รายงานผู้ป่วย

The first two cases of mandibular osteotomy and genioglossus advancement for Sleep-disordered Breathing (SDB) in Thailand

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Sleep - disordered Breathing (SDB) consists of primary or habitual snoring, upper airway resistance syndrome (UARS) and obstructive sleep apnea syndrome (OSAS). Treatment of SDB consists of weight reduction, modification of body position during sleep, positive airway pressure treatment (CPAP) and surgery. Although CPAP is the gold standard of treatment, the patients' compliance to use this equipment is poor due to many factors.

Site-specific surgery is the alternative and highly effective treatment for SDB. Uvulopalatoplasty is the popular procedure for treatment of SDB but is effective only for retropalatal obstruction cases. If the obstruction sites are located at retrolingual or combined retropalatal and retrolingual levels, hypopharyngeal surgery is necessary.

The first two cases of Mandibular Osteotomy and Genioglossus Advancement for SDB in Thailand are reported here in.

Key words : *Sleep-disordered Breathing (SDB), Obstructive Sleep Apnea Syndrome (OSAS), Mandibular Osteotomy and Genioglossus Advancement, Hypopharyngeal Surgery.*

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ประกอบเกียรติ หิรัญวิวัฒน์กุล. รายงานผู้ป่วย 2 คนแรกในประเทศไทยที่ได้รับการผ่าตัดรักษาเพื่อแก้ไขความผิดปกติของการหายใจขณะนอนหลับ โดยวิธีผ่าตัดกระดูกขากรรไกรล่างและดึงกล้ามเนื้อลิ้นมาทางด้านหน้า. จุฬาลงกรณ์เวชสาร 2543 ก.ค; 44(7): 525 - 35

ความผิดปกติของการหายใจขณะนอนหลับ เป็นกลุ่มอาการอันประกอบด้วย การนอนกรนธรรมดา (Primary or Habitual Snoring) กลุ่มอาการทางเดินหายใจส่วนบนมีความต้านทานสูง (Upper Airway Resistance Syndrome or UARS) และกลุ่มอาการหยุดหายใจจากทางเดินหายใจอุดตันขณะนอนหลับ (Obstructive Sleep Apnea Syndrome or OSAS). การรักษาความผิดปกติทั้งหมดนี้ประกอบด้วย การลดน้ำหนัก การจัดท่าการนอนให้ถูกต้อง การใช้เครื่อง CPAP (Continuous Positive Airway Pressure treatment) และการผ่าตัด ถึงแม้ว่า CPAP จะเป็นการรักษาที่ดีที่สุดในปัจจุบัน แต่ยังมีปัญหามากมายที่เป็นอุปสรรคในการใช้เครื่อง

การผ่าตัดเพื่อแก้ไขภาวะอุดตันของทางเดินหายใจที่ตรงกับตำแหน่งที่เป็นปัญหาจริงๆ เป็นวิธีที่ได้ผลดีไม่แพ้การใช้เครื่อง CPAP การผ่าตัดเพื่อลดขนาดของเพดานอ่อนและลิ้นไก่ (Uvulopalatoplasty) เป็นวิธีที่ได้รับความนิยมและได้ผลดีในกรณีที่การอุดตันอยู่เฉพาะที่บริเวณหลังเพดานอ่อน ถ้าการอุดตันของทางเดินหายใจเกิดที่ระดับหลังโคนลิ้นหรือที่ระดับหลังโคนลิ้นร่วมกับระดับหลังเพดานอ่อน การผ่าตัดรักษาจำเป็นต้องใช้วิธีการผ่าตัดเพื่อเพิ่มเนื้อที่ของทางเดินหายใจบริเวณคอหอยส่วนล่าง

รายงานฉบับนี้ ได้รายงานผู้ป่วย 2 คนแรกในประเทศไทยที่ได้รับการผ่าตัดรักษาเพื่อแก้ไขความผิดปกติของการหายใจขณะนอนหลับ โดยวิธีผ่าตัดกระดูกขากรรไกรล่างและดึงกล้ามเนื้อลิ้นมาทางด้านหน้า

Sleep - disordered Breathing (SDB) consists of primary or habitual snoring, upper airway resistance syndrome (UARS) and obstructive sleep apnea syndrome (OSAS). The presenting symptoms of OSAS are chronic disruptive snoring, choking during sleep, apneas observed by a bed partner, frequent awakening, restless sleep, excessive daytime sleepiness, morning headache or an unrefreshed feeling upon awakening and poor cognitive functions.[1] The diagnosis of OSAS requires the presence of elevated apnea and hypopnea index (AHI or RDI). When untreated, significant OSAS and UARS are associated with increased mortality largely due to cardiovascular causes (hypertension, coronary heart diseases, stroke) [2-7] and automobile accidents.[8] The diagnosis of UARS requires nocturnal polysomnography with intraesophageal pressure monitoring and a complaint of excessive daytime sleepiness.[9-12] Asian patients with OSAS seemingly have greater severity of their illness compared to Caucasian patients matched for age, gender and body mass index (BMI).[13]

Case report

Case 1.

A 36-year-old male patient presented with symptom of excessive daytime sleepiness (EDS) with snoring for two years. The eight questions of the Epworth Sleepiness Scales were asked.

Watching TV = 2

Sitting and reading = 1

Sitting, inactive in a public place (theater, meeting) = 1

Sitting and talking to someone = 0

In a car, while stopped for a few minutes in traffic = 3

As a passenger in a car for an hour without a break = 3

Sitting quietly after a lunch without alcohol = 2

Lying down to rest in the afternoon when the circumstances allow =3

Total score = 15

In addition to EDS, the patient had problems of sleep fragmentation, choking during sleep, abnormal motor movement during sleep, nocturnal enuresis, poor cognitive functions, and an unrefreshed feeling on awakening.

Physical examination showed :

Blood pressure = 120/80

Neck circumference = 13 inches

Body weight = 57.0 kg , height = 1.60m meter, BMI = 22.27 kg/m^2

The facial contour was assessed and slight retrognathia and micrognathia was found

Slight deviated nasal septum and slight hypertrophied interior turbinates but adequate nasal airflow

Normal adenoid and palatine tonsils

Moderately redundant palatal mucosa and long uvula

Mildly redundant lateral pharyngeal wall

Moderately large base of tongue

Fiberoptic Nasopharyngoscopy and Muller's maneuver was done and showed 50% obstruction at the level of the retropalatal and retrolingual areas. Protrusion of the mandible improved the visualization of the laryngeal inlet

A Standard Nocturnal Polysomnography was done. Polysomnography with the Sensormedics system was used. Electroencephalogram (EEG) of the

international electrode placement was recorded on C3/A2, C4/A1, O1/A2 and O2/A1. Electrooculogram (EOG) was recorded with ROC/A1 and LOC/A2. Electromyogram (EMG) was recorded at the chin and both legs. Electrocardiogram (ECG) was done on modified V-2 leads. Respiration was investigated by oronasal airflow, thoracic and abdominal movements (inductive plethysmography), snoring sounds taped on the neck and oxygen saturation (pulse oximetry). The record was automatically scored and manually rechecked following the Rechtschaffen and Kales[44] international criteria for sleep/wake determination, and abnormal breathing patterns were also automatically scored and manually rechecked using the current criteria for identifying sleep apnea and hypopnea.

Sleep study result showed total sleep time = 284 minutes, sleep time efficiency = 95%, apnea index = 1.70, respiratory disturbance index = 1.90, and lowest oxygen saturation = 93 %, but there were frequent episodes of arousal and paradoxical movement of the chest and abdomen. The interpretation of this study was Upper Airway Resistance Syndrome.

The patient refused CPAP treatment and wanted surgery to be conducted.

The surgery was scheduled for Uvulopalatopharyngoplasty (including tonsillectomy), Mandibular Osteotomy and Genioglossus Advancement. The procedure is described in detail here in later in Surgical Technique. No complication occurred during or after the surgery. The surgical result was assessed clinically. Improvement of excessive daytime sleepiness was achieved and snoring sounds disappeared postoperatively.

Case 2.

A 45-year-old female patient presented with loud snoring for 2 years. The eight questions of the Epworth Sleepiness Scales were asked.

Watching TV = 0

Sitting and reading = 0

Sitting, inactive in a public place (theater, meeting) = 0

Sitting and talking to someone = 1

In a car, while stopped for a few minutes in traffic = 0

As a passenger in a car for an hour without a break = 3

Sitting quietly after a lunch without alcohol = 2

Lying down to rest in the afternoon when the circumstances allow =1

Total score = 7

In addition to EDS, she had problems of sleep fragmentation, choking during sleep and abnormal motor movement during sleep.

Physical examination showed:

Blood pressure = 140/100

Neck circumference = 12 inches

Body weight = 59.0 kg , height = 1.60m meter, BMI = 23.05 kg/m^2

The facial contour was assessed and retrognathia and micrognathia was found

Normal nasal septum and normal interior

Normal adenoid and palatine tonsils

Mildly redundant palatal mucosa and long uvula

Moderately redundant lateral pharyngeal wall

Moderately large base of tongue

Fiberoptic Nasopharyngoscopy and Muller's maneuver was done and showed 50% obstruction at the level of the retropalatal and retrolingual areas. Protrusion of the mandible improved the visualization of the laryngeal inlet.

Lateral Cephalometry X-Ray showed small mandible and marked narrowing of the posterior airway space at hypopharyngeal level.

Standard Nocturnal Polysomnography was done with the same technique as the first case.

Sleep study result showed total sleep time = 339 minutes, sleep time efficiency = 82%, apnea index = 24.00, respiratory disturbance index = 24.00, lowest oxygen saturation = 91%. The interpretation of this study was Obstructive Sleep Apnea Syndrome.

The patient refused CPAP treatment and wanted surgery to be performed.

The surgery was scheduled for Uvulopalato-pharyngoplasty (including tonsillectomy), Mandibular Osteotomy and Genioglossus Advancement. No serious complication occurred during surgery. In the immediate postoperative period there was upper airway obstruction after extubation but immediate improvement after a nasopharyngeal airway was inserted. During the first and second post-op day the patient was kept in the intermediate ICU for monitoring and close observation. The nasopharyngeal airway was removed on the next day without any problems. There was a small amount of hematoma on the floor of mouth on the first postoperative day but it resolved spontaneously a few days later. The surgical result was assessed clinically. Improvement of snoring was achieved. She attained better sleep after the first week post-op. The second polysomnogram was advised for the patient 3 months after the surgery.

Surgical Technique of Mandibular Osteotomy and Genioglossus Advancement (GA)

When a preoperative evaluation implicates the base of tongue and hypopharyngeal region as the cause of upper airway collapse, a procedure to widen the retrolingual area is indicated. The procedures directed at enlarging the retrolingual region, inferior sagittal mandibular osteotomy and genioglossus

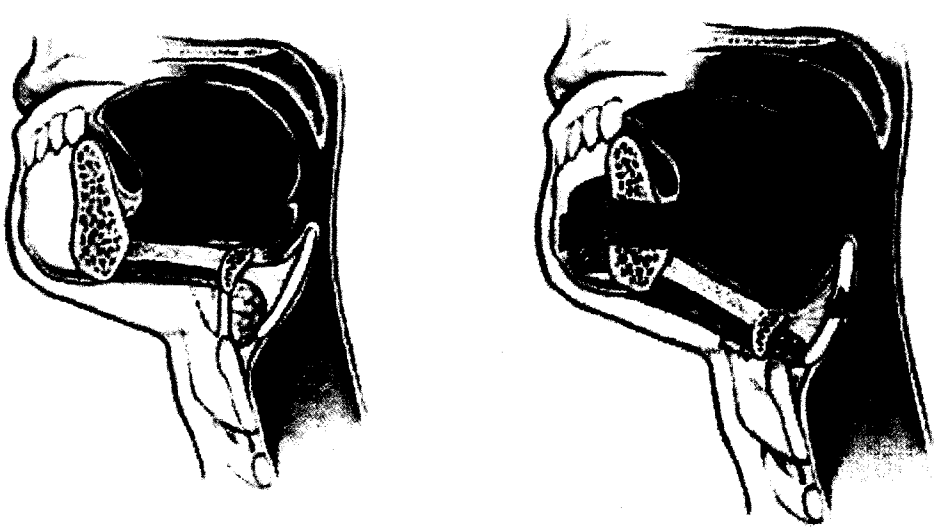


Figure 1. Surgical Technique of Mandibular Osteotomy and Genioglossus Advancement

advancement, with or without hyoid myotomy and suspension, appear to be the most promising. The rationale for the GA procedure is that the main protrusion muscle of the tongue, the genioglossus, is placed under tension. During sleep-induced hypotonia or atonia, this tension restricts the collapse of the tongue into the airway. This technique has undergone several modifications to obtain the best cure and the least morbidity. Presently, after the anterior mandibular osteotomy isolates the geniotubercle, this fragment is rotated to allow minimal bony collapse. The advancement is about 14 mm (= the thickness of the symphysis of mandible). Complete rotation of the fragment can detach the genioglossus muscle from its insertion and cause necrosis of the osteotomy fragment. After rotating the fragment, it is immobilized with the mandible by a 2.0 titanium screw with lag screw technique. Prior to advancing the fragment, it is pushed backwards for hemostasis on the bone cut surface with electric cautery and gelfoam placed in the marrow. (Fig.1)

Discussion

Treatment of SDB consists of weight reduction, modification of body position during sleep, positive airway pressure treatment (CPAP or BIPAP)[14-15] and surgery. Although CPAP is the gold standard of treatment, the patients' compliance to use this equipment is usually poor due to many factors.[16-25]

Treatment criteria for positive airway pressure treatment of adult obstructive sleep apnea patients[14] is as follows :

- all OSAS patients with an RDI > 30 events per hour, regardless of symptoms, based on the increased risk of hypertension.

- patients with RDI of 5-30 events per hour accompanied by symptoms of excessive daytime sleepiness, impaired cognition, mood disorders, insomnia, or documented cardiovascular diseases, to include hypertension, ischemic heart disease, or stroke.

Presurgical valuation should include complete ENT examination, fiberoptic nasopharyn-goscopy, lateral cephalometric analysis and polysomnography.[26]

Fiberoptic nasopharyngoscopy is more effective to evaluate the sites of obstruction in the airway. It can easily detect posterior septal deviation, enlargement of the posterior aspect of the inferior turbinate, nasopharyngeal lesions, and the position of the soft palate, uvula and base of the tongue as they relate to the lateral and posterior pharyngeal wall while sitting and lying supine. As part of the evaluation, the two following maneuvers should be done. (1) the Muller maneuver,[26-28] which is performed by inhaling against a closed oral and nasal passageway, evaluates collapse of the retropalatal and the retroglossal area. The negative pressure with this maneuver attempts to simulate the sleep-related pressure changes during sleep, (2) to protrude the mandible forward. If this maneuver improves visualization of the endolarynx, a tongue base surgical procedure such as genioglossus advancement should improve the obstruction.[28]

Lateral cephalometric analysis is used to assess the facial skeletal anomalies such as maxillary or mandibular retrusion. It can also be used to assess the posterior airway space (PAS), and hyolingual complex abnormalities.[29]

Because of the fact that clinical impression alone is not sufficient to reliably identify patients with

or without sleep apnea (sensitivity = 60 % and 63 % respectively)[30], standard nocturnal polysomnography (NPSG) is indicated for the diagnosis of possible OSAS[30-36]. It includes recording and analysis of the following parameters: EEG, EOG, EMG, oronasal airflow, chest and abdominal wall effort, body position, snore microphone, ECG, oxyhemoglobin saturation, and intraesophageal pressure monitoring in some cases. The duration of a diagnostic NPSG is at least 6 hours with the exception of the diagnostic portion of a split-night study, which is at least 2 hours in duration.[14] Limited-channel diagnostic NPSG may be indicated for patients with a high pretest probability of OSAS based on validated screening algorithms. This technique should include the following minimum parameters: oronasal airflow, chest wall effort, ECG, and oxyhemoglobin saturation. However, it is not effective in distinguishing sleep from wake or determining sleep stage and is less accurate than a standard NPSG in determining the number of obstructive respiratory events.[14]

Surgical treatment philosophy: [26,28,29,37,38]

- Treatment to cure
- Site-specific correction
- Staged management if necessary
- Full patient disclosure of options and risks

Surgical indications for treatment:[28]

- RDI > 20
- Oxyhemoglobin desaturation < 90 %
- Altered daytime performance and excessive daytime sleepiness
- Significant associated cardiac arrhythmias
- Specific anatomic abnormality identified
- Refused or rejected positive airway pressure treatment and desire for surgery
- Medically stable enough to undergo the recommended procedure

Classification of disease severity :

1. Primary snoring

Diagnostic criteria:

- RDI < 5
- no O2 saturation < 90 %
- Pes less than -10 cm H2O
- no complaint of excessive daytime sleepiness & fatigue

2. Upper airway resistance syndrome (UARS)

Diagnostic criteria:

- RDI < 5
- No O2 saturation < 90%
- Pes more than -10 cm H2O
- Presence of short, transient arousal (2-3 sec duration) on PSG
- Complaint of excessive daytime sleepiness & fatigue

3. Obstructive sleep apnea syndrome (OSAS)

Diagnostic criteria:

- RDI > 5
- O2 saturation < 90%
- Pes more than -10 cm H2O
- Presence of short, transient arousal (2-3 sec duration) on PSG
- Complaint of excessive daytime sleepiness & fatigue

Surgical procedures for treatment of OSAS at designated levels[26,28,29,37,38,40-43]

Bypass all upper airway obstructions → Tracheostomy

Selectively eliminate specific abnormalities in the upper airways

1. Nose → nasal reconstruction, radiofrequency

ablation of inferior turbinate

2. Velopharyngeal sphinctor → UPPP, LAUP, somnoplasty (radiofrequency ablation of soft palate)

3. Base of tongue → mandibular osteotomy and genioglossus advancement, base of tongue resection, radiofrequency ablation of base of tongue

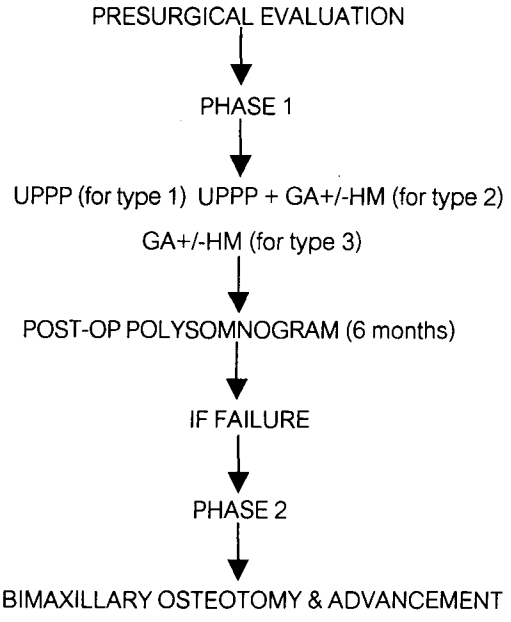
4. Hyoid → hyoid myotomy and suspension

5. Bimaxillary osteotomy and advancement

Anatomic site of obstruction[39-41]

- Type 1 : retropalatal
- Type 2 : retropalatal + retrolingual
- Type 3 : retrolingual

Riley-Powell-Stanford surgical protocol[26,28]



The author makes the first report of two cases of Mandibular Osteotomy and Genioglossus Advancement procedure done for treatment of Sleep-disordered Breathing in Thailand, and hopes that this procedure will be widely used because of its safety and effectiveness.

Regarding the present status of practice for sleep-disordered breathing in Thailand, a good team approach that consists of pulmonologists, psychiatrists, neurologists, pediatricians, ENT surgeons and anesthesiologists is required because most SDB patients have compromised airways. Every patient should received a full disclosure of all treatment options, risks and benefits with no bias or prejudice from specialty physicians. Patients with obstructive sleep apnea syndrome or upper airway resistance syndrome should be advised to try to use CPAP before surgical consideration because CPAP is the gold standard of treatment. Surgery is only the alternative treatment and the result after surgery is not better than CPAP results.

Finally, nocturnal polysomnography is the most important tool to diagnose and to assess the severity of Sleep-disordered Breathing. Localization of the obstruction site is the most important consideration before the surgeon makes a decision which surgical procedures to use. Laser-assisted uvulopalatoplasty was once the popular procedure in the U.S. but nowadays it is unaccepted for treatment of Obstructive Sleep Apnea Syndrome, especially when retrolingual level obstruction is identified, because it does not improve the apnea / hypopnea index or the nadir of oxygen desaturation of the patients. On the other hand, it makes the obstruction persist but more silently.[45-46]

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