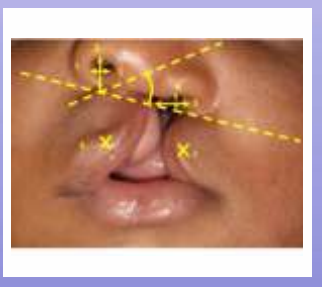
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PAR and self reported improvement of malocclusion



Orthodontic considerations of children with special needs



Pre-surgical infant orthopedics experience in LUTH



Management of bimaxillary protrusion

Orthodontic management of severe crowding



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Presurgical Infant Orthopaedics: The Lagos University Teaching Hospital Experience

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Abstract

Background: Presurgical infant orthopedics is an important aspect of the orthodontic management of children with cleft lip and palate. The aim of this study was to review the literature on its use and to highlight our experiences in the treatment of infants with different PSIO techniques at the Lagos University Teaching University (LUTH).

Methods: A literature review on PSIOs was conducted, in addition, a review of our experiences in the management of children with clefts at LUTH using different PSIO techniques such as the NAM and DNAM appliances was also done.

Results: A wide variety of PSIO appliances exist and the appropriate use of each technique is dependent on some factors including: the type of cleft, available resources, patient factors and operators' skill. The challenges experienced in LUTH in the use of these appliances include, inadequate record taking, loss of patients to follow-up, financial pressures, lack of patients' compliance, language barrier and irritation of the soft tissues in the children among others.

Conclusion: The article has presented a detailed review of different types of PSIO techniques and their clinical applications. The Lagos University Teaching Hospital experience in the use of PSIOs has been a learning curve, various challenges have been highlighted, and lessons have been learned. These lessons have helped in improvement of our treatment protocol.

Keywords: PSIO, LUTH, Experience

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ORIGINAL ARTICLE

Introduction

Cleft lip and palate (CLP) are the most common congenital malformation caused as a result of variation in the development of facial structures during gestation.[1] These anomalies affect 1 in every 700-1000 live births worldwide. [2,3] Butali et al [4] estimated the prevalence rate as 0.5 per 1000 in Nigeria.

Although causes of orofacial clefts are not completely understood, it has been suggested that it occurs as a result of interplay between multiple genes and environmental factors which influence the risk of orofacial clefts (OFC) either individually or through their interactions in complex biological pathways.[5]

Clefts involving lip, alveolus and palate result in disfigurement, distorted growth and development. Facial features depend on the severity of the cleft, the unilateral cleft deformity is characterized by a wide nostril base, separation of the upper lip on the cleft side, lateral and inferior displacement of the affected lower lateral cartilage, a depressed dome, increased alar rim, oblique columella, an overhanging nostril apex, shift in the base of the nose and deviation of the septum to non-cleft side. [1,6,7]

The bilateral cleft is characterized by a procumbent or rotated premaxilla, significant increase of the alar base width, widely separated lip segments, the flattened nasal tip is tethered directly to the prolabium by a severely deficient or absent columella, the lower lateral cartilages are flared or concave where they should be convex.[6–8]

Management of children with cleft lip and palate involves a multidisciplinary team who provides comprehensive diagnosis, planning and treatment. The cleft team includes the orthodontist, maxillofacial surgeon, plastic surgeon, pediatrician, paedodontist, speech therapist, ENT surgeon, prosthodontist, and psychologist. [2,9] All members of the team are equally important and they all work in tandem with each other to improve the patient's appearance, speech and function. The orthodontist is involved in the management pathway at a number of different stages from birth to adulthood[10] including:

Prenatal stage: To provide counselling and support to parents.

Neonatal and Infancy (Birth to 2 months): Prior to lip repair, an orthodontist is involved in a phase of oral orthopaedics to align the displaced cleft segment. This is known as Presurgical infant Orthopedics treatment.

Deciduous/Primary Dentition (2 years to 6 years) : no/minimal orthodontic intervention is done when there is a functional or psychological challenge.

Mixed dentition stage (7 to 12 years)

Early mixed dentition- fabrication of an upper removable appliance to procline the upper incisors and push them out of anterior crossbite**.**

Late mixed dentition- expansion of the narrow upper arch form to improve access for surgery prior to alveolar bone graft.

Permanent dentition stage (>12 years)

Early permanent dentition- orthodontic treatment at this stage may be conducted to relieve crowding, facilitate the eruption of canine, attempt correction of a developing skeletal class lll relationship, align the dentition, correct dental midline.

Late permanent dentition- most patients will present with moderate to significant class lll skeletal relationship, due to maxillary hypoplasia hence the need for orthognathic surgery or distraction osteogenesis.[10]

Presurgical Infant Orthopedics

Pre Surgical Infant Orthopaedics (PSIO) has been defined as the use of forces to reposition tissues secondarily displaced due to a cleft deformity.[1,11] Treatment is initiated in infants as soon as possible after birth usually within 7 days of life.[12] It engages the use of orthopaedic appliances for correction of Cleft Lip and Palate (CLP) defect using compressive and tensional forces or passively guiding growth and aims at reducing the width of the cleft defect, allowing a favorable alignment in the cleft segments within the first few months of infancy prior to surgical repair of the lip. It ultimately allows ease of surgical repair with minimal tension. [1]

Mode of action of PSIO

Matsuo[1,13–15] carried out a series of research on moulding of neonatal nasal cartilage and nostril using silicone tubes. He recognized that the cartilage in newborn is soft and the high level of estrogen at the time of birth correlates with the increased hyaluronic acid which in turn inhibits the linking of the cartilage intercellular matrix resulting in reduction in the elasticity of the cartilage, ligament and connective tissues.[6,12] The amount of plasticity in neonatal cartilage is highest after birth and gradually reduces as infant grows, the cartilage subsequently loses its pliability at around 6 weeks. Therefore, Presurgical Nasoalveolar Moulding is most successful within the first 3-4 months of life.[12]

Techniques of PSIO

Lip strapping

Cleft of the lip and alveolus of not more than 5mm can be managed using lip taping. This uses medical adhesive tapes such as Steri-strips or Durapore 3M tape placed across the upper lip, the tape offers outer pressure which approximate the upper cleft lip and alveolar gap segment, decreases the width of the nasal base and exerts a retracting and backward pressure against the protruding premaxilla in bilateral clefts thereby permitting the cleft segments to be anatomically located.[1,16] This ultimately assist lip repair under less tension so that healing is improved and scarring is minimized.

Dynacleft and Nasal elevators

Dynacleft ® is a prefabricated nasal and alveolar moulding device used to mould the upper lip, alveolus and nose prior to lip repair. Unlike the

traditional surgical tapes that were used in the past, Dynacleft® provides a constant approximation force with an elastic center that allows it to conform to the baby's mouth better because of its ability to contract and expand.[1]

The controlled force provided to the prolabium and premaxilla improves surgical result and reduces the need for early lip adhesion surgery. Dynacleft with a nasal elevator has provided results similar to NAM appliance, however unlike the NAM appliance, it does not require adjustments with growth of the infant.[1]

Nasoalveolar Moulding

The paradigm shifts in the PSIO treatment came in 1993, when Grayson and Cutting[17] introduced Nasoalveolar molding (NAM) in which presurgical molding of the alveolus, lip and nose is carried out in newborns with Cleft lip and palate.

The older PSIO appliances that were used for most cleft treatment protocol were designed to correct the alveolar cleft only with no recognition and management of the nasal deformity. Most nasal deformities are addressed surgically and the patients usually require quite a number of secondary surgical revisions as they grow up. [1,18]

Grayson[17] described a new technique to mould the lip, alveolus and nose in infants born with CLP before surgery. The NAM appliance is the only Presurgical orthopedic technique that directly addresses both the cleft nasal and alveolar deformity.[18,19] It works on Matsuo's principle which described that the nasal cartilage could be moulded due to increased plasticity concurrent to increased level of maternal oestrogen if treatment is initiated within the first six weeks of life.[1,13,15,20]

The NAM appliance has an intraoral moulding plate and a nasal stent. The intraoral moulding plate gradually approximates the displaced alveolar segments resulting in approximation of the cleft segments as well as the medial and lateral aspects of the nose. The nasal stent reshapes and projects the nasal tip while correcting the corresponding deformity of the lower lateral alar cartilage in unilateral CLP and lengthening of the columella in bilateral CLP.[18]

Indications for the use of the nam appliance

Patient selection -

Age - Satisfactory results are better achieved if treatment is commenced within six weeks of life. This is because of high levels of maternal estrogen in the fetal circulation which triggers an increase in hyaluronic acid which in turn increases the plasticity of the neonatal cartilage, The use of NAM appliance takes advantage of the high plasticity of the neonatal cartilage in the early postnatal period.

Cleft type - NAM appliance is indicated in patients with complete unilateral cleft lip and palate with associated nasal deformity.

Also indicated in cases of complete bilateral cleft lip and palate with severe displacement of the premaxillary segment, a wide nasal tip and alar base and a severely deficient columella.

Parental compliance- because of the dependent nature of the infants on the parents /caregiver, the success of NAM appliance depends largely on the parents' positive attitude and commitment. The use of NAM appliance is indicated when parents who are taking care of the babies with cleft do not see the procedure as an additional burden on them but are willing to comply.

Parental availability- Since treatment with NAM requires frequent hospital visits for adjustment of the moulding plate and monitoring of treatment progress, patients' selection should be done based on the availability of the parents/caregiver.

Objectives of the NAM appliance

In unilateral cleft lip and palate, the objectives of NAM appliance are: mainly to reduce the severity of the original cleft deformity by reducing the width of the alveolar cleft segments and aligning the base of the nose and the lip segments.[1,17]

As the alveolar ridge and maxilla improve in relation to each other, there is a concurrent improvement of the overlying soft tissues.

The alar rim which was initially stretched over a wide alveolar cleft deformity becomes lax enabling it to be elevated into a symmetrical and convex form.[1,17–19,21]

In Bilateral cleft lip and palate, objectives are:

To achieve non-surgical elongation of the columella.

To center the premaxilla along the mid sagittal plane.

To retract the premaxilla, to ensure continuity with the posterior alveolar cleft segments.

To reduce the width of the nasal tip and improve nasal tip projection.[1,17–19,21]

Benefits of NAM

It allows for proper alignment of the lip, nose and alveolus before primary lip and nose repair. This ultimately enables the surgeon to achieve a better and a more predictable outcome with less tension and less scar tissue formation.[1,6,22,23]

The stable change in nasal shape with less scar tissue, allows for better lip and nasal form. [6,24]

It reduces the number of surgical revisions for excessive scar tissue, oronasal fistulas, nasal and labial deformities[1,7].

It allows for better positioning of the alveolar segments, and increased bone bridges across the clefts, thereby allowing the permanent teeth have a better chance of eruption in a good position with adequate periodontal support. [1,6,21]

Complications of NAM

Complications associated with NAM therapy can be classified into soft tissue, hard tissue and appliance related.

Soft tissue complications

Mucosal ulceration- may be caused by under relieved areas of the fitted surface of the moulding plate, excessive activation of the internal surface of the appliance resulting in impingement of the mucosa[21,25]. The area under the prolabium may also get ulcerated if the strapping is too tight[1]. Ulcerations can be resolved by lubricating the mucosa interface, smoothening rough surfaces and relieving overcontoured areas[25].

Tissue fungal infections- the full-time wear of the NAM appliance and contact with the oral microflora results in a potential reservoir for infectious organisms. If the moulding plate is not removed daily and cleaned regularly, a candida infection may occur[25,26].

Fungal infection is treated with Nystatin or Amphotericin ointment.

Skin ulceration - frequent application and removing of the extraoral surgical tape used to approximate the lip segment and secure the appliance may cause irritation of the facial epidermal tissues especially in the zygomatic process area. [1,25] This can be prevented by using Duoderm® or 3MTegaderm® underneath the strapping. In addition, the overlying retraction and horizontal tape must be removed once a day usually at bath time and hydration cream should be applied to the cheek. Each time the tape is changed, its position on the patient's face should be varied, this allows epidermal tissue to ventilate.

Tape removal solvent or warm water can facilitate the removal of the tape.

Mega-nostril - is the distended nasal aperture created from excessive tension on the flexible lower nasal alar cartilage[25,27]. This may occur when the stent application is initiated before the size of the cleft gap is adequately reduced, the premature nasal stenting exerts excessive force against the nasal tissue resulting in excessive alar expansion[1]. It may also occur due to improper stent positioning, or nasal over contouring which causes the stent activated forces to be concentrated on the alar rim rather than the nasal dome[25,28]. This is avoided when the nasal stent is applied and activated by a qualified practitioner.

Hard tissue complications

Locked out segment- occurs due to poor and involuntary moulding process, wherein the greater segment moves more rapidly without a change in the position of the lesser segment. As a result, the lesser segment gets locked out behind the greater segment. [1]

Premature eruption of the maxillary incisors as a result of pressure exerted by the moulding plate.[25,29]

Appliance related.

Dislodgement of the acrylic plate- occurs if the arms of the appliance are tapped too horizontally or the appliance is under activated causing obstruction of the airway[1,6]. A 5mm diameter hole placed in the center of the moulding plate at fabrication allows for

passage of air in the event that the molding plate drops down from the rear and causes obstruction.[6]

Treatment protocol

Following arrival in the outpatient cleft clinic at the Lagos University Teaching Hospital, the parents of the infants with clefts are seen by the cleft team. Those infants who require PSIO are referred to the Cleft Orthodontic clinic.

At the Orthodontic clinic, proper documentation is done:

A detailed history of each child is taken.

Detailed examination (extraoral and intraoral examination) is carried out. This includes detailed measurement of the cleft size.

Parents are counselled, educated and informed on the benefits, proper usage and duration of wear of the NAM appliance.

Informed consents for photographs and PSIO treatment are taken from the parents/guardian.

Records are taken, this includes baseline photographs (extraoral and intraoral views), primary impression for fabrication of study cast and special tray.

Extraoral views taken include Frontal, Right and Left profiles, Basal view while the intraoral view taken is the maxillary arch using the palatal mirror.

Treatment consists of two main parts: a period of alveolar moulding which is ideally initiated within 7 days after birth, this period lasts for about 6-8 weeks or when the cleft is reduced to 5mm or less and a subsequent period of nasal moulding (6-8weeks) after the main correction of the greater alveolar segments have been achieved.

Procedure for fabrication and use of NAM

A heavy bodied poly vinyl siloxane impression material (PVS) is used to take the primary impression as soon as after birth as possible when the cartilage is plastic and moldable for the fabrication of study cast and special tray. The tray is seated until the impression material is observed to just begin to extrude past the tray borders, the head of the infant is inclined forward to keep the tongue in a forward position and to allow fluids to drain out of the oral cavity.

Once the impression material is set, the tray is removed and the mouth is examined for any residual impression material that may be left behind. The impression is poured with dental stone and a study cast is fabricated. A special tray is fabricated on the study cast, and a secondary impression is taken at the patient next visit.

Following this, a secondary impression is taken, a working/master cast is made and NAM (the moulding plate) is fabricated on the working cast. It is made of a clear acrylic and is lined with a thin layer of denture material. The plate is inspected carefully for areas of overextension which may cause soft tissue ulceration if not relieved.

The appliance is secured extraorally to the cheeks bilaterally by surgical tapes which have orthodontic elastic band at each end. The retention arm is positioned at an angle of 40 degrees down from the horizontal to prevent unseating of the appliance from the palate.

Instructions are given to the parents to keep the plate in the baby's mouth full time, to take it out for cleaning at least once in a day, and to change the tape once wet or dirty. The patient visits the clinic weekly to modify the moulding plate to gradually approximate the alveolar segments and reduce the cleft size (the modification is achieved through the selective removal of acrylic from the region where one desires the alveolar bone segment to move while at the same time, soft denture liner is added to the appliance in the region from which one desires the alveolar segment to be reduced) and to monitor treatment progress.

Closure of the alveolar gap brings the lip segments together, reduces the nasal base width and allows for laxity of the alar rim. The nasal stent should not be added before achieving laxity of the alar rim to prevent an increase in the nostril circumference known as mega nostril. When the alveolar segment is reduced to 5mm or less, the nasal stent is incorporated.

The nasal stent is made from 0.036 guage round Stainless-Steel wire with an acrylic bulb. It is attached to the labial flange of the moulding plate

near the base of the retention arm. It extends forward and then curves backward in the form of a swan neck entering 3-4mm past the nostril aperture. As the wire extends to the nostril, it curves back on itself to create a small loop for retention of the intranasal portion of the nasal stent. This hard acrylic portion is shaped into a bilobed form, a layer of soft denture liner is added to the hard acrylic for comfort. The upper lobe enters the nose and gently lifts the dome until a moderate amount of tissue blanching is seen while the lower lobe lifts the nostril apex and defines the top of the columella.

Patients' instructions

The following instructions are given to the parents for the care of the babies and the NAM appliance:

The appliance must be worn by the patients 24 hours a day, 7 days in a week including when they are feeding.

The appliance is secured extraorally to the cheek and bilaterally by surgical tapes which have an orthodontic elastic at each end. The elastics are attached to the retention arm of the appliance.

Surgical tapes are placed to approximate the cleft segments. The tape should be applied at the base of the nose and not low on the lip near the vermillion border, to prevent undesirable horizontal lengthening of the lip overtime.

The tape should be applied to the non-cleft side first and then pulled over and adhered to the cleft side.

The appliance must be cleaned regularly preferably every morning using water and Milton® tablets.

The tape should be replaced every morning or once it is dirty from feeds.

A thin layer of petroleum jelly should be added on the nasal stent at the time of every insertion to prevent irritation of the nasal mucosa.

The patient should be brought to the clinic every 1 to 2 weeks to modify the molding plate to guide the alveolar cleft segments into the desired position.

Assessment of treatment progress

In order to objectively assess treatment progress, certain standardized measurements are done both on the patients and on the casts pre and post treatment using a pair of dividers and a metre rule.

Extraoral measurements carried out as described by Rau et al [30]

Intersegmental lip distance (ISLD): This is the distance from the right lip (RL) to the left lip (LL) segment or the corresponding premaxilla lip margin.

Nostril height of the cleft (NHc) and non-cleft side (Nhnc): This is the distance from the highest point of the nostril to the alar base (AB) line (connecting line between the right and the left alar base).

Nostril width of the cleft (NWc) and non-cleft side (Nwnc): The distance from the point farthest right to the point farthest left of the nostril.

Columella deviation angle (CDA): angle between the columella axis and the alar base line in unilateral CL/P.

Premaxilla deviation angle (PDA): angle between the premaxilla axis and a vertical line to the vomer in bilateral CLP.

Columella length (CL): distance from the base of the nose to the highest point of the columella in bilateral CLP.

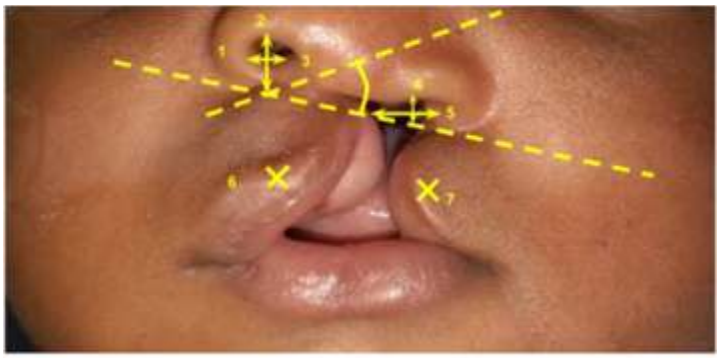


Figure 1: Landmarks on Unilateral Cleft Lip and Palate

1. - Nasal width on the non-cleft side
2. - Nasal height on the non-cleft side
3. - Columellar Deviation Angle
4. - Nasal height on the cleft side
5. - Nasal width on the cleft side
6. - Right Lip
7. - Left Lip



Figure 2: Landmarks on Bilateral Cleft Lip and Palate

1. - Right Lip
2. - Right Premaxillary Lip
3. - Left Premaxillary Lip
4. - Left Lip
5. - Columellar Length

Table 1: Measurement on the patient

Measurements	Explanantion
Bialar Width	Measurements between right and left ala of the nose
Columellar Width	Width of columella at base
Columellar Length	Distance from the base of the columella to the most anterior and inferior point of the nasal tip
Nostril Height	Maximum vertical Length from the floor to the inner aspect of the ala of the nose
Nostril Width	Maximum horizontal length at alar base
Columellar angle on cleft side	Angle formed by joining a line from nasal tip to intercolumellar line and calculated from the cleft side.

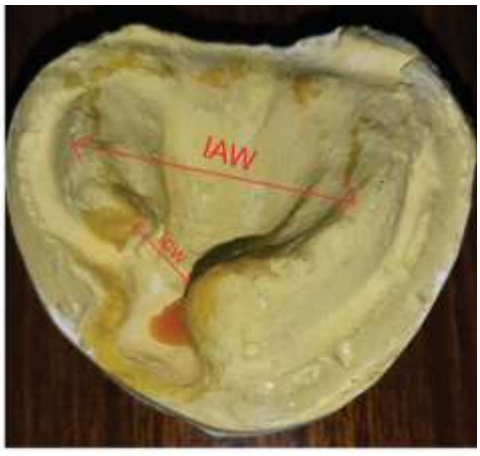


Figure 3: ICW - Intraoral Cleft Width, IAW - Intraoral Arch Width

Table 2: Measurement on the cast

Measurements	Explanation
Intraoral cleft width	Distance from the right alveolar crest to the left alveolar crest
Intraoral arch width	Maximum horizontal measurement at crest of alveolar ridge

Lagos University Teaching Hospital Experience: Challenges Encountered and Lessons Learned

Challenges:

Operational Factors

Availability of Equipment and Instruments - Lack of scanning machine which can be used in making extraoral and intraoral scans needed for making study models and the NAM appliance.

Doctor Factors

Deficient Views of Photographs

Initially, not all the required profiles of extraoral photographs were taken, so we had incomplete records of patients extraoral photographs.

Intraoral photographs (maxillary arch views) were taken without palatal mirrors.

Unstandardized Measurements

Standardized measurements were not taken pre and post treatment to assess treatment progress, we simply eyeballed and compared final result with previous photos that were taken before the initiation of treatment. This method is subjective and forestalled objective assessment of treatment progress.

Patient Factors

Loss Of Patients to Follow Up - few patients came into the clinic, intraoral impressions were taken for fabrication of special tray and NAM but they did not come back to complete these procedures while others were lost after the appliance was fitted.

Some of the patients are living a far distance from the hospital and had to travel a long distance before arrival at the clinic

Finances available for transport also affected some parents' compliance

Language Barrier- we had parents that do not speak and understand English language, so we had difficulties communicating effectively with these parents. This in turn affected our treatment.

Few parents also attempted to boycott PSIO treatment and presented themselves to the Oral and maxillofacial Surgery clinic for cheiloraphy while they were still on PSIO treatment without being certified ready for surgery by the orthodontist.

Some patients could not tolerate the NAM appliance when fitted, the appliance irritated the soft palate, she kept retching and so there was a need to modify the appliance.

Lessons learnt

We learned to take the complete profiles of extraoral radiographs. This includes frontal, right profile, left profile, basal views.

We now have suitably sized palatal mirrors for newborns, allowing for standard maxillary arch view.

Standardized measurements are now taken pre and post treatments using a pair of divider and meter rule in order to allow for objective assessment of treatment progress.

Enquires must be made about parents' availability and financial ability to be able to bring the babies to

the clinic for treatment and financial help should be provided for those that lack finances for transportation.

Speak the language they understand for effective communication and a good treatment outcome.

Constant communication with patients through phone calls, whatsapp videos, photos sent by parents encourage parents and help to show parents we are in this together as this is often a traumatic period for them.

There is a need to work as a team, the full cooperation of the oral and maxillofacial surgeon with the orthodontists is required. The Orthodontist must confirm a patient eligibility for surgery having completed the PSIO treatment before the surgeon carries out the lip repair.

Treatment should be individualized, there may be a need to modify or change the appliance type in some cases.



Pretreatment



Naso-alveolar moulding Appliance



Appliance in patient's mouth



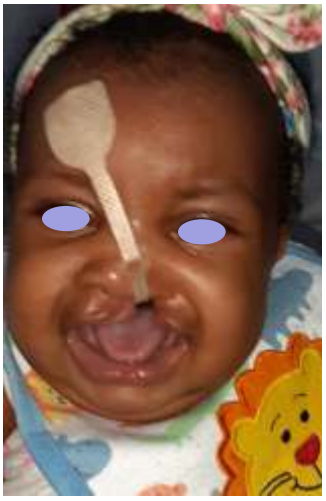
Post Treatment



Pretreatment



Aligner Appliance



Appliance in patient's mouth



Post Treatment

Conclusion

This article has presented a detailed review of the different types of PSIOs and their clinical applications in the orthodontic preparation of children born with clefts for surgical repair. We have also highlighted our experience at the Lagos University teaching hospital in the use of PSIOs, citing the challenges experienced and the lessons learnt so far.

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