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Assessment of Maxillary Growth in Patients with Anterior Palatal Fistula Managed by Distraction Osteogenesis With and Without Bone Graft

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ABSTRACT

Background: Palatal fistula is probably the commonest complication associated with cleft palate surgery. Traditional surgical and orthodontic treatment for these patients often fall short of expectations.

Patients and Methods: This study was performed on 30 patients with anterior palatal fistula and maxillary hypoplasia and were divided into 3 groups according to the size of fistula and methods of closure of the bony gap. Group I closure was done by cancellous bone graft. Group II closure was done by application of internal maxillary distractor. Group III closure was done by application of internal maxillary distractor and cancellous bone graft.

Results: In 60% of patients of Group I; closure of the gap occurred after 6 months. In 50% patients of Group II; closure of the gap occurred after 6 months and this percent raised to 90% after 1 year. While in 50% patients of Group III; closure of the gap occurred after 6 months and this percent raised to be 80% after 1 year as shown by Cone Beam CT.

Conclusion: Closure of anterior palatal fistula either by distraction osteogenesis with and without bone graft or by bone graft alone may be a stimulus for growth of the maxilla at suture sites.

Key Words: *Maxillary – Anterior palatal fistula – Distraction – Osteogenesis.*

INTRODUCTION

Palatal fistula is probably the commonest complication associated with cleft palate surgery. The rate of palatal fistula varies from 4-35% **[1]**.

The primary causes of development of palatal fistula is repair under tension, postoperative infection which is hardly seen in small children and vascular accidents during palatoplasty can also cause flap loss. Besides these, inadvertent use of diathermy, particularly near the greater palatine pedicle can compromise the blood supply of the mucoperiosteal flap and can result in fistula **[2]**.

Amaratunga, 1988 **[3]** advised thatsurgical closure of palatal fistula should be attempted at least six months after the previous surgery.

In addition to performing soft tissue flaps, Posnick and Tompson, 1992 **[4]** described the use of segmental Le Fort I osteotomies for maxillary hypoplasia and the closure of alveolar defects and residual palatal fistulas. The major advantage of this technique is its ability to close simultaneously dead cleft space, fistulas, and alveolar defects. However, it is more invasive than distraction osteogenesis.

Le Fort I osteotomy with direct advancementhas many disadvantages when used for correction of hypoplastic maxilla in CLP patients, such as limited advancement, requirement for bone graft, negative-effect on velopharyngeal closure, and high risk of bonenecrosis andrelapse **[5]**.

Yen SL et al., 2003 **[6]** performed Distraction Osteogenesis to close a palatal fistula in a cleft patient and transported a small palatal disk consisting of a tooth and the surrounding bone to narrow the cleft space. This was the first use of Distraction Osteogenesis to close a palatal fistula in a cleft patient.

Maxillary anterior segmental Distraction Osteogenesis was first used in a cleft patient to advance a hypoplastic maxilla by Karakasis D, Hadjipetrou L, 2004 **[7]**. They performed a two-stage procedure; initially closure of the unilateral palatal fistula with adjacent mucosal flaps, and after a

recovery period of 3 weeks they advanced the maxilla with anterior segmental Distraction Osteogenesis.

Maxillary anterior segmental Distraction Osteogenesis would be an alternative technique to reduce the size of the fistula whenthe palatal fistula is too large to close with mucosal flaps. While the segments are advanced anteromedially, this technique has the additional advantage of advancing a hypoplastic maxilla without inducing velopharyngeal incompetence and stimulating its growth **[8]**.

Palatoplasty could inhibit the vertical growth of posterior region of maxilla **[9]**. It also may inhibit forward displacement of the maxillary base and anteroposterior development of the maxillary dentoalveolus in unilateral cleft lip and palate patients **[10]**.

Cleft patients with anterior palatal fistula suffer from maxillary hypoplasia that leads to facial deformity and class III malocclusion with collapse of the maxillary segment on the cleft side. Also when the size of the fistula is large (>5mm), it may be difficult to close it by traditional methods due to insufficient soft tissue coverage.

The aim of this work is to assess the potential growth of the maxilla in patients with anterior palatal fistula at the age of mixed dentition after soft tissue repair and attaching the two maxillary parts by distraction alone or by distraction and bone graft.

PATIENTS AND METHODS

This study was carried out in the Department of Plastic and Reconstructive Surgery, Ain Shams University in the period between 2013-2015 on 30 patients with anterior palatal fistula either true unrepaired or previously repaired but at least 6 months after the previous surgery at the age of mixed dentition (6-12 years). Patients were divided into three groups.

Group I: Included 10 patients with small anterior palatal fistula (<3mm) who were managed by closure of the fistula and application ofiliac crest cancellous bone graft Fig. (1).

Group II: Included 10 patients with medium sized anterior palatal fistula (3-5mm) who were managed by closure of the fistula and application of internal maxillary distractor Fig. (2).

Group III: Included 10 patients with large sized anterior palatal fistula (>5mm) who were managed by closure of the fistula and application of both internal maxillary distractor and bone graft Fig. (3).

Active distraction was begun on the 6th postoperative day at a rate of 0.5mm twice daily until the desired amount of maxillary movement has been achieved (closure of the osteotomy gapdistally). Once the appropriate distraction was achieved, the device was left in place for 12 weeks to permit bone consolidation. The fistula and the 2 maxillary arches are evaluated both clinically and radiologicallypreoperatively and every six months postoperatively for 1 year through;

A- Occlusal view X-ray skull.

B- Cephalometrypostoperative to assess the potential growth of the maxilla with increase in SNA and decrease in ANB angles.

C- Cone Beam CT scanto assess closure of the bony gap with attachment of the two maxillary segments to achieve one piece of maxilla in spite of being clefted Fig. (4).

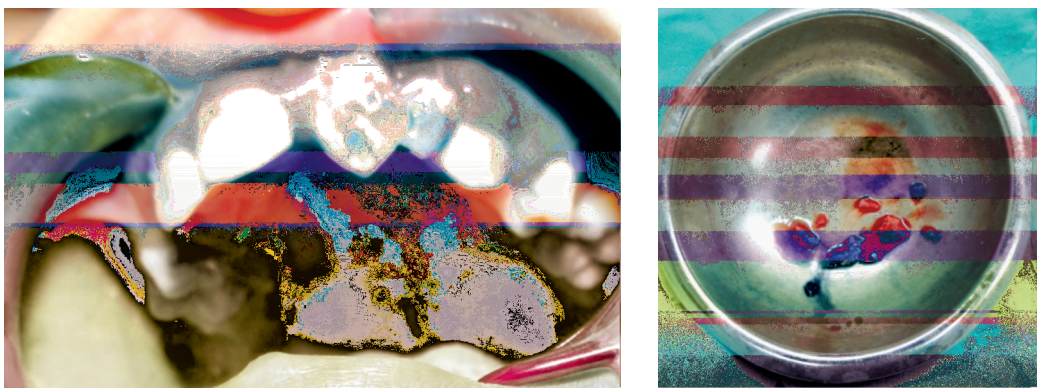

Fig. (1): Showing on the left side closed anterior palatal fistula, on the right side showing iliac crest cancellous bone graft used for closure in a 7 years old male patient.

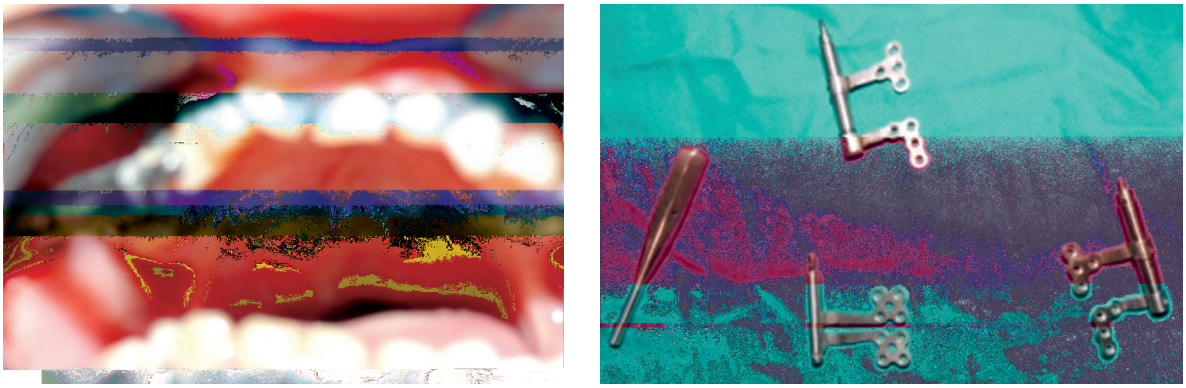

Fig. (2): Showing on the left side closed anterior palatal fistula and the distractor in place in 7 years old male patient while on the right side the internal maxillary distracters used.

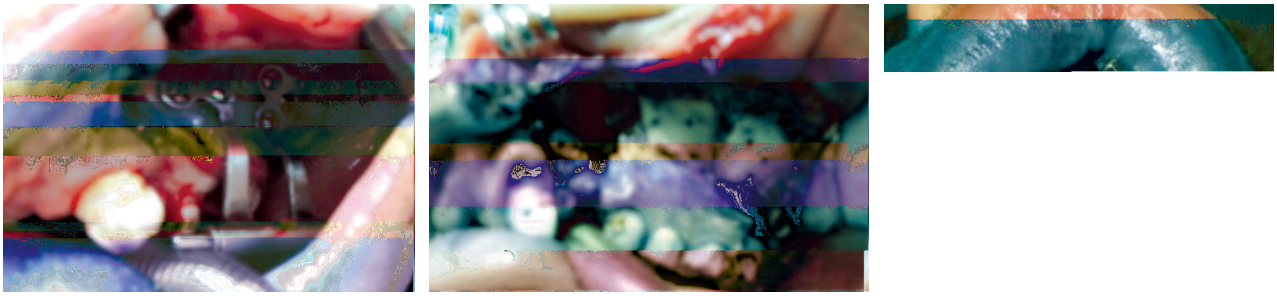

Fig. (3): Showing on the left side large anterior palatal fistula and maxillary osteotomy done, on middle showing closure of the gap partly by compression using distractor and the remaining gap filled by bone graft while on the right side shows closure of anterior palatal fistula and the distractor in place 3 months postoperative.

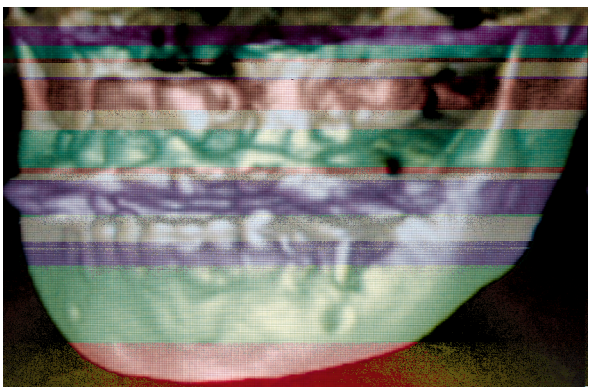

Fig. (4): Showing cone beam ct scan on maxilla and mandible of 7 years old male patient with closure of anterior palatal fistula by distraction osteogenesis.

RESULTS

I- *Closure of the bony gap after 6 months and after 1 year:*

In Group I 60% of patients (six of ten patients) showed closure of the bony gap at 6 months and this percent raised to 70% after 1 year.

In Group II 50% of patients (five of ten patients) showed closure of the bony gap after 6 months and this percent raised to 90% after 1 year.

In Group III 50% of patients (five of ten patients) showed closure of bony gap after 6 months and this percent raised to 80% after 1 year.

But as shown in (Table 1). The difference between the outcome (closure of the gap) and the different performed procedures was no statistically different.

Table (1): Assessment of the outcome (closure of the gap after 6 months and after 1 year) in the different performed procedures.

	Procedure						*p*-value*
	Bone graft N=10		Distractor N=10		Distractor and bone graft N=10		
	N	%	N	%	N	%	
Closure of gap after 6 month:							
No	4	40	5	50.0	5	50	1.000
Yes	6	60	5	50.0	5	50	
Closure of gap after 1 year:							
No	3	30	1	10.0	2	20	0.845
Yes	7	70	9	90.0	8	80	

*: Fisher's Exact Test.

II- *Occurrence of complication (bleeding and infection) in different performed procedure:*

In Group I, postoperative bleeding occurred in 10% (one of ten patients) while infection occurred in 30% (three of ten patients) of patients.

In Group II, postoperative bleeding occurred in 10% (one of ten patients while infection occurred in 10% (one of ten patients) of patients.

In Group III, no case was complicated by post-operative bleeding while infection occurred in 20% (two of ten patients) of patients.

But as shown in (Table 2), the difference between the occurrence of complication and the different performed procedure was not statistically significant.

Table (2): Assessment of the complication (post-operative bleeding and infection) in the different performed procedures.

Post op. Bleeding:			
	No		
	Yes		
Infection:			
	No		
	Yes		

IV- *Relation between age of the patients, fistula size and closure of the gap after 6 months:*

Comparing the age by mean and standard deviation between patients with closure of the gap after 6 months and without closure after 6 months there was no statistically significant difference between the two groups (Table 4).

Comparing the fistula size by mean and standard deviation between patients with closure of the gap after 6 months and without closure after 6 months there was no statistically significant difference between the two groups (Table 4).

V- *Relation between age of the patients, fistula size and closure of gap after 1 year:*

Comparing the age by mean and standard deviation between patients with closure of the gap after 1 year and without closure after 1 year there was no statistically significant difference between the two groups (Table 5).

Comparing the fistula size by mean and standard deviation between patients with closure of the gap after 1 year and without closure after 1 year there was no statistically significant difference between the two groups (Table 5).

Table (5): Comparing the age and fistula size by mean and standard deviation between patients with closure of the gap after 1 year and without closure after 1 year.

	Closure of gap after 1 year				Student *t*-test	*p*-value
	No		Yes			
	Mean	Standard deviation	Mean	Standard deviation		
• Age	8.33	1.86	7.58	1.28	1.171	0.252
• Fistula size	4.50	2.26	4.42	1.56	0.107	0.915

VI- *Relation between fistula size and the occurrence of complications:*

In comparing fistula size by mean and standard deviation in patients who suffered from complications as infection and those who didn't suffer from complication, there was no statistically significant difference between the two groups (Table 6).

Table (6): Comparing fistula size by mean and standard deviation in patients who suffered from complications (either post-operative bleeding or infection) and those who didn't suffer from complication.

	Post op. bleeding				Student *t*-test	*p*-value
	No		Yes			
	Mean	Standard deviation	Mean	Standard deviation		
• Fistula size	4.46	1.71	4	1.41	0.442	0.724

	Infection				Student *t*-test	*p*-value
	No		Yes			
	Mean	Standard deviation	Mean	Standard deviation		
• Fistula size	4.42	1.56	4.5	2.26	–1.07	0.915

VII- *Assessment of maxillary growth after 1 year by cephalometric evaluation of patients:*

There is no statistically significant difference in increase of SNA between different performed procedures (Table 7) due to limited period of follow-up (1 year). But minimal promising changes were observed with increase in SNA angles and decrease in ANB angles. This will be our goal in the next few years of follow-up. Thus we will do serial cephalometric evaluation of those patients every 6 months in the next 5 years.

Table (7): Assessment of SNA increase after 1 year as an indicator for maxillary growth in the different performed procedures.

	Procedure						*p*-value*
	Bone graft		Distractor		Distractor and bone graft		
	N	%	N	%	N	%	
Increase in SNA:							
• No increase in SNA	5	50.0	5	50.0	4	40.0	1.000
• Increase in SNA	5	50.0	5	50.0	6	60.0	

*: Fisher's Exact Test.

DISCUSSION

Cleft lip and palate is a condition that is fairly common. It is one of the most common congenital defects found all over the world. Along with a plethora of other problems such as speech problems, hearing problems, feeding problems, dental defects and psychological problems, they also present with maxillary hypoplasia **[11]**.

Patients with severe maxillary deficiency are difficult to treat with traditional surgical/orthodontic approach. These patients present with maxillary hypoplasia in all the three dimensions along with thin and structurally weak bones. This is also compounded by residual palatal and alveolar fistulae, absent and aberrant dentition and scarring of palatal and pharyngeal tissue **[12]**.

The early treatment of mid-face retrusion in children with cleft lip and palate has to be considered a major goal. Early correction of the maxillary hypoplasia minimizes the psychological problems and provides benefits related to improved occlusion.

In the past it has been virtually impossible using maxillary advancement alone to treat patients with

severe maxillary deficiency. With the use of distraction, severely hypoplastic maxilla can be repositioned and maintained to desired horizontal and vertical positions without the use of bone grafting and hardware fixation **[13]**.

Contrary to the use of protraction facemask with or without osteotomy, maxillary advancement with distraction can be done with minimal or no alteration in mandibular position **[8]**.

For patients with maxillary hypoplasia, do may be of even greater importance as they lack bone and soft tissue. The slowly moving bony structures of the midfacial region are used as a framework for the overlying and expanding soft tissues **[14]**.

This process induces new bone formation along the vector of pull without the use of bone graft. Distraction forces applied to bone creates tension in the surrounding soft tissues, initiating sequences of adaptive changes in the soft tissues allowing larger skeletal movements while minimizing the relapse **[12]**.

The new bone that is formed by distraction osteogenesis is of the same morphology as the bones of the midface and maxilla **[15]**.

The decision of fistula closure either by bone graft alone or distraction osteogenesis or both was based upon the size of the fistula. Small sized fistulae (<3mm) were closed by bone graft alone, medium sized fistulae (3-5mm) were closed by distraction osteogenesis while large fistulae (>5mm) were closed by both distraction osteogenesis and bone graft.

For small sized fistulae <3mm in which there is adequate soft tissue coverage we did bone graft alone that was enough for closure of the gap. According to Semb, **[16]** mixed dentition bone grafting doesn't seem to have any adverse effect on maxillary growth in the years following the procedure.

In case of inadequate soft tissue coverage as in medium sized fistulae (3-5mm) Posnick and Thompson **[4]** did surgical gap closure through segmental repositioning of maxilla (Le Fort I osteotomies) with advancement of the maxillary arch anteriorly to close the gap and placing cancellous iliac crest bone graft posteriorly. The disadvantages of this technique were disturbance of maxillary and mandibular teeth relationship, a posterior dental gap that was created and possible worsening of velopharyngeal function which was borderline in these patients.

In our study segmental maxillary osteotomies and mobilization of the maxillary segment with attached palatal mucosa anteriorly for gap closure was done then the distractor device was applied at that position (by contraction). After a latency period of 7 days, this segment was mobilized posteriorly by the distraction device (reverse of distraction) thus avoiding the disturbance of maxillary teeth relationship to the mandibular ones and also to fill the gap that was created posteriorly after osteotomies and anterior mobilization of the maxillary segment.

For large fistulae (>5mm) there were inadequate soft tissue cover for bone grafting and limited mobilization of the maxillary segment anteriorly to close the gap due to the limited mobility of the attached palatal mucosa. Segmental osteotomies of the maxilla and mobilization of the maxillary segment anteriorly as much as possible was done followed by placement of iliac crest cancellous bone graft in the remaining bony gap and then after a latency period of 7 days, this segment was mobilized posteriorly by the distraction device (reverse of distraction).

Follow-up after 6 months and 1 year was done by clinical evaluation, X-ray skull occlusal view, lateral cephalometry and Cone Beam CT to assess the maxillary growth and closure of the gap with bone formation.

As regarding closure of the gap, in 60% of patients of Group I that were managed with bone graft alone, closure of the gap occurred after 6 months and this percent raised to be 70% after 1 year. In 50% patients of Group II that were managed with distractor alone, closure of the gap occurred after 6 months and this percent raised to 90% after 1 year. While in 50% patients of Group III that were managed by distrator and bone graft, closure of the gap occurred after 6 months and this percent raised to be 80% after 1year as shown by Cone Beam CT.

In patients of Group I, infection occurred in 30% (3 of 10 patients). Those patients received systemic antibiotics with antiseptic mouth wash but resorption of the grafted bone occurred with disruption of sutures leading to recurrence of fistula. These patients were re-operated upon 6 months later with re closure of the gap by another bone graft. All patients were closed successfully. In patients of Group II, infection occurred in 10% (1 of 10 patients) and along with failure of conservative management by systemic antibiotics and mouth wash for two weeks, loosening of the device