Journal of Maxillofacial and Oral Surgery

Impact of Mandibular Distraction Osteogenesis on the oropharyngeal airway in adult patients with obstructive sleep apnea secondary to retroglossal airway obstruction

--Manuscript Draft--

Manuscript Number:	MAOS-D-12-00336R1
Full Title:	Impact of Mandibular Distraction Osteogenesis on the oropharyngeal airway in adult patients with obstructive sleep apnea secondary to retroglossal airway obstruction
Article Type:	Clinical Paper
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Abstract:	The pharyngeal airway is a complex musculo-mucosal passage that is suspended form the skull base and has an indirect anchorage to the cranial base by the insertion of its musculature into the mandible. Any change in the three dimenssional position of the mandible in relation to the cranial base can have a significant impact on the volume and the patency of the oropharyngeal airway. The aim of this paper is to provide an anatomic (3D volumetric CT) and a physiologic (Polysomnographic) evidence of the changes in the oropharyngeal airway after Mandibular distraction osteogenesis by two different techniques in adult patients with obstructive Sleep apnea secondary to retroglossal airway obstruction
Response to Reviewers:	Sir, We have Provided the necessary clarifications as requested in the manuscript. Mandibular distraction in our cases was performed after addression the ankylosis and reconstructing the joint. Patients who underwent body distraction also underwent orthodontic alignment. No tongue reduction surgery was necessary in any of our patients as satisfactory results were obtained with distraction. Over correction by 20% (3-4mm) to compensate for relapse. All of these have been addressed in the manuscript as per your comments

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TITLE PAGE

Title: Impact of Mandibular Distraction Osteogenesis on the Oropharyngeal airway in adult patients with Obstructive Sleep Apnea secondary to retroglossal airway obstruction.

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Funding: Cleft Children International (Zurich)
No Competing interests
Key words: Mandibular Distraction Osteogenesis; Oropharyngeal airway; Obstructive Sleep Apnea; Retroglossal airway

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INTRODUCTION

Obstructive Sleep Apnea Syndrome (OSAS) is a complex disorder that not only affects the quality of life but can also have undesirable cardiorespiratory sequelae. The hypoplastic and/ or retro positioned mandible along with an inefficient genial muscular apparatus[1] are the strong factors that cause retroglossal airway obstruction leading to OSAS.

Anatomically the pharynx is formed by the soft tissues suspended from the cranial base. The mandible indirectly acts as a major anchorage point for the hyoid, the laryngeal cartilages, pharynx and the tongue musculature which help maintaining the airway patency. Thus the mandible is the key skeletal unit in preserving airway patency.

More than 52% of the upper pharyngeal airway obstructions occur at the base of the tongue[2]. This is secondary to disproportionate mandibular anatomy and mandibular advancement in such situations is shown to have 97.0 % of success in paediatric patients and 100%'success rate in adult patients[3,4,5,6,7,8,9,10] Mandibular advancement can be achieved by osteotomies or distraction and our choice of the latter was based on the magnitude of advancement required and its influence on post operative stability. [11,12]

Numerous studies have been performed to evaluate the structural oropharyngeal airway (OPA) changes in two [4,7,8,9] and three dimensions [5, 13,14] . Physiologic improvement of the same has been shown using polysomnography. However a combination of both the anatomic and physiologic changes in adult patients has seldom been addressed in a single issue. The aim of this study is to quantify the three dimensional volumetric changes in the oropharyngeal airway and correlate it to the functional improvement using polysomnography after Mandibular

Distraction Osteogenesis(MDO)(Corpus distraction and/or Orthomorphic distraction) in adult patients with retroglossal airway obstruction.

Materials and Methods: A prospective study was carried out at our Centre between 2007 -2010 after procuring clearance from the institutional ethical board and all the patients with TMJ ankylosis who reported with snoring, day time somnolence with clinical and radiographic evidence of retroglossal airway obstruction secondary to mandibular hypoplasia were included in the study. 13 patients (9 female and 4 male patients) around the median age of 21 years were included in the study. 11 patients had unilateral TMJ ankylosis and 2 patients had bilateral ankylosis. None of them had any specific adaptations to avoid snoring however, few patients claimed to be comfortable sleeping in prone position. The following were criteria followed to enrol the patients in the study.

Inclusion criteria: 1.Adult patients of TMJ ankylosis with clinical evidence of OSA (snoring and day time somnolence) secondary to Mandibular hypoplasia and retroglossal airway compromise.

2. Radiologic evidence of a linear dimension of the retroglossal airway less than 10mm

3. AHI index greater than or equal to 5.

A pre operative Echocardiogram with evaluation of pulmonary artery pressure was performed in all the patients to rule out any the cardiorespiratory co- morbidities. All the patients were moderately built except for patient number 3 who had mild degree of obesity.

Lateral cephalograms were taken for all these patients to evaluate the position of the mandible to the maxilla and cranial base. The linear dimension of the retro-glossal airway was assessed based on Mcnamara's analysis. Patients with a linear depth of the retro-glossal airway less than 10 mm

with clinical evidence of snoring secondary to mandibular retrognathism were included in the study.

A 3D CT was performed and the volume of the OPA was assessed using the following landmarks:

Cranial – the hard palate plane,

Caudal – the glottis,

Ventral – the line joining the dorsum of tongue base and the uvula ,

Dorsal – the posterior pharyngeal wall, and

Lateral – the right and left lateral pharyngeal walls.

The functional status of the OPA was evaluated by pre operative Polysomnography and the Apnea- Hypopnea Index (AHI) was calculated. All the patients with an AHI greater than 5 were taken up for Mandibular distraction osteogenesis with informed consent of the patient. Mandibular distraction in all our patients was performed after ankylosis release and reconstruction of the condyle with costochondral graft.

The surgical procedure involved conventional mandibular body distraction in 8 patients. 5 patients with unilateral ankylosis had stable occlusal segment hence we chose to do orthomorphic distraction[15,24] as performing a body distraction in these patients would worsen the existing occlusion. Thus body distraction was performed in patients willing to undergo orthodontic alignment.

Distraction was performed as per the standard protocol to achieve the desired mandibular position as decided by the pre operative lateral cephalogram. An overcorrection of 3-4 mm was performed in all the cases considering the possibility of relapse.

Post operative 3D CT and polysomnography was performed 3 months after distraction to evaluate the volumetric and the functional changes in the OPA.

RESULTS

The results of this study have been tabulated under four sub-headings for a clear understanding of the dimensional and functional changes in the pharyngeal airway.

1. The Mandibular advancement achieved (Table 1) - The mean mandibular advancement achieved was 11.07mm with a range of 7 to 14mm. (Table 1)

2. The changes in the linear dimensions of the OPA (Table 2) - An increase in the OPA antero-posterior dimension was observed ranging from 1mm to 4.5mm and a mean increase of 2.5mm.

3. The volumetric change in the OPA (Table 3) - Quantitative assessment of the OPA volume before and after advancement demonstrated an average increase by19.3% with a range of 7.7% - 29.8%.

4. The AHI index and the oxygen saturation trends (Tables 4, 5) - 12 patients had shown a significant improvement in the AHI post operatively with a decrease in the mean AHI from 44.8 (Range: 84.8 - 12.9) to 13.2. (Range: 45.5 - 2.8) (Table 4).The average pre operative lowest oxygen saturation was 68.3% (Range: 50% - 92%) and post advancement, the average lowest oxygen saturation was 80.97% (Range: 50 - 93%). The pre operative average oxygen saturation ranging from 80% to 98.5% in the pre op improved to 94.4% to 99.7% post operatively, which was clinically significant in terms of reducing snoring and day time somnolence (Table 5).

DISCUSSION

Micrognathia and retrognathia are common features seen in a number of congenital and acquired craniofacial anomalies including Treacher Collins syndrome, Hemifacial Microsomia, Nager syndrome, Pierre Robin sequence and like TMJ ankylosis.[7,8] Abnormalities in the position or morphology of the skeletal tissues have a direct impact on the surrounding envelope of soft tissues of the upper airway, thereby indirectly influence the airflow through it. True micrognathia with associated retro-glossal airway compromise is a significant cause for airway obstruction, and was the common feature in all our patients.

Obstructive Sleep Apnea Syndrome is a condition caused by upper airway obstruction, characterized by apneic and/ or hypopneic episodes during sleep, which in extreme conditions may be accompanied by severe secondary cardio-respiratory problems. These cardiorespiratory issues are secondary to an increase in the negative intra thoracic pressure with an increase in the right ventricular pressure and volume.[2] The priority in patients suffering from OSAS is to relieve them of the causative factor at the earliest. The modality of treatment used is determined by the site of obstruction (upper airway secondary to nasal, nasopharyngeal obstruction or lower pharyngeal airway secondary to retroglossal obstruction) with due consideration to the patient's systemic status.

To be successful, the surgical procedure must bypass the obstructive area (Tracheostomy) or enlarge the airway (mandibular advancement) or prevent collapse of the soft tissues at the site of obstruction (tongue reduction).

Total Mandibular advancement was the first orthognathic surgical procedure used for the treatment of retroglossal airway obstruction[16]. A definitive explanation for an improvement in the airway status after Mandibular advancement is still ambiguous. However, greater acceptance has been given to the fact that there is repositioning of the genioglossus along with the advancing

segment which pulls the tongue forwards from the posterior pharyngeal wall leading to better airway patency [7,10] Recent studies reveal that the genial musculature in these patients show an abnormal neuromuscular activity which improves with mandibular advancement leading to a significant improvement in the airway patency.

Numerous surgical options have been described for the management of OSAS secondary to hypoplastic mandible of which distraction osteogenesis has gained significant acceptance. The magnitude of advancement that can be achieved and the predictability of outcome make MDO the treatment of choice over conventional osteotomies in adult cases of OSAS[3,4,5,6,7,8,10,11,17] Genial Advancement with Hyoid Myotomy[18,19] and Suspension, and genial distraction[20] are the alternate options that have been reported in the literature to improve the retroglossal airway patency.

MDO has been our choice of treatment in these patients as we can achieve better control over the proximal and distal segments with MDO than with osteotomies. Apart from this, MDO does not have secondary morbidities associated with the harvest of bone grafts and the problems associated with bone graft fixation. Mandibular body distraction and Orthomorphic distraction[15] have been the two techniques that have been used for the correction of OSAS in our patients. Both these techniques increase the space within the body of the mandible which can accommodate the tongue better and thus prevent the retroglossal airway obstruction. However Mandibular body distraction brings about changes in the occlusal table which may worsen pre-existing occlusion in adult patients. This has prompted us to use isolated basal bone distraction[24] (Orthomorphic distraction patients 8, 10,11,12,13). Orthomorphic distraction brings about an increase in the retroglossal airway volume by increasing the space within the mandible and also exerts a forward traction over the tongue musculature. In these patients, the facial asymmetry

and the management of ankylosed joint can be addressed in a single stage. Tongue reduction procedures were not necessary in our patients as advancing the mandible had provided a successful outcome. Adjuvant procedures involved advancement genioplasty that was performed in 1 patient with bilateral ankylosis exclusively to improve the cosmetic outcome (Performed after the study).

Numerous studies have been performed to assess the airway dimensions after mandibular advancements using lateral cephalograms [4,5,8,9] but these do not demonstrate the actual volumetric improvement at the pharyngeal inlet. Computed tomography provides valuable data regarding the 3 dimensional changes in the airway space and volume which gives us credible proof for the outcome of the surgical treatment.[5,3,14] Perlyn et al[7] used CT scans and demonstrated an increase in the space within the body of the mandible and upper airway volume by quantitative assessment after MDO.[13,21] Our patients revealed a greater increase in the lateral dimensions of the airway than the antero posterior dimensions similar to that reported by Fairburn et al[14] who stated that lateral dimensions get enhanced to a greater extent than antero-posterior dimensions in the retroglossal region. This could be probably due to the traction on the mylopharyngeus component of the superior constrictor along with the traction on the base of the tongue. Though 3D CT provides an accurate analysis of the pharyngeal airway, the limiting factor is that it interprets a dynamic functional anatomic unit in a static manner, as the scanning time of the CT unit is greater than 4 seconds (=8 seconds) with in which one breathing cycle would be complete. An important addition in this study is to evaluate the physiological response and functional outcome of this procedure using polysomnography which is the gold standard in the assessment of OSAS. This enabled us to correlate the anatomical and volumetric changes of the airway to the functional outcome for the correction of OSAS.

Surgical success in the management of OSAS can be evaluated using two parameters - An AHI less than 20 or a 50% decrease in the AHI with a near normal peripheral oxygen saturation [22,23] (Spo_2). 12 patients in our series showed a successful post-surgical outcome with significant improvement in the AHI status which could be predictably correlated with an overall increase in the upper pharyngeal airway dimension and volume on 3D CT. The lowest oxygen saturation improved in all patients except one (Patient 3) whose lowest oxygen saturation remained to be 50% despite of a mandibular advancement of 10mm and an increase in the pharyngeal airway volume by 12.44% (Table 3) (which is similar to the rest of the successful cases in the group). This may be attributed to the obesity and large neck circumference (18 inches) which would lead to the collapse of the adipose tissue into the airway. A decrease in Functional Residual capacity of the lungs secondary to obesity leading to hypoxemia has also been reported to be a cause of OSA in these patients. Both the techniques of mandibular distraction that have been used in our series have shown equivocal results in the anatomic and the physiologic improvement of the oro pharyngeal airway status.

None of the patients complained of any recurrence of snoring episodes during the subsequent follow up phase of 2 years and volumetric 3DCT evaluation of the OPA performed 1 year after MDO in two of our patients did not reveal any significant change as compared to the immediate post consolidation OPA volume which confirmed the stability of the results achieved with MDO.

To conclude, OSAS is a disease with multifactorial aetiology and varied treatment options. The success in the treatment depends on the identification of the pathologic etiological factor and addressing the same. Mandibular distraction Osteogenesis (total mandibular DO or distraction of mandibular basal bone) is one of the best and a viable option in adult patients with OSAS.

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Tables

Table 1 The amount of mandibular advancement measured on Lateral Cephalograms

Table 2 Comparison of the pre-operative and post-operative (After 3 months) Linear OPA space on a Lateral Cephalogram (McNamara's Airway analysis)

Table 3 Changes in the OPA volume determined pre and post-operatively (After 3 months)

Table 4 Apnea and Hypopnea Index calculated pre and post-operatively (After 3 months)

Table 5 Pre-operative and Post-operative values of average and Lowest Oxygen Saturation

Legends for pictures

Figure1: Pre operative profile picture of a patient with Unilateral TMJ Ankylosis

Figure 2: Pre-operative lateral cephalogram of the same patient

Figure 3: Pre-operative CT tracing of OPA volume after digitising the relevant anatomical landmarks

Figure 4: Post-operative profile of patient

Figure 5: Post-operative lateral cephalogram

Figure 6: Post-operative CT showing the OPA

Table 1: **The amount of mandibular advancement measured on Lateral Cephalograms**

Sl no.	Patient name	Pre-op SNB	Pre-op mandibular length (in mm)	Post-op SNB	Post-op Mandibular Length (in mm) (After 3 months)	Total Amount of advancement (Lateral Ceph Assessment)
1.	P.K	62	59	62	70	11
2.	K.S	58	49	70	60	11
3.	T. K	59	46	64	56	10
4.	V.H	63	54	68	63	9
5.	R. V	68	62	73	69	7
6.	S. M	51	56	65	70	14
7.	R. K	55	57	70	66	9
8.	A. P	67	62	67	72	10
9.	M. S	67	52	67	63	11
10.	M.N	60	55	63	65	10
11	A.N	66	61	66	71	10
12	B.C	66	63	66	74	11
13	MB	68	65	69	76	11

Table 2 Comparison of the pre-operative and post-operative (After 3 months) Linear OPA space on a Lateral Cephalogram (McNamara's Airway analysis)

Sl no.	Patient name	Pre-oporopharyngeal airway (in mm)	Post-Operative oropharyngeal airway (in mm)	Change in oro pharyngeal airway (in mm)
1.	P. K	4.0	5.0	1.0
2.	K.S	2.5	5.5	3.0
3.	T. K	1.0	4.5	3.5
4.	V. H	6.0	7.0	1.0
5.	R. V	5.0	7.0	2.0
6.	S. M	2.0	6.0	4.0
7.	R. K	8.0	9.0	1.0
8.	A. P	4.0	6.0	2.0
9.	M. S	4	5.5	1.5
10.	M.N	3.5	6	2.5
11	A.N	4	7	3
12	BC	4.5	9	4.5
13	MB	3	7	4

Table 3 Changes in the OPA volume determined pre and post-operatively (After 3 months)

Sl no.	Patient Name	Pre-Op Airway Volume in cm^2	Post-Op Airway Volume in cm^2	Percentage of increase in airway volume
1	P.K	15.2	18.6	22.0
2.	K.S	14.6	16.4	12.7
3.	T.K	17.1	19.2	12.4
4.	V.H	23.6	27.3	15.6
5.	R.V	26.8	34.3	29.6
6.	S.M	22.8	24.6	7.7
7.	R.K	17.3	22.5	29.7
8.	A.P	17.5	20.3	19.1
9.	M.S	23.3	27.3	15.6
10.	M.N	22.6	24.9	29.8
11	AN	18	21.5	29
12	B.C	24	26.3	15.6
13	MB	17.1	19.2	12.4

Table

Table 4 Apnea and Hypopnea Index calculated pre and post-operatively (After 3 months)

Sl No.	Name	Age of the patient in years	Diagnosis	Pre-op AHI	Treatment done	Post-op AHI
1	P. K	19	Unilateral TMJ Ankylosis	20.6	Orthomorphic DO	2.8
2.	K.S	18	Unilateral TMJ Ankylosis	43.5	Mandibular Body DO	4.6
3	T. K	41	Unilateral TMJ Ankylosis	50.5	Mandibular body DO	45.5
4	V. H	20	Unilateral TMJ ankylosis	33.3	Mandibular body DO	4.6
5.	R.V	18	Unilateral TMJ Ankylosis	30.3	Mandibular body DO	3.5
6	S. M	20	Bilateral TMJ Ankylosis	84.8	Mandibular body DO	38.8
7	R.K	21	Unilateral TMJ Ankylosis	40.4	Mandibular body DO	11.6
8	A.P	22	Unilateral TMJ Ankylosis	33.2	Orthomorphic DO	5.1
9	M. S	26	Bilateral TMJ Ankylosis	12.9	Mandibular Body DO	4.2
10	M.N	25	Unilateral TMJ ankylosis	78.7	Orthomorphic DO	13.2
11	AN	27	Unilateral TMJ ankylosis	34	Orthomorphic DO	6
12	B.C	19	Unilateral TMJ ankylosis	76	Orthomorphic DO	17
13	MB	27	Unilateral TMJ ankylosis	45	Orthomorphic Distraction	10.6

Table 5 Pre-operative and Post-operative values of average and Lowest Oxygen Saturation

Sl No.	Patient name	Pre Op Average Oxygen Saturation	Pre Op lowest Oxygen Saturation	Post Op average Oxygen Saturation	Post Op lowest Oxygen Saturation
1.	P. K	97.0	76.0	99.7	92.0
2.	K.S	97.8	73.0	99.5	89.0
3.	T. K	80.0	50.0	95.3	50.0
4.	V. H	96.8	82.0	99.2	90.6
5.	R. V	97.0	79.2	99.4	90.3
6.	S.M	92.6	50.0	96.9	74.0
7.	R. K	93.4	65.4	96.5	72.6
8.	A.P	98.5	92.0	99.4	93.0
9.	M.S	96.8	52.0	99.4	70.4
10	M.N	90.2	57.0	94.4	77.0
11	AN	96.6	90.o	95.4	93
12	BC	91.2	60	97.4	72.4
13	MB	95.4	62.4	97.4	88.4

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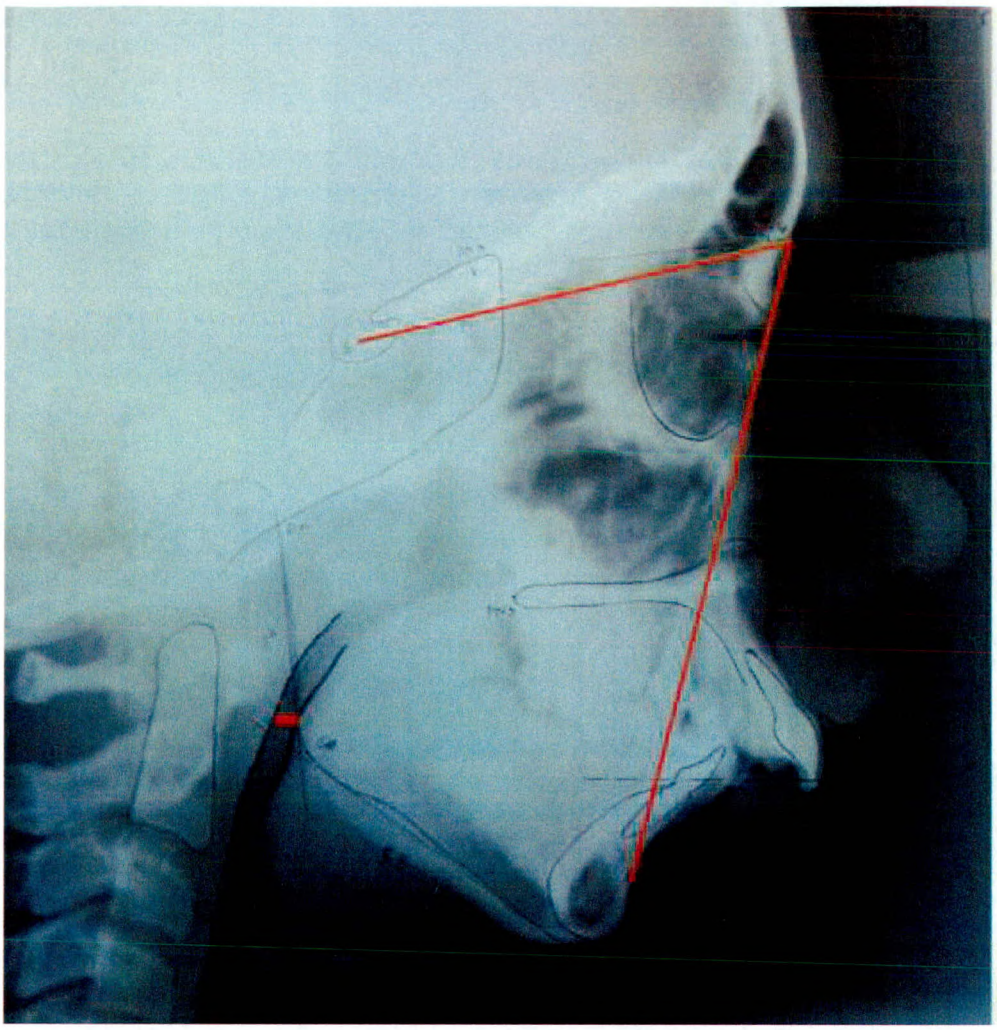


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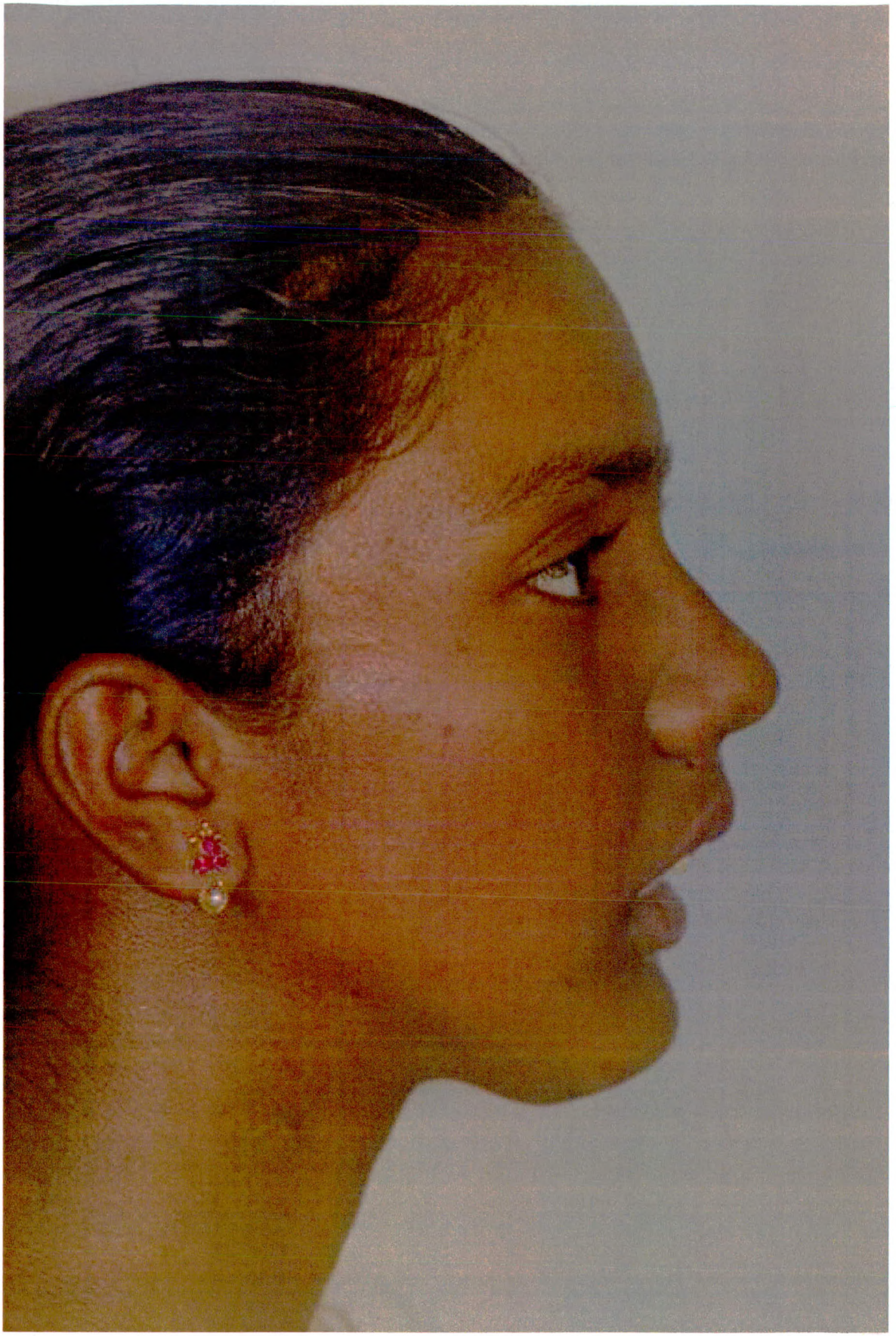


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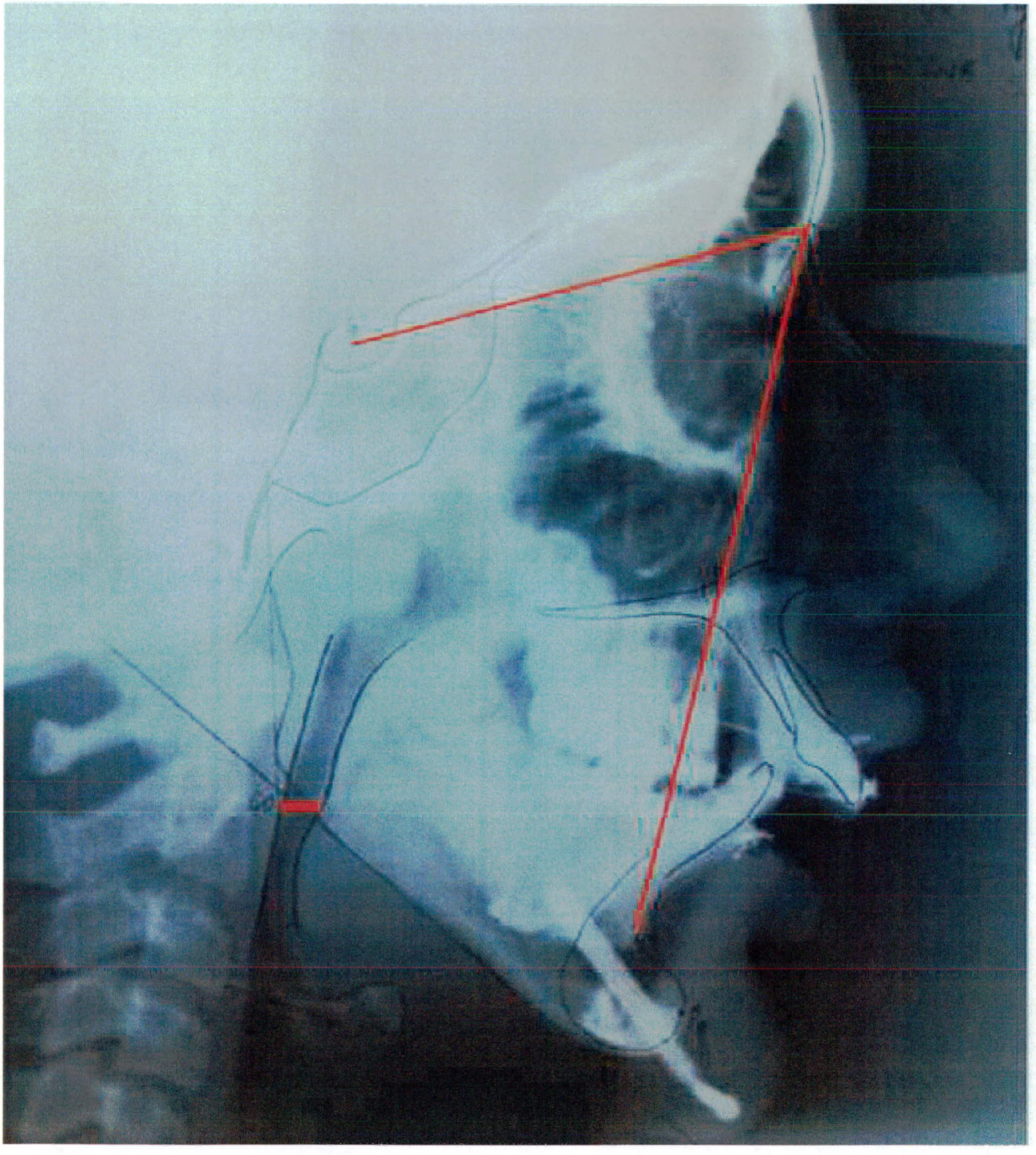


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