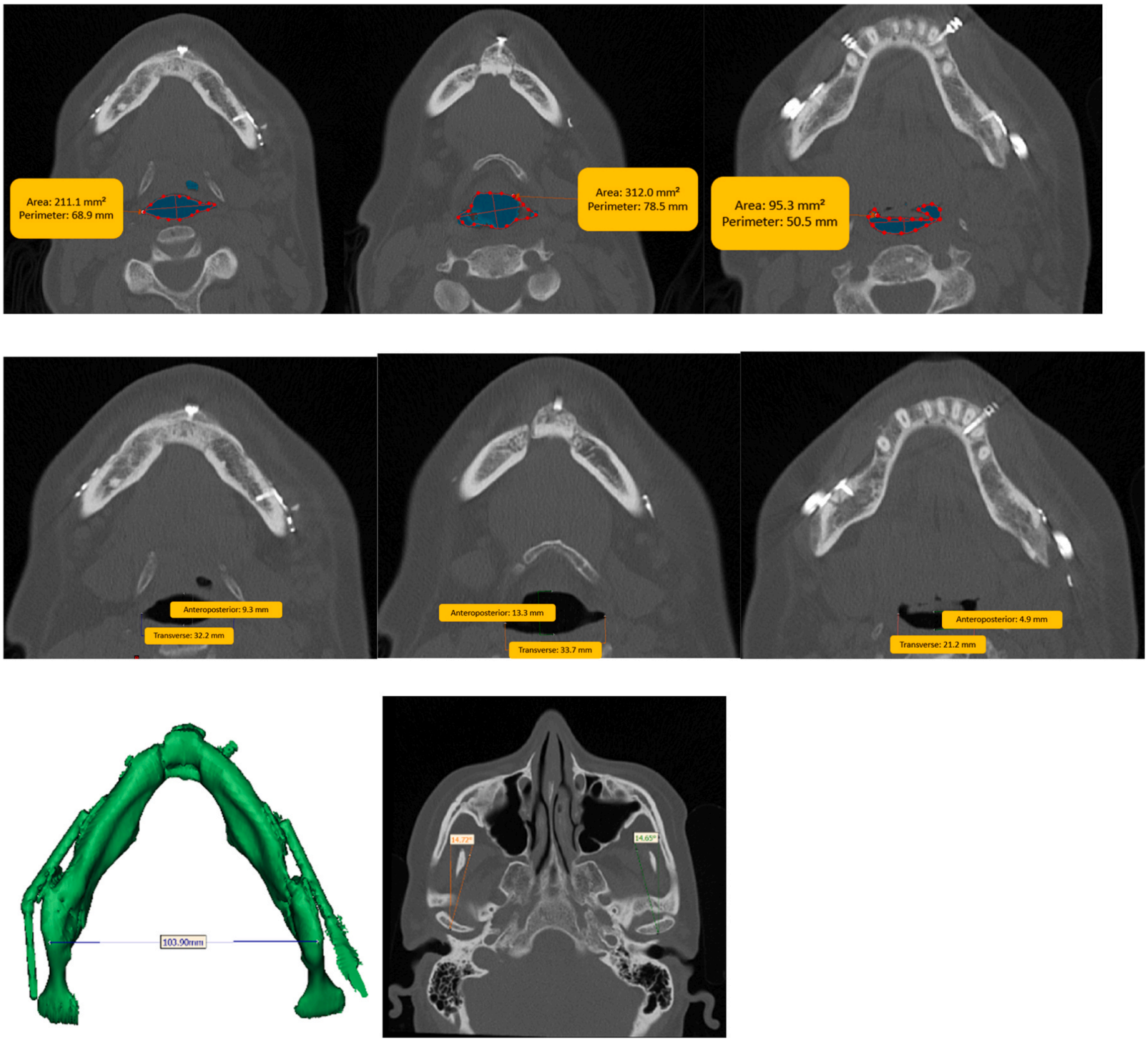

Fig. 5. Post-operative airway results at 6 months. Point B (5.1), pogonion (5.2) and minimum cross-sectional area (MCA) (5.3). Anterior-posterior results in Point B (5.4), pogonion (5.5) and MCA (5.6) at 6 months. Condylar angulation in 3D reconstruction and axial CT section (5.7- 5.8).

consensus and discussion between the two reviewers, along with a third and fourth reviewer who acted as a judge to address any unresolved disagreements.

2.2.6. *Data extraction*

To collect and extract data from the included studies, various variables were considered and tabulated using Microsoft Excel, and the results were presented in detail in tables and figures.

3. Results

3.1. *Case report*

Various parameters were analyzed to evaluate mandibular dimensional changes and obtain different values between the preoperative and postoperative controls (three, six, and nine months) (Figs. 3–6). Preoperative and 9-month postoperative polysomnography and cephalometry were made to evaluate and compare the parameters of AHI, lowest oxygen saturation (LSAT) and Sella-nasion-B point (SNB) angle (SN and NB plans). The results of this angle in the preoperative and postoperative examinations at three and six months were 74º, 82º and 81°, respectively, finally increasing by 7º (which was maintained at the 9-month control). Bilateral mandibular body advancement achieved an airway increase, improving the AHI from 32 to 9, 13 and 12 (pre-surgery and 3-, 6- and 9-month control). LSAT from 82 % to 96, 94 and 93 % (pre-surgery and 3-, 6- and 9-month control, respectively) (Table 1).

The gains at 3 months in anteroposterior airway were 54 % in B point, 265 % in Pogonion, and 221 % in MCA. These relapsed in the ninth month in 19.8, 30, and 19.6 %, respectively (for B point, Pogonion, and MCA). The changes in the airway area showed an increase at 3 months of 132 % in B point, 515 % in Pogonion, and 337 % in CMA. Like the anteroposterior dimension, they relapsed at 16.4, 29.1, and 53.1 %, respectively (for B point, Pogonion, and MCA) (Table 2)

In the analysis of bone advancement, the distraction gap achieved at 3 months was an average of 18.4 mm (right segment: 18.6 mm/left: 18.3 mm). This advancement was maintained without significant changes in the 9-month CT control (right segment: 18.5 mm/left: 17.8 mm) (Fig. 7). The authors of this study believe, based on the agreement of evidence from bone measurements, that the relapse does not belong to the mandibular advancement bone but to the soft tissue (Figs. 8 and 9).

Condylar angulation is represented in (Figs. 3.9, 3.10, 4.7, 4.8, 5.7, 5.8, 6.9-6.10), in 3D reconstruction and axial CT sections for the preoperative state and at 3, 6, and 9 months postoperatively, and is summarized in "Table 2 – Condylar Angulation". The condylar angulation change was less than 5°, with both condyles remaining in their position within the fossa in all three spatial directions—axial, coronal, and sagittal—during the 9-month postoperative follow-up.

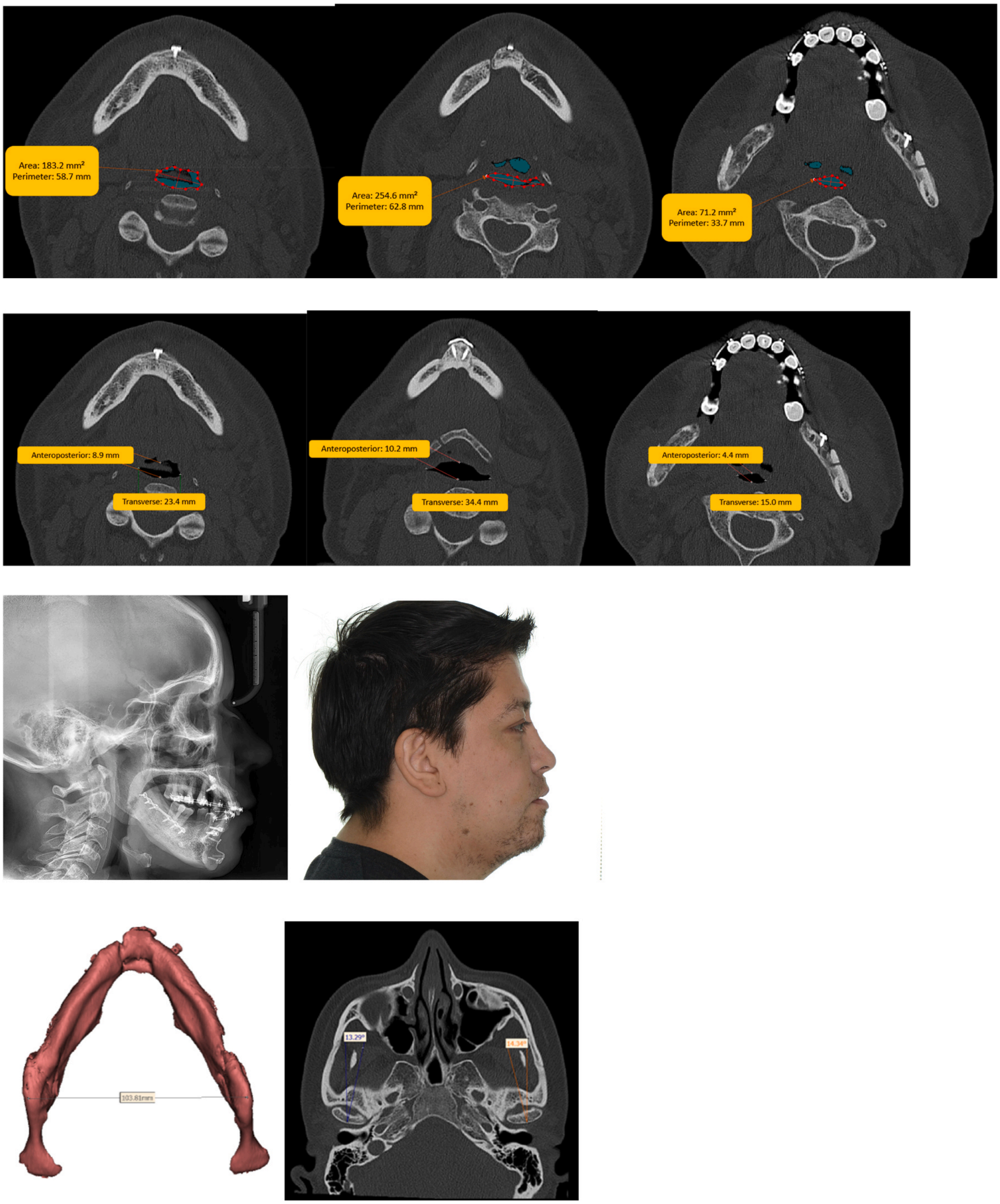

Fig. 6. Post-operative airway and physical results at 9 months. Point B (6.1), pogonion (6.2) and minimum cross-sectional area (MCA) (6.3). Anterior-posterior results in Point B (6.4), pogonion (6.5) and MCA (6.6) at 9 months. Post-operative lateral profile teleradiography (6.7) and facial photography (6.8). Condylar angulation in 3D reconstruction and axial CT section (6.9- 6.10).

Regarding immediate complications, the patient experienced post-operative oedema and localized pain, which are common symptoms of this surgical procedure, both with complete resolution. As for late complications, only a slight expected mandibular relapse of minimal magnitude was observed.

The patient continues attending his respective controls, with a follow-up period of 9 months to date. As determined by the surgeon, the duration will be long-term.

In the third postoperative month, a light elastic on each side was used for two weeks to improve occlusal conditions. Once the orthodontic treatment progresses and the alignment and dental malocclusion allow optimal results, orthognathic surgery will be planned. Figs. 3.7–8 and 6.7–8 show the pre-surgical and 9-month post-operative lateral profile teleradiograph and facial photographs, demonstrating the physical changes and aesthetic results.

3.2. Systematic review

In the initial identification process, 1628 potential articles were found for review, of which 152 duplicates were removed from the databases. Thus, 1476 publications were subjected to an in-depth review of titles and abstracts, resulting in 85 potential manuscripts for full-text

Table 1
Pre-surgical and post-surgical mandibular airway changes at 3, 6 and 9 months.

	AHI (/h)	LSAT (%)	SNB (°)	B Point				Pogonion				Minimum cross-sectional area (MCA)			
				Area	Perimeter	Antero-posterior	Transversal	Area	Perimeter	Antero-posterior	Transversal	Area	Perimeter	Antero-posterior	Transversal
Pre-surgical	32	82	74	91.7 mm2	42.6 mm	7.2 mm	17.8 mm	53.4 mm2	50.7 mm	4.0 mm	26.8 mm	30.6 mm2	33.3 mm	1.9 mm	16.1 mm
Post - surgical (3 months)	9	96	82	213.3 mm2	76.4 mm	11.1 mm	33.7 mm	328.8 mm2	88.5 mm	14.6 mm	36.1 mm	134.0 mm2	74.8 mm	6.1 mm	26.7 mm
Post - surgical (6 months)	13	94	81	211.1 mm2	68.9 mm	9.3 mm	32.2 mm	312.0 mm2	78.5 mm	13.3 mm	33.7 mm	95.3 mm2	50.5 mm	4.9 mm	21.2 mm
Post - surgical (9 months)	12	93	81	183.2 mm2	58.7 mm	8.9 mm	23.4 mm	254.6 mm2	62.8 mm	10.2 mm	34.4 mm	71.2 mm2	33.7 mm	4.4 mm	15.0 mm

Abbreviations: AHI: apnoea-hypopnea index; LSAT: lowest oxygen saturation; SNB: sella-nasion-b point angle; MCA: Minimum cross-sectional area.

evaluation. Applying the exclusion and inclusion criteria excluded 81 articles, leaving 4 for analysis (Fig. 10). No additional studies were identified through manual searching. Of the included articles, two were prospective studies and two were case reports. The articles included a total of 31 patients who underwent MDO. The demographic data and patient outcomes are described in Tables C and D. Of the total sample size of 31 patients, 65 % were male (20 patients) and 35 % female (11 patients), with an average age of 34.45 years and a standard deviation of 13.98. In 100 % of the studies, the patients had OSAHS, 75 % had mandibular micrognathia and 50 % had temporomandibular joint ankylosis (Table 3).

Regarding MDO, an intraoral distractor was used in 75 % of the cases, and an extraoral distractor was used in 25 %. The average latency period was 5.75 days, with a distraction rate of 1 mm/day in 48.4 % and 0.8 mm/day in 51.6 % of the cases.

Advancement is defined as the gain in newly formed bone tissue resulting from applying traction forces provided by osteogenic distraction devices. The average mandibular advancement achieved was 11.06 mm, within a range of 5.5–18 mm.

The distractor size variable will determine the size in millimeters of the opening of the osteogenic distraction device, whether the distraction system is of the intraoral or extraoral type, which has an average of 14.63 mm within a range of 10–28 mm.

The average AHI, identified in two studies, decreased from approximately 30.68 to 3.62, with an 88 % improvement, indicating a significant reduction in the frequency of apnea and hypopnea events. The RDI, identified in two studies, decreased from approximately 45.8 to 3.57, with a 92 % improvement. The LSAT increased from approximately 82.02–92.43 %, increasing by 10.41 %, indicating improved oxygenation during sleep. The SNB angle, defined as the angle formed by the S-N plane and N-B point, indicates the anteroposterior relationship of the mandible to the skull. This angle was identified in two studies and increased from approximately 68.05° to 73.45°, resulting in an increase of 5.4° and a 7.94 % improvement.

Complications such as temporomandibular joint symptoms, such as pain and noises, were associated in 50 % of the studies. Infections were associated with 50 %, and distractor failure and alveolar nerve paresis in 25 %. The average follow-up of the four studies was 19.09 months (Table 4).

No study has been found that combined MDO, BSSO, and the box genioplasty technique.

4. Discussion

OSAHS is a public health challenge as it is a highly underdiagnosed condition despite its high prevalence, affecting approximately 2 %–4 % of the adult population worldwide [1]. During sleep, effects influence the permeability of the UA and ventilatory control, while a reduction in the electrical activity of the medullary neurons of the abductor muscles is also observed. The activity of the genioglossus muscle decreases, leading to tongue descent and, consequently, airway obstruction in individuals with anatomical abnormalities in the airways [1,35].

This syndrome is often associated with narrow UA, increasing the risk of collapse during sleep and usually manifests in adults with mandibular retrognathia, resulting in a convex profile (Class II) and a short distance between the chin and the posterior cervical region, indicating the need for interventions to correct micrognathia associated with OSAHS [13,23].

Several surgical procedures, specifically osseous treatments such as maxillomandibular osteotomy (MDO), MMA, genioplasties, orthognathic surgery or a combination of these, have been proposed to correct craniofacial anomalies associated with OSAHS [16].

The purpose of this study is to present the combination of surgical techniques in the mandible: bilateral sagittal split ramus distraction plus box genioplasty to optimize the increase in the anteroposterior diameter of the airway as initial treatment in an adult patient with OSAHS and

Table 2
Mandibular dimensional changes pre-surgical and post-surgical at 3, 6 and 9 months; including soft tissue measurements, anterior tubercle of the hyoid to the mandibular plaxne.

	Pre-surgical		Post-surgical (3 months)		Post-surgical (6 months)		Post-surgical (9 months)	
	Right	Left	Right	Left	Right	Left	Right	Left
Condyle - Angle	56 mm	56.7 mm	55.2 mm	55.6 mm	55.32 mm	53.47 mm	53.1 mm	53.2 mm
Angle - Coronoid	60.8 mm	64.6 mm	63.3 mm	63.9 mm	60.87 mm	63.69 mm	60.1 mm	62.5 mm
Cóndilo - Spix	35.6 mm	34.6 mm	41.3 mm	38.1 mm	41.44 mm	40.2 mm	40.2 mm	40 mm
Coronoid - pregonial	68.4 mm	72.5 mm	74.2 mm	78.7 mm	73.93 mm	78.23 mm	73 mm	78.2 mm
Condyle - molar distal	64.2 mm	58.1 mm	78.7 mm	75.3 mm	78.64 mm	75.3 mm	75.1 mm	73.9 mm
GAP distraction	-	-	18.6 mm	18.3 mm	18.5 mm	18.1 mm	18.5 mm	17.8 mm
Hyoid - Mandibular plane	9.5 mm		5.3 mm		5.4 mm		7.4 mm	
Condylar angulation	10.59º	14.76º	12.99º	13.56º	14.72º	14.65º	13.29º	14.34º

skeletal class II dentofacial anomaly.

In this systematic review of the four selected studies, the evaluated variables were AHI and LSAT, which showed improvements of 88 and 10.41 %, respectively. This case report analyzed the same variables, obtaining a percentage improvement of AHI (from severe to mild category) and saturation enhancement of 11 %, respectively (the nine months).

Three studies describe surgical complications, mainly moderate pain, mechanical failure of the distractors, inflammation and local infection, paresthesia of the inferior alveolar nerve, occlusal alterations and discomfort of the temporomandibular joints such as joint noises. All were temporary except for bilateral condylar resorption and mandibular relapse, which were reported in only one study.

The literature indicates that major and minor complication rates for MMA were 10 % and 31 %, respectively, [36]. As for infection at the distractor site and distractor failures, these could be mitigated by improving the design, manufacture, and handling of the distractors and implementing methodical cleaning measures by patients or their caregivers. Removing distraction devices should follow specific protocols to minimize the risk of postoperative complications such as infections or relapses.

Following an osteotomy, a gradual tension is applied to the callus, connecting the separated bone segments using an external or internal fixation device, thus elongating the bone tissue [37]. The device is suggested to be removed in the presence of mature and healthy bone after the consolidation period [37,38]. In maxillofacial OD, the average consolidation period is 8–12 weeks; in long bone OD, the average healing index (days/cm) is 20–30 weeks [39].

In this case report, a period of 6 months was considered for removal, and immediate complications such as localized pain and oedema and a late relapse were reported. Regarding the pre-and postoperative dimensional changes in the 6 months control, the results were condyle-angle -0.68 mm and -3.23 mm, angle-coronoid $+0.07$ mm and -0.91 mm, condyle-Spix $+5.84$ mm and $+5.6$ mm, coronoid-regional $+5.53$ mm and $+5.73$ mm, condyle-distal molar $+14.4$ mm and $+17.2$ mm, respectively. To summarize, the postoperative dimensional changes at 6 months compared to the preoperative state were $+25.16$ mm on the right and $+24.39$ mm on the left.

The average follow-up in the selected studies was 19.09 months, compared to this case report, where a 9-month follow-up was conducted.

Long-term follow-up is crucial in assessing the durability of these interventions, particularly considering the high relapse rates observed in some mandibular surgeries. Extended monitoring allows for a comprehensive evaluation of the structural and functional stability of the surgical outcomes, providing insight into the long-term efficacy of combined techniques like MMA and BSSO.

Among the limitations in the search for studies for this review, it was found that most studies on patients with OSAHS treated with mandibular OD were conducted in pediatric populations, representing a significant percentage of patients with syndromic or craniofacial deformities such as Pierre Robin syndrome, hemifacial microsomia, cleft lip and palate, craniosynostosis, Treacher Collins syndrome, Crouzon syndrome and Alagille syndrome, among others. Due to the continuous growth of the craniofacial complex in pediatric patients, mandibular OD is one of the first-line treatments to increase the airway in OSAHS, with orthognathic surgery rarely performed in this population.

Another limitation lies in the varied methods of airway measurement used to evaluate the success or failure of a surgical intervention in the literature search.

A narrow and elongated airway, as observed through CT imaging, is strongly associated with increased collapsibility, emphasizing the importance of evaluating turbulence in airway dynamics rather than solely measuring expanded airway volume.

The lack of standardized methods makes comparing success between studies difficult, as each study may use different criteria and evaluation techniques. This methodological variability hinders the uniform and accurate evaluation of results, complicating comparisons between studies and data synthesis and drawing definitive conclusions about treatment efficacy.

In treating patients with class II mandibular hypoplasia, both mandibular OD and BSSO are used to achieve immediate and progressive mandibular advancement. However, mandibular OD has primarily been applied to patients requiring large advancements ($\geq$10 mm), proving stable over time. By contrast, BSSO has been associated with a higher incidence of relapses in advancements $>$ 6 mm [40].

The box advancement or genioplasty technique consists of displacing the suprahyoid musculature anteriorly, allowing it to be associated with conventional mandibular advancement techniques such as BSSO to achieve airway expansion through a complete block advancement.

This technique ensures immediate airway patency during the latency period of the OD, mitigating risks of obstruction, and serves as a vital complement to mandibular OD and BSSO.

In this case, the mandibular osteogenic distractor was maintained for six months, ensuring sufficient consolidation of the newly formed bone tissue. After removing the distractor, a single osteosynthesis plate stabilized the left mandibular ramus. This approach facilitated a stable occlusion, contributing to a reduced osseous relapse rate of approximately 12 %. These results underscore the importance of adequate stabilization techniques post-distraction. Incorporating a method for forward movement and fixation, such as sagittal split ramus osteotomy (SSRO) combined with osteosynthesis, demonstrates a viable strategy to enhance the predictability of mandibular positioning outcomes. The indication for box genioplasty has already been detailed within this manuscript. This technique serves as an initial intervention to increase the diameter of the oropharyngeal airway during the latency period of the OD, which spans seven days post-surgery, when distractors are not yet activated. The box genioplasty ensures critical airway expansion, mitigating risks of airway obstruction during this latency phase.

Additionally, mandibular advancement procedures, such as maxillomandibular advancement (MMA), are generally indicated for cases with significant airway obstruction and pronounced craniofacial anomalies. Box genioplasty, as a complementary technique, offers immediate improvements in airway patency, distinguishing its application

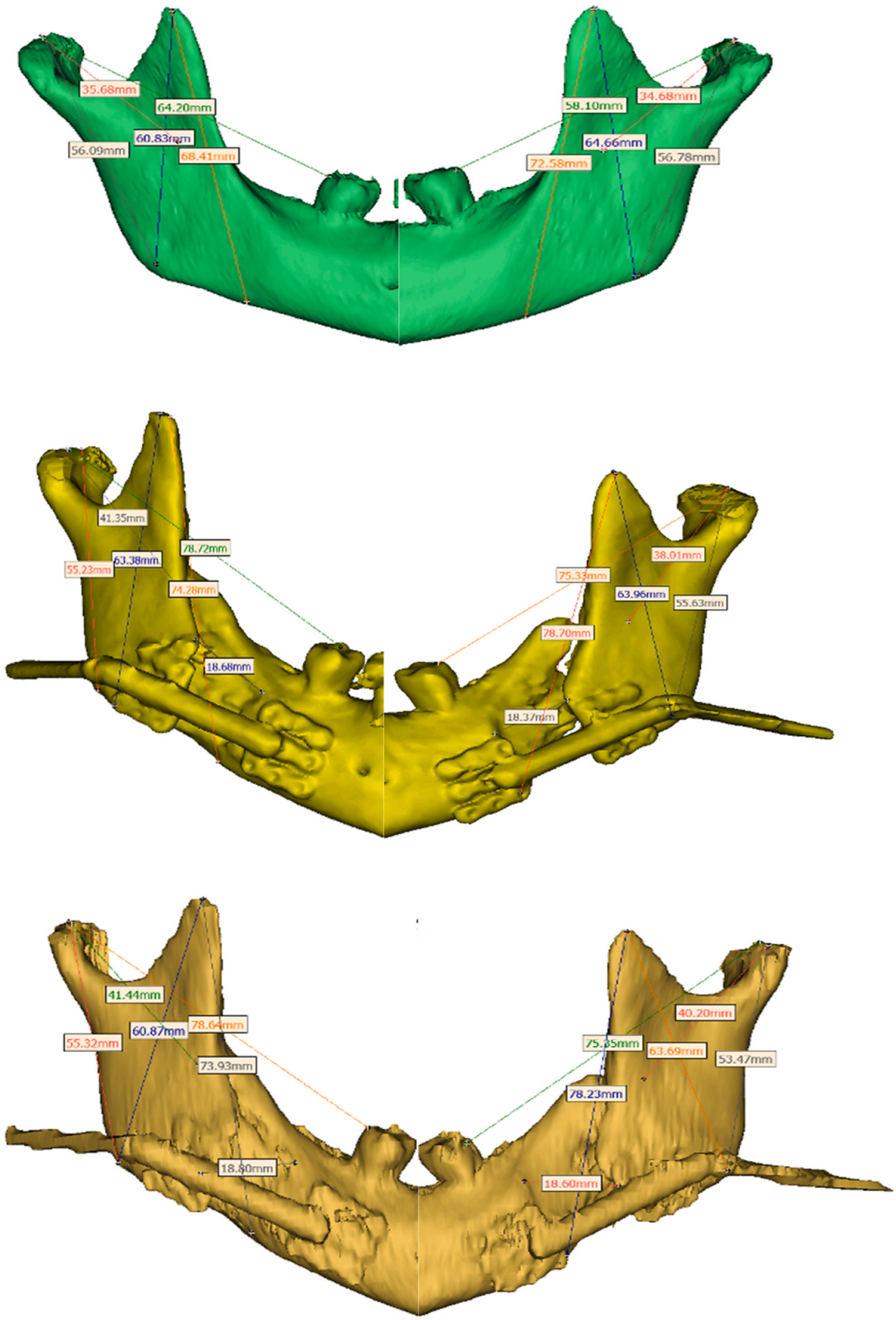

Fig. 7. 3D pre-surgical reconstruction (7.1 green), post-surgical reconstruction at 3 months (7.2 yellow) and post-surgical reconstruction at 6 months (7.3 orange) showing dimensional mandibular changes.

from broader indications of MMA. This distinction highlights the tailored use of each surgical technique, emphasizing that genioplasty's role is not merely supplementary but essential in addressing specific airway and structural requirements during the early stages of treatment.

To optimize OD results and minimize relapse risk, a technique that triangulates vectors and distributes traction forces more effectively is proposed. This study performed a bilateral sagittal split ramus distraction technique plus box genioplasty.

The purpose of complementing Bilateral Sagittal Split ramus OD with a box genioplasty lies in its capacity to achieve an immediate increase in the anteroposterior airway diameter. This advancement of the genial process and its associated musculature, provides an advantage not achievable with OD alone, contributes to reducing the risk of airway collapse and obstruction. Consequently, it permits the patient's post-surgical extubation without requiring admission to intensive care units, which optimises post-operative management and significantly

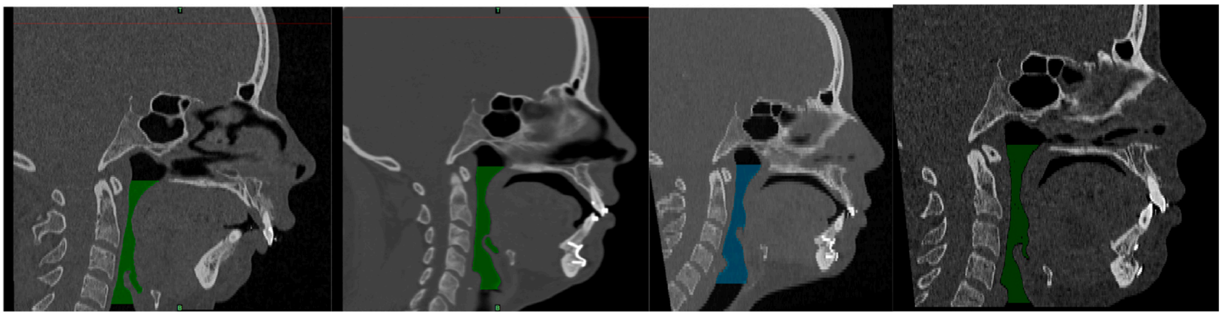

Fig. 8. CT in sagittal section showing comparative pre-surgical airway (8.1) and post-surgical airway at 3 (8.2), 6 (8.3) and 9 months (8.4).

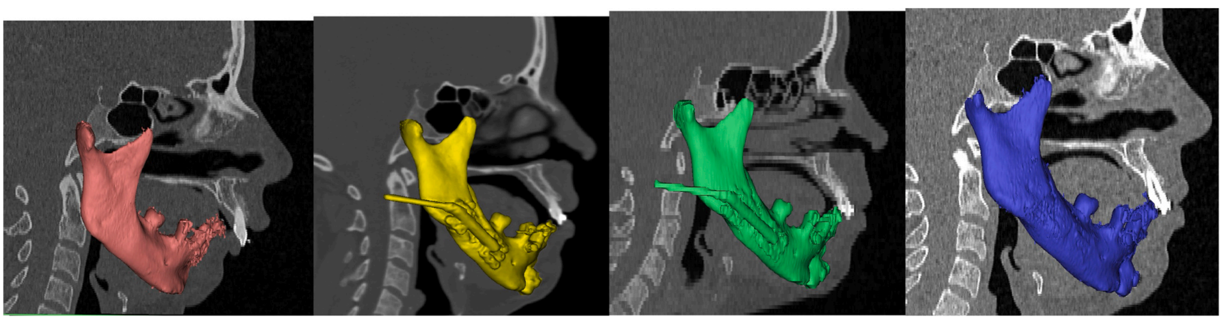

Fig. 9. CT 3D reconstruction in sagittal section showing comparative pre-surgical jaw (9.1) and post-surgical jaw at 3 (9.2), 6 (9.3) and 9 months (9.4).

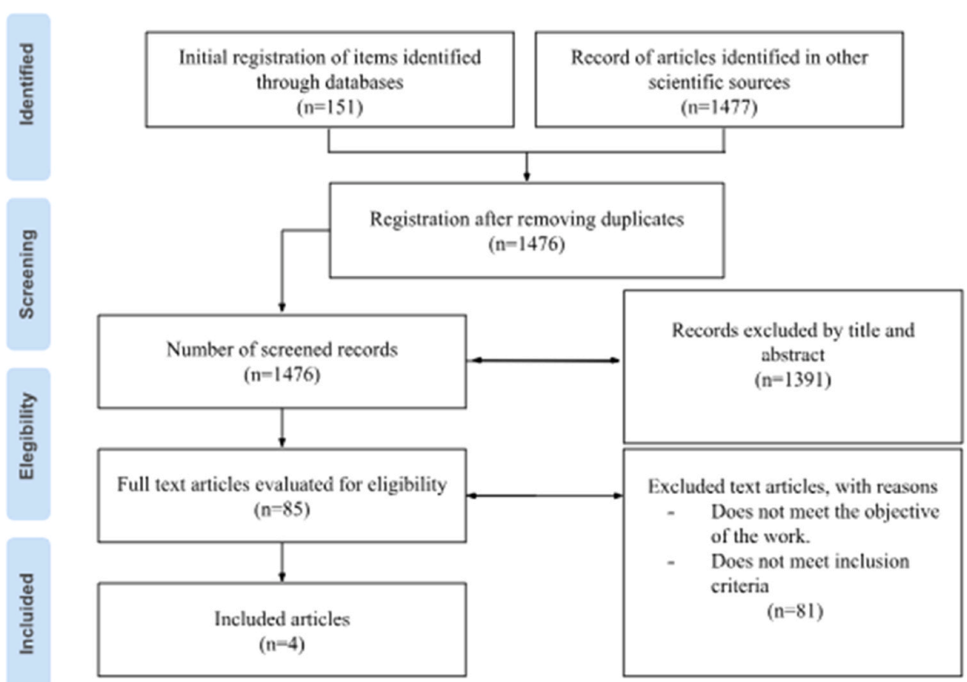

Fig. 10. PRISMA flow diagram for article selection.

Table 3
Demographic data of the studies included in the final review.

Author	Year	Type of study	Sample size	Age (years)	Gender	Morbid history
Li et al.	2002	Prospective	3	58 (51–68)	1 F, 1 M	OSAHS
Hamada et al.	2007	Case report	1	31	M	OSAHS, mandibular micrognathia, Class II
Feiyun et al.	2010	Case report	16	27 (18–43)	6 F, 10 M	OSAHS, bilateral TMJ ankylosis, mandibular micrognathia
Andrade et al.	2018	Prospective observational	11	21.81 (18–26)	N/A	OSAHS, uni/bilateral TMJ ankylosis, mandibular micrognathia

Abbreviations: F: Female, M: Male, N/A: Not available, OSAHS: Obstructive sleep apnoea and hypopnea syndrome.

Table 4
Results of the studies included in the final review.

Author	Type de distractor	Latency period (days)	Distraction range (mm/day)	Advance (mm)	Distractor size (mm)	AHI (/h)		RDI		LSAT (%)		SNB (°)		Complications		Follow up (months)
						Pre - s	Post - s	Pre - s	Post - s	Pre - s	Post - s	Pre - s	Post - s	Temporary	Permanent	
Li et al.	Extraoral	7	1	5.83 (5.5–6.0)	10	N/A	N/A	44.3 (25.9–69)	5.03 (0–11.6)	83.67 (80–87)	87.67 (83–90)	N/A	N/A	Inflammation and local infection, TMJ discomfort, moderate pain, alveolar nerve paresthesia	No	10,67 (6–18)
Hamada et al.	Intraoral	5	1	18	N/A	29.9	5.8	N/A	N/A	77	87	67.4	69.3	Moderate bilateral TMJ pain, joint noises, sensitivity/pain and difficulty opening, occlusal alterations	Bilateral condylar resorption, mandibular relapse	12
Feiyun et al.	Intraoral	7	0.8	N/A	18.9 (12–28)	N/A	N/A	47,3	2.1	75.4	98.2	68.7	77.6	No	No	29,7
Andrade et al.	Intraoral	4	1	9.36	15	31.45	1.43	N/A	N/A	92.01	96.84	N/A	N/A	Distractor failure, intraoral distractor infection	No	24

Abbreviations: AHI: apnoea-hypopnea index; RDI: respiratory alteration index; LSAT: lowest oxygen saturation; SNB: sella-nasion-b point angle; Pre/Post-s: Pre/Post-surgical.

reduces hospitalisation days. Studies demonstrate significant mandibular advancements with OD, obtaining significant results in AHI, LSAT and SNB, but when compared with the OD through BSSO combined with genioplasty, even greater mandibular advancement can be achieved, allowing for greater airway stabilization, demonstrating a stable and effective technique.

A two-dimensional cephalometric analysis of Delaire was also performed between preoperative and postoperative results. The posterior tilt concerning F2 and F3 significantly improved the mandibular ramus position, and concerning F1, the bony chin improved in its anteroposterior position, advancing considerably along with the airway.

In a future case, it would be ideal to perform a CT one week postoperatively before the distractors are removed to quantify the immediate airway advancement achieved with the box technique without the immediate postoperative inflammation of the surgery.

5. Conclusions

The technique of MDO by BSSO and box genioplasty in this OSAHS and skeletal dentofacial abnormality class II report achieves a decrease of the AHI classification from severe to mild, with a final relapse in anteroposterior airway segment of 19.8 % in B point, 30 % in Pogonion and a 19.6 % in CMA; without mandibular bone retrusion. Using the triangular vector achieved with two posterior osteotomies (right and left) and one anterior and medial osteotomy allows for a box advancement, becoming an excellent therapeutic option for this type of pathology.

Contributions from the authors

All authors contributed to the design of the study. S.D.A. D. and F.J. P. prepared the materials, collected and analysed the data. Disagreements were resolved by consensus and discussion with a third and fourth reviewer, C.R.A. and P.T.C., who acted as judges to resolve the disagreements. All authors read and approved the final manuscript.

Consent to participate

The use of the clinical case with its respective images is supported by the informed consent obtained and signed by the patient included in this study.

Consent to publication

The authors state that the patient participating in the human research gave informed consent for the publication of the images in Figs. 1–6.

Ethical approval – statements

None

Ethics

This study was conducted in accordance with the principles of the Helsinki Declaration, ensuring the patient's identity remained anonymous.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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