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AESTHETIC

Case Report

BEYOND THE STANDARD: ACHIEVING FUNCTIONAL AND AESTHETIC EXCELLENCE IN UNILATERAL MACROSTOMIA REPAIR WITH BUCCAL FLAP AND MULTI Z-PLASTY TECHNIQUE IN INFANCY

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ABSTRACT

Introduction: Macrostomia is a rare congenital craniofacial anomaly that significantly affects feeding, speech, aesthetics, and psychosocial well-being. Early surgical intervention using advanced techniques is essential to restore anatomical integrity and improve functional outcomes. This case report presents the management of a five-month-old female infant with a unilateral left-sided macrostomia *(Tessier Cleft-7)* using a combination of *buccal flap* and multiple *Z-plasty* techniques.
Case Presentation: The patient presented with a unilateral left-sided macrostomia since birth, accompanied by feeding difficulties characterized by choking and audible "*blup-blup*" sounds during feeding. Physical examination confirmed a moderate unilateral macrostomia without signs of infection or systemic illness. Preoperative assessments, including laboratory tests and multidisciplinary consultations, confirmed the patient's fitness for surgery.
Discussion: Surgical repair was performed with a multi-layer closure approach, utilizing a buccal flap for the inner layer and multiple *Z-plasty* for the outer layer to achieve tension-free closure and optimal aesthetic results. This technique aligns with contemporary trends favoring individualized, anatomy-driven repairs over traditional methods. The absence of intraoperative and postoperative complications and the satisfactory functional and cosmetic outcomes underscore the effectiveness of this approach. Compared to conventional linear closure techniques, multiple *Z-plasty* minimize scar contracture and enhance commissural symmetry, particularly in complex cases of macrostomia. This case highlights the critical role of multidisciplinary collaboration and tailored surgical planning in addressing the anatomical challenges of macrostomia and achieving optimal reconstructive outcomes.
Conclusion: Early, multidisciplinary management of macrostomia using combined *buccal flap* and multiple *Z-plasty* techniques can achieve excellent functional and aesthetic outcomes with minimal complications. This case contributes to the growing literature supporting anatomy-specific surgical approaches and comprehensive perioperative care in macrostomia reconstruction.

***Keywords**: Macrostomia; Buccal flap; Z-plasty; Labial reconstruction*

Latar Belakang: Makrostomia merupakan kelainan kraniofasial kongenital yang jarang terjadi dan secara signifikan memengaruhi fungsi makan, bicara, aspek estetik, serta kesejahteraan psikososial. Intervensi bedah dini dengan teknik yang mutakhir sangat penting untuk memulihkan integritas anatomis dan meningkatkan luaran fungsional. Laporan kasus ini menyajikan tata laksana seorang bayi perempuan usia lima bulan dengan makrostomia unilateral sisi kiri *(Tessier Cleft-7)* menggunakan kombinasi teknik *buccal flap* dan *multiple Z-plasty*.
Presentasi Kasus: Pasien datang dengan keluhan makrostomia unilateral sisi kiri sejak lahir, disertai kesulitan menyusu berupa episode tersedak dan bunyi "*blup-blup*" yang terdengar saat proses pemberian makan. Pemeriksaan fisik mengonfirmasi adanya makrostomia unilateral derajat sedang tanpa tanda infeksi maupun gangguan sistemik. Evaluasi prabedah, termasuk pemeriksaan laboratorium dan konsultasi multidisipliner, menyatakan pasien dalam kondisi layak menjalani pembedahan.
Hasil: Rekonstruksi dilakukan dengan pendekatan penutupan berlapis (multi-layer closure), menggunakan buccal flap pada lapisan dalam dan multiple *Z-plasty* pada lapisan luar untuk mencapai penutupan tanpa tegangan serta hasil estetik yang optimal. Teknik ini sejalan dengan tren bedah rekonstruksi kontemporer yang mengutamakan pendekatan individual berbasis anatomi dibandingkan metode konvensional. Tidak ditemukannya komplikasi

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intraoperatif maupun pascaoperatif, serta diperolehnya hasil fungsional dan kosmetik yang memuaskan, menegaskan efektivitas pendekatan ini. Dibandingkan teknik penutupan linear konvensional, multiple *Z-plasty* mampu meminimalkan kontraktur jaringan parut dan meningkatkan simetri komisura labialis, terutama pada kasus makrostomia yang kompleks. Kasus ini menegaskan peran krusial kolaborasi multidisipliner dan perencanaan bedah yang terpersonalisasi dalam mengatasi tantangan anatomis makrostomia serta mencapai luaran rekonstruksi yang optimal.

Simpulan: Penatalaksanaan makrostomia secara dini dan multidisipliner dengan kombinasi teknik *buccal flap* dan multiple *Z-plasty* dapat menghasilkan luaran fungsional dan estetik yang sangat baik dengan angka komplikasi minimal. Laporan ini menambah bukti ilmiah yang mendukung pendekatan bedah berbasis anatomi serta perawatan perioperatif komprehensif dalam rekonstruksi makrostomia.

Kata Kunci: *Makrostomia; Buccal flap; Z-plasty; Rekonstruksi labial*

Conflicts of Interest Statement:
The authors listed in this manuscript declare the absence of any conflict of interest on the subject matter or materials discussed.

INTRODUCTION

Lateral cleft of the lip, or macrostomia, also known as Tessier cleft type 7, is one of the rarest craniofacial anomalies and is defined as an abnormal widening of the mouth at the oral commissure. The incidence varies between 1 in 60,000 to 1 in 300,000 live births[1]. Macrostomia can present as a unilateral or bilateral anomaly with a partial or complete cleft that significantly affects to functional, aesthetic, and psychosocial implications for patients[1,2,3]. This lateral cleft is due to failure of fusion of the maxillary and mandibular processes of the first and second branchial arch or due to disruption in the processes after fusion during the 5th to 8th week of embryogenesis[1,3,4]. It is commonly associated with macrostomia and defects of the first branchial arch. The patients commonly also have deformities like preauricular tags, sinuses and Goldenhar syndrome (bilateral lateral clefts). Approximately 80–90% of cases are unilateral, often affecting the right side and more commonly seen in males[4]. While genetic and environmental factors contribute to its etiology, early surgical intervention remains the cornerstone of management to restore anatomical integrity and improve quality of life[5,6].

The timing and technique of macrostomia repair are critical for optimizing outcomes. Recent advances emphasize layered closure techniques, such as multi-layer reconstruction with buccal flaps and Z-plasty, which enhance both structural stability and aesthetic results[7]. Despite standardized protocols, perioperative strategies-including antibiotic use, suture materials, and postoperative care-vary significantly across centers, underscoring the need for evidence-based approaches[8,9]. For instance, studies demonstrate that early primary repair in infancy minimizes long-term maxillary growth disturbances while maintaining low complication rates[10].

This case report highlights the successful management of a five-month-old female infant with a unilateral left-sided macrostomia (Tessier Cleft-7) using a combination of buccal flap reconstruction and multiple Z-plasty. The patient's presentation characterized by feeding challenges and absence of familial predisposition-aligns with typical clinical profiles of isolated macrostomia cases[2]. The surgical approach, supported by multidisciplinary preoperative assessments, reflects contemporary practices prioritizing functional restoration and minimal scarring[11]. By detailing this case, we aim to contribute to the growing literature on optimized surgical techniques and perioperative protocols for macrostomia repair, particularly in resource-constrained settings where delayed presentations remain common[12].

CASE PRESENTATION

A five-month-old female infant was brought to Dr. Moewardi General Hospital by her family with the chief complaint of a left-sided macrostomia, which had been apparent since birth. The family reported that the patient experienced frequent choking and produced a "blup-blup" sound when feeding, indicating difficulties during the feeding process. There was

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no family history of similar congenital anomalies. Informed consent was obtained from the patient's parent for documentation in publication and research purposes. The patient was planned for labial reconstruction surgery to correct the defect. Upon admission and during the preoperative period, her general condition was moderate with stable vital signs, and she remained able to feed despite the noted difficulties. No comorbidities or other aggravating medical conditions were identified during the assessment.

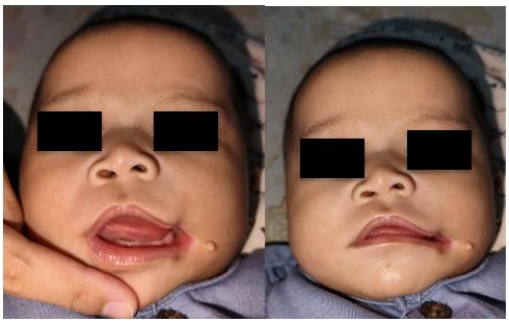

Figure 1. Cracks on the lips before labial reconstruction surgery

On physical examination, the patient was found to be in a moderate general condition with compos mentis. which are appropriate for her age of five months. Local examination of the facial region revealed a distinct defect in the left labium oris, consistent with a unilateral macrostomia (Tessier Cleft-7). There were no signs of local infection, edema, or other complications in the perioral area. These physical findings support the diagnosis of a moderate degree facial cleft without accompanying systemic disturbances or aggravating conditions.

The supporting examinations performed in this patient included a routine preoperative blood laboratory assessment, which revealed results within normal limits, indicating the absence of active infection or hematological abnormalities that could hinder surgical intervention. No additional radiological imaging was conducted, as there were no clinical indications necessitating further diagnostic imaging. As part of the preoperative preparation, consultations with the departments of anesthesiology and pediatrics were also completed, both confirming that the patient was fit to undergo general anesthesia and labial reconstructive surgery. Overall, these supporting examination findings corroborated the diagnosis of facial cleft (Tessier Cleft-7) and confirmed the patient's readiness to proceed with the planned surgical procedure without other medical impediments.

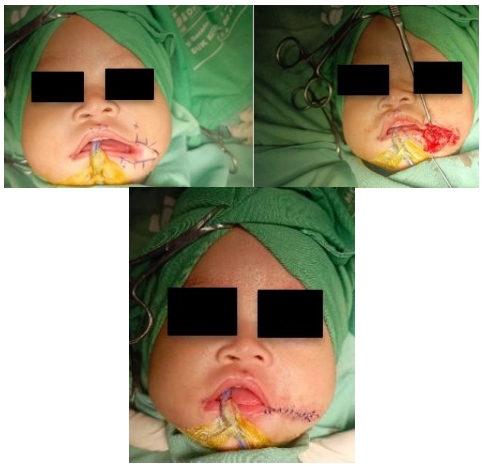

Figure 2. Durante labial reconstruction surgery

The surgical procedure for this patient was performed in April 2025. The technique involved closure of the inner layer using a buccal layer, while the outer layer was reconstructed with multiple Z-plasty, followed by layered suturing to ensure both structural strength and optimal aesthetic outcome. Throughout the procedure, there were no intraoperative complications or difficulties encountered. No implants were used, and no tissue specimens were sent for anatomical pathology examination. Intraoperative bleeding was well controlled, with a total of 10 gauze pads used.

Z-plasty is a plastic surgery technique used to improve the function and appearance of scars. This technique involves transposing two triangular flaps of equal size, forming a "Z" shape, to reduce skin tension and enhance the scar's appearance. A flap is a section of skin (and underlying tissue) that is moved while maintaining its blood supply. Based on vascularization, flaps are classified as either random flaps or axial flaps. In this case. Z-plasty make the flaps an 60° angle with equal limb lengths. This technique can lengthen the scar by about 25–30% and improve its cosmetic appearance, especially in contracture scars or localized small scars. For longer scars, multiple Z-plasty may be required.

The operation was completed without incident, resulting in a stable and satisfactory reconstruction of the lip. Postoperatively, the patient was admitted for inpatient care with

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instructions to maintain a semi-Fowler's position, without the placement of drains or nasogastric tubes. The patient received intravenous ampicillin at a dose of 20 mg/kgBW every 8 hours and paracetamol at a dose of 15 mg/kgBW every 8 hours to prevent infection and manage pain.

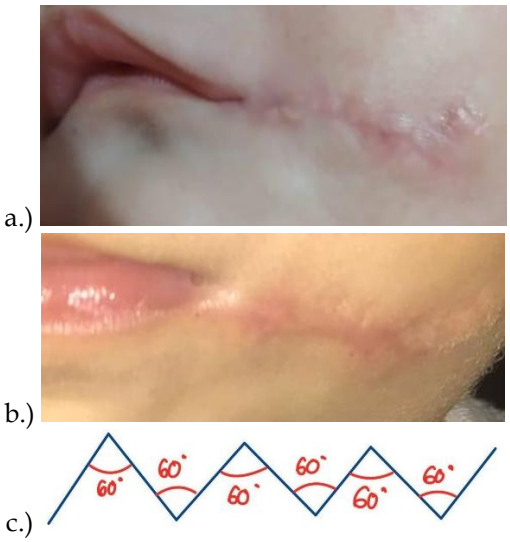

Figure 3. a) The patient's condition 1 week after surgery b) The patient's condition 3 months after surgery c) Illustration of Parallel Multiple Z-Plasty on this Patient

DISCUSSION

Macrostomia also known as commissural or lateral facial cleft (Tessier no. 7) is a rare congenital anomaly resulting from failed fusion between maxillary and mandibular processes during embryogenesis[1]. The pathogenesis of macrostomia is considered multifactorial. Proposed etiological mechanisms include mutations in craniofacial regulatory genes (TCOF1, POLR1C/D, HOXA2, OTX2), intrauterine vascular disruptions, and teratogenic exposures such as alcohol or retinoic acid during early pregnancy[4,13,14]. The clinical impact extends beyond the visible deformity, as it can compromise feeding, increase the risk of aspiration, and lead to speech and dental problems if not addressed promptly. Early intervention is crucial to minimize these risks and support normal facial development and psychosocial[15]. The management of macrostomia is not only a surgical challenge but also a multidisciplinary endeavor, as it influences feeding, speech, psychosocial development, and overall quality of life. Each case presents unique anatomical and clinical considerations, and reporting individual experiences contributes to the refinement of surgical techniques and perioperative care, especially in complex or atypical presentations[15].

The gold standard for macrostomia repair involves early surgical intervention, typically performed between three to six months of age to optimize both functional and aesthetic outcomes[12]. The most widely adopted techniques include the Millard rotation-advancement and Tennison-Randall triangular flap, both designed to reconstruct the orbicularis oris muscle, restore lip continuity, and achieve symmetry of the Cupid's bow and philtrum. Recent modifications, such as the incorporation of Z-plasty or additional flaps, aim to prevent vermillion notching, reduce scar contracture, and address nostril deformities. Multidisciplinary care, including perioperative orthodontic, pediatric, and anesthetic support, is essential for comprehensive management and to ensure optimal healing and long-term function[16].

In the present case, a five-month-old female infant with a unilateral left-sided macrostomia underwent labial reconstruction using a multi-layer closure technique, with the inner layer repaired using a buccal flap and the outer layer closed with multiple Z-plasty, followed by meticulous layered suturing. This approach was chosen to provide a tension-free closure, optimize anatomical restoration, and minimize the risk of linear scar contracture. Compared to the more conventional Millard or Tennison-Randall techniques, the use of multiple Z-plasty in this case allowed for better redistribution of tissue, improved camouflage of the scar, and enhanced aesthetic outcomes, as supported by recent literature[16]. Postoperative management included semi-Fowler positioning and administration of intravenous ampicillin and paracetamol, with no drains or nasogastric tubes used, reflecting a trend toward less invasive postoperative care.

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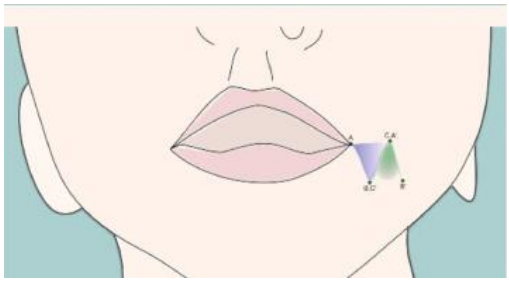

Figure 4. *Cupid's Bow Diagram for Multiple Z-Plasty on Macrostomia Patient.*

The surgical approach in this case aligns with contemporary trends favoring individualized, anatomy-driven repairs over a one-size-fits-all technique. While the Millard and Tennison-Randall techniques remain the mainstay for most unilateral cleft lips, modifications such as Z-plasty have been shown to further reduce the risk of vermillion notching and unsightly scarring, particularly in complex or wide clefts. Studies highlight that these modifications can result in more natural lip contours and improved long-term outcomes, particularly in terms of symmetry and scar appearance. The suturing technique for patients undergoing multiple Z-plasty aims to prevent scarring, relieve tension, and maintain the symmetry and continuity of the skin line. For the re-aproximation, the first flap is transposed in the opposite direction to the next one (zig-zag). After all the flaps are positioned, check the length of the new line, because escess skin can be 'dog-ear' (skin protrusion at the tip. Skin re-approximation begins at the midpoint of each Z (intersection) and proceeds laterally. In contrast, cases managed with traditional straight-line closures or without layered repair may face higher revision rates due to functional or aesthetic dissatisfaction[17].

This case is notable for the anatomical complexity of the Tessier Cleft-7, involving both labial and palatal elements, which required precise surgical planning and execution. The combination of buccal flap for the inner layer and multiple Z-plasty for the outer layer is relatively uncommon and demonstrates the adaptability of cleft repair techniques to unique anatomical challenges. The absence of intraoperative and postoperative complications, as well as the stable, aesthetically pleasing result, underscores the value of a tailored, multidisciplinary approach. Furthermore, the decision to forgo routine use of drains or nasogastric tubes reflects evolving best practices aimed at minimizing patient discomfort and promoting faster recovery. This case reinforces the importance of individualized surgical planning, meticulous technique, and comprehensive perioperative care in achieving optimal outcomes for complex cleft lip presentations[17]. Z-plasty's flexibility makes it highly adaptable to the unique needs of macrostomia cases, allowing it to be used alone or alongside other reconstructive methods. Its proven ability to improve both function and appearance is well-supported by research[18]. After a three-month postoperative evaluation, a complication was observed in the form of scar formation in this patient; however, the scar was minimal and did not affect tissue elasticity. The patient was able to laugh freely, and no impairment in mouth opening was noted.

CONCLUSION

In summary, this case highlights that early and multidisciplinary management of a five-month-old female infant with a unilateral left-sided macrostomia (Tessier Cleft-7), utilizing a combination of buccal flap for the inner layer and multiple Z-plasty for the outer layer, can result in optimal functional and aesthetic outcomes with minimal complications. The patient's clinical presentation-characterized by feeding difficulties and the absence of systemic comorbidities-was managed through careful preoperative assessment, individualized surgical planning, and evidence-based perioperative care, reflecting current best practices in cleft lip reconstruction. This approach not only addressed the anatomical complexity of the defect but also minimized postoperative morbidity, this surgical strategy contributes to the growing literature supporting tailored techniques aimed at improving the quality of life in patients with rare congenital craniofacial conditions. The single-case nature of this report limits generalizability; future studies should evaluate long-term outcomes in larger cohorts. Broader Implications: This case supports the use of multiple Z-plasty in infancy for macrostomia, highlighting its potential to enhance functional, minimal complication and aesthetic outcomes in complex craniofacial reconstructions.

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