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A CASE REPORT: ENHANCING TASAL AND MUSCULAR SUPPORT FOR ECTROPION CORRECTION IN TESSIER 3 AND 5 FACIAL CLEFT

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ABSTRACT

Introduction: Facial cleft is a rare and challenging craniofacial malformation with a low incidence ranging from 0.75 to 5.4 per 1000 common cleft. Due to the high variance in its occurrence, good techniques have not been established yet.

Case Illustration: In this case study, we present the medical history of a 45-year-old woman who previously underwent multiple surgeries for the reconstruction of her lower eyelid. The initial surgeries were performed to address a unilateral facial cleft classified as Tessier 3 and 5, utilizing ear cartilage grafts for the tarsal plate. However, she subsequently experienced complications, including ectropion, epiphora, and soft tissue deformities around her left eye. The goal of the current surgical intervention is to rectify the ectropion and enhance overall facial aesthetics for improved outcomes.

Discussion: The primary objectives of this surgical techniques are to enhance the tarsal support through the tarsal strip technique and to provide muscle support using the mid-face lifting technique. Additionally, we removed the scar and excess tissue from the lower lid to adjust the tarsal's proper length. These methods aim to address the ectropion and prevent its recurrence.

Conclusion: This combination of techniques can be a potential alternative for rectifying ectropion by reinforcing both tarsal and muscular support structures.

Highlights:

1. The oblique facial cleft is an uncommon and intricate craniofacial anomaly characterized by a low occurrence rate and significant variability.
2. The treatment is intricate and relies on the surgeon's expertise and discernment.
3. Utilizing a combination of Tarsal Strip and Midface Lifting techniques could serve as an alternative approach to address ectropion by enhancing both tarsal and muscular support.

INTRODUCTION

Facial cleft is a rare and challenging craniofacial malformation.[1] Its prevalence varies, ranging from 0.75 to 5.4 per 1000 cases of common clefts.[2] In 1976, Paul Tessier established a classification system for craniofacial malformations.[3] This system relies on direct anatomical observations derived from clinical examinations and surgical dissections. Malformations are assessed based on their association with either the lip and upper jaw system or the eyelid and orbital cavity system.[4]

Tessier number 3 cleft, referred to as the medial orbito-maxillary cleft, extends diagonally from the lacrimal groove, encircling the base of the nose and upper lip, resulting in a cleft lip. In severe instances, the frontal process and medial wall of the maxilla may be entirely absent.[3] In the case of Tessier Number 5, known as a lateral orbito-maxillary cleft, a gap initiates near the oral commissure on the upper lip, extending as a groove across the cheek before concluding at the junction of the middle and lateral thirds of the lower eyelid. Bone involvement typically encompasses an alveolar cleft in the premolar region, extending across the maxilla to the lateral side of the infraorbital nerve, reaching up to the infraorbital rim and orbital floor. This lack of formation results in a shortened distance between the oral commissure and the lower eyelid, with the medial cantus usually positioned normally, while the lateral canthus is generally dystrophic.[5]

The etiology of craniofacial clefts remains unclear, with the most plausible explanation emerging from developmental factors. The loss of essential elements in craniofacial bone formation leads to the failure of bone formation and fusion, resulting in a significant defect. Additionally, various genetic and environmental factors contribute to craniofacial development.[6]

Due to the rarity and variability in occurrence, coupled with the low incidence of these cases, the development of effective techniques has been hindered. Consequently, the selection of treatment approaches is highly individualized, presenting a complex and challenging task that heavily relies on the expertise and judgment of the surgeon.[4]

CASE ILLUSTRATION

A 45-year-old female patient visited our clinic reporting issues including ectropion, epiphora, and soft tissue deformities affecting her left eye. Her medical history revealed prior surgeries related to Tessier classifications 3 and 5 for a unilateral facial cleft, involving multiple attempts to reconstruct the lower lid through ear cartilage grafts for the tarsal plate. Upon physical examination, we observed an ectropion-like condition in her left eye, marked by a scar and residual graft tissue within the lower lid's middle third. Additionally, there was evident incompleteness in the tarsal plate, accompanied by flattened mid-cheeks, accentuating the tear-trough area and elongating the lid height, contributing to an older, fatigued appearance. The patient's diagnosis was established according to the Tessier classification of 1976.[3]

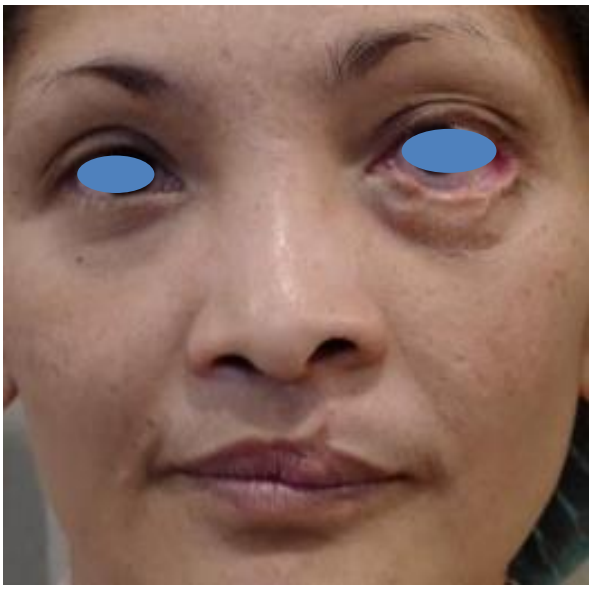

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Figure 1. Pre-surgery picture of a patient with uncorrected Tessier 3 and 5 facial cleft with a moderate degree of ectropion on the left. [7]

In this particular case, the surgical intervention was focused on addressing the ectropion by reinforcing the tarsal support through the application of a tarsal strip technique. Simultaneously, a midface lifting technique was employed to provide muscle support.

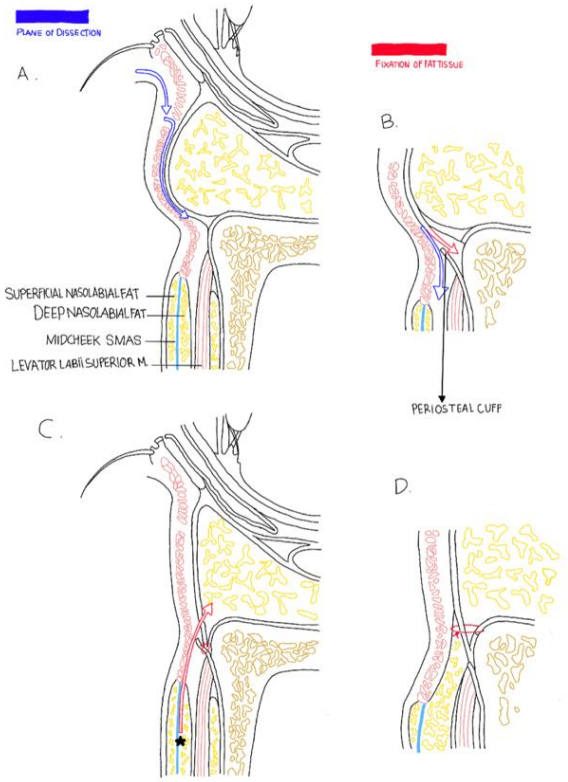


Figure 2. A, B, C, and D show a muscle flap dissection which was made onto the orbital rim then the orbital fat was transposed and placed under the periosteal cuff.[9]

For the midface lifting aspect, the author adopted a modified version of the fat preservation technique introduced by Mendelson in 2001[8], as outlined by Lee et al.[9] The surgical procedure was conducted under light sedation, complemented by a local anesthetic solution containing lidocaine and epinephrine in a 10:1 ratio. An infraciliary incision, executed with a number 15 blade in a bevelled fashion, was made to initiate the procedure. Initially, the skin and the orbicularis muscle flap were meticulously separated from the orbitalis septum along the preseptal space until the orbital rim was exposed. Subsequently, the premaxillary space was dissected, and a periosteal cuff was fashioned.

Upon completion of the dissection, the medial and central orbital fat were transposed and positioned beneath the periosteal cuff. This was followed by suturing them together using Polydioxanone (PDS II) 5-0. The next step involved separating the orbicularis muscle flap from the skin. The muscle flap was then pulled and advanced in a superolateral vector to elevate the midface area, providing the necessary muscle support. Finally, the flap was secured to the inner aspect of the lateral orbital rim with PDS II 5-0 sutures.

To ensure the proper length of the tarsal, we conducted a wedge resection to excise excess scar tissue and utilized a remaining cartilage graft to reconstruct the tarsus. The application of the tarsal strip technique, as derived from Anderson et al.[10] was employed to fortify the tarsal support. The technique involved a lateral canthotomy and cantolysis, achieved by making an incision on the outer corner of the eye and separating it from the surrounding tissue. Subsequently, the appropriate crus of the lateral canthal tendon was transected. Tarsal strips were then crafted from both the lower and upper lids. The eyelid was split into anterior and posterior lamellae, and the tarsal strip was fashioned from the posterior lamella. To prevent epithelial trapping, it was crucial to thoroughly clean the epithelium from the tarsal strip. Using scissors, an excess margin from the anterior lid and eyelash follicle was removed. The tarsal strip was then sutured to the periosteum at the lateral orbital wall, allowing for the adjustment of height and tension of the lateral cantus. Finally, the lower lid skin

was draped over the lower eyelid, excess skin was conservatively marked and excised, and the incision was closed with nylon 7-0 sutures.

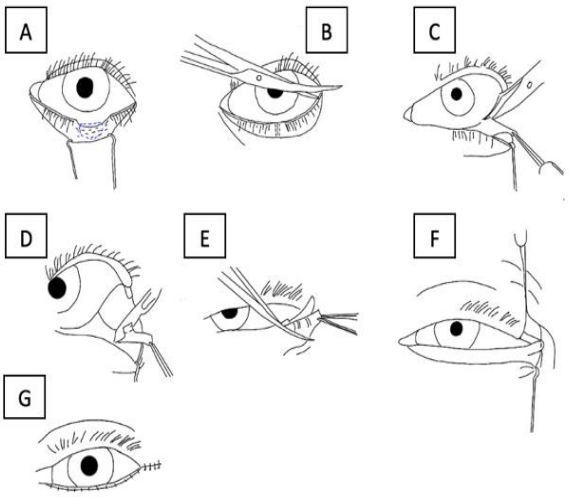
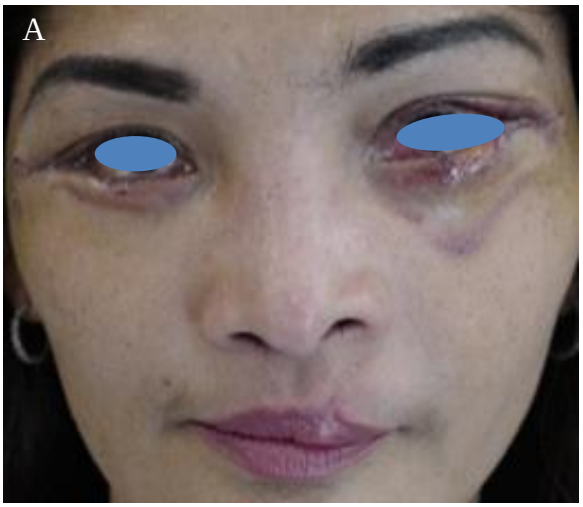


Figure 3. (A) Excised excess scar tissue with a wedge resection. (B) and (C) A lateral canthotomy and a lateral cantolysis were performed, then a lower lid was split into anterior and posterior lamellae, and the tarsal strip was fashioned from the posterior lamella. (D) and (E) Palpebra conjunctiva was shaved from the tarsal strip, then the Anterior lamella was excised. (F) and (G) Tarsal strips are sutured together to the periosteum at the lateral orbital wall.[7]

These approaches are intended to overcome the ectropion and prevent recurrency and give better facial aesthetic result. After seven days post-surgery (Figure 4A), the ectropion was successfully corrected with minimal swelling and bruising. All sutures were removed, and although there was a slight slant in the appearance of the eyes at this point, it was expected to return to normal over time, with no restrictions on normal activities. At the three-month mark following the surgery (Figure 4B), there were no indications of recurrent ectropion. The initial complaint about constant tearing had gradually improved to a normal condition, and overall aesthetic appearance was notably enhanced.



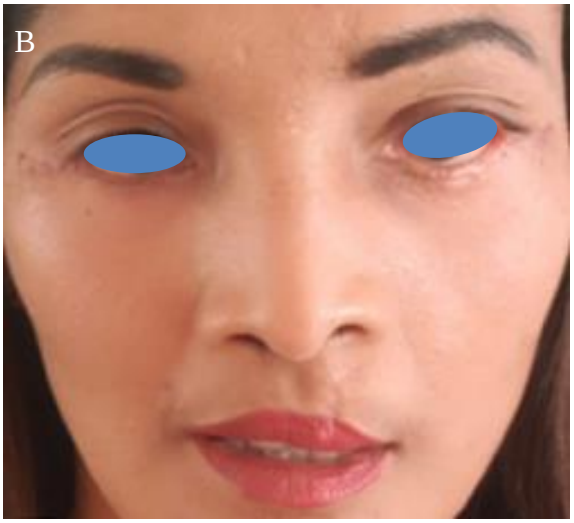


Figure 4. (A) 7 Days after surgery, (B) 3 months following surgery.

DISCUSSION

Craniofacial congenital clefts represent abnormalities affecting the cranium and face, characterized by defects along the anatomic lines of fusion. These malformations exert adverse effects on patients' lives, encompassing functional, psychosocial, and aesthetic aspects. Among orofacial anomalies, midline mandibular cleft stands out as one of the rarest conditions.[11-13]

The etiology of craniofacial clefts refers to the underlying causes or origins of these congenital anomalies, and as mentioned, it remains unclear. Craniofacial clefts are rare birth defects characterized by abnormal openings or gaps in the tissues of the head and face. The most common types involve the lip and/or the palate, and they can vary in severity. Several factors may contribute to the development of craniofacial clefts, but a precise understanding of the etiology is still elusive.

Genetic mutations passed down from parents to their children can play a role in craniofacial clefts. Some cases may be linked to specific genetic syndromes that affect facial development. Exposure to certain environmental factors during pregnancy, such as maternal smoking, alcohol consumption, nutritional deficiencies, or exposure to toxins, may increase the risk of craniofacial clefts. Teratogens are substances that can interfere with normal fetal development. Exposure to certain teratogens during pregnancy, such as drugs, alcohol, or infections, may increase the risk of craniofacial clefts. Interaction of Genetics and Environment are often considered that craniofacial clefts result from a complex interplay between genetic predisposition and environmental factors. Certain individuals may have a genetic susceptibility, and environmental influences during critical periods of development could contribute to the manifestation of clefts.Irregularities during the embryonic development of the face and skull can lead to cleft formation. Failures in the fusion of facial structures during early pregnancy can result in clefts.Problems with the blood supply to developing facial tissues may contribute to anomalies in craniofacial development.In some instances, the cause of craniofacial clefts remains unknown and is classified as idiopathic. These cases may involve a combination of genetic and environmental factors that are not yet fully understood.

Research in genetics, embryology, and molecular biology continues to shed light on the underlying causes of craniofacial clefts. Improved understanding of these factors may contribute to better preventive measures, early detection, and management strategies. Additionally, a multidisciplinary approach involving genetics counselors, surgeons, and other healthcare professionals is often necessary for comprehensive care in cases of craniofacial clefts.

In 1976, Tessier introduced an anatomical classification system for craniofacial clefts, categorizing them into 15 groups numbered from 0 to 14.[3,14,15] This classification encompasses clefts that either involve or do not involve the orbit, differentiating those in the middle parts of the face (clefts 0, 1, 2, and 14) and lateral parts of the face (cleft 7). The remaining nine clefts impact the orbit and periocular area, with clefts 3–6 described as southbound and clefts 8–13 as northbound. While an equivalent counterpart exists for almost every cleft at the top part of the orbit, each cleft is independently created.[15-19]

Tessier cleft number 5 remains difficult to classify. According to earlier publications, there's been documentation of a dual phenotypic manifestation related to cleft number 5.[20,21] The first form, considered the more severe presentation, involves maxillary medial dysplasia alongside a considerable soft tissue defect.[22] On the other hand, the second, milder form exhibits a vertical sclerodermic furrow along with a bony cleft located on the lateral aspect of the maxilla.[23,24] Tessier's initial publication noted this dual presentation in just one patient who had a bilateral facial cleft number 5.[3]

Racz, C. (2018)[25] identified the existence of two distinct variations within Tessier cleft number 5. Tessier's classification from 1976 delineated several

distinctive features that differentiate cleft number 5 from neighboring clefts (such as numbers 4 and 6). These distinctions encompass its position in relation to the infraorbital nerve (medial for cleft number 4 and lateral for cleft number 5), its location on the eyelid (medial third for cleft number 4 and lateral third for cleft number 5), its positioning on the alveolus (behind the lateral incisor for cleft number 4, behind the canine for cleft number 5, with no alveolar involvement for cleft number 6), its placement on the lip (lateral to the philtrum for cleft number 4, lateral third and on the oral commissure for cleft number 5), and its orbital rim involvement (present in cleft 5 but positioned more laterally on the zygomatic corpus for cleft number 6). Furthermore, an added complexity arises with the inclusion of cleft number 6 in Treacher Collins syndrome. Isolated cleft number 6 occurrences are infrequent and are typically classified as cleft number 5 according to Tessier's 1976 classification.[3,25]

Facial Tessier Cleft type 3, a form of craniofacial malformation, affects various structures including the vermilion border of the upper lip and the nasal wing, leading to the absence of an oral vestibule in this region. The cleft extends through the sidewall of the nose to the medial angle of the eye, medially in relation to the lower lacrimal point. This condition is accompanied by a fissure in the lower eyelid, downward dislocation of the medial angle of the eye, microphthalmia, hypertelorism, improperly formed nasopharyngeal tract, a cleft in the medial orbital wall, and a cleft in the hard palate and the maxillary alveolar process.[26]

The soft tissue characteristics of Tessier 3 cleft begin superiorly at the lacrimal portion of the lower eyelid, extend around the alar base within the naso-labial groove, and terminate inferiorly as a cleft lip. In Tessier 3 cleft, the tissue deficiency is situated along the medial cleft margin, leading to constriction of the nasal soft tissues. Conversely, there is tissue excess along the cheek on the lateral cleft margin. Tissue excess is present along the medial cleft margin, but the nose is correctly positioned. Excess tissue can be released from the skin next to the nose on the medial cleft margin. In the case of Tessier 3 cleft, the incision for the opening is placed within the supra-alar crease, and the triangle is designed using the skin excess along the lateral cleft margin. For Tessier 3 cleft, the initial incision is made within the supra-alar crease, and the triangular shape is crafted utilizing the surplus skin along the lateral margin of the cleft. [3,27]

The surgical treatment of these cases poses a significant challenge due to the rarity of these anomalies and the absence of established standard care guidelines.[28] Additionally, the traditional management approach entails intricate markings that can be challenging for surgeons to memorize, discouraging many from attempting the surgery. Surgeons also grapple with complexities such as determining the optimal age for surgical intervention and devising methods to minimize scarring in these cases.

Each uncommon craniofacial cleft is distinct and presents a unique set of challenges. The infrequency of the condition and the varied manifestations make the development of a universal diagnostic and treatment algorithm particularly difficult.[3,6] Several techniques have been suggested for managing craniofacial deformities.[29,30] The prevalent approach employed by surgeons to address Tessier craniofacial clefts is the use of local flaps.[4,31] However, the complexity of the malformations poses challenges in accurately marking flaps, and in some instances, this technique results in scarring that does not align with the natural folds of the anatomical unit.[32] The utilization of the tissue expansion technique emerges as a potentially more effective solution, offering tension-free reconstruction with tissue quality comparable to that of the

surrounding area.[33] Unfortunately, the availability of tissue expanders in Indonesia is constrained. As a result, we are proposing an alternative method that not only has the potential to yield superior results but also conceals scars within the natural contours of the face. This research focuses on employing a dual approach involving midface lifting and a tarsal strip technique as an innovative alternative for lower eyelid reconstruction. This unique combination of techniques represents a groundbreaking modality, never before utilized to address soft tissue issues in patients with facial clefts. Additionally, it offers the advantage of simplifying the intricate marking process for surgeons and enhancing overall surgical outcomes.

The main objective of the surgical procedure is to establish robust support for the lower eyelid by enhancing both muscle and tarsal support. This goal is achieved through the implementation of the midface lifting technique, which involves dissecting from the perceptual space to the premaxillary space, allowing the midface area to move when the muscle flap is pulled towards the superolateral vector, creating an anti-gravity effect. Consequently, this effect provides additional support to the orbicularis muscle.

In this particular case, the patient had undergone multiple surgeries, including the reconstruction of the tarsal plate using an ear cartilage graft, leading to the presence of scar tissue and remnants of the cartilage graft. To address this issue, a wedge excision technique was employed to remove the scar tissue, and the remaining graft was secured to the tarsal, effectively forming a unified plate. In the final step of the procedure, tension and strength were restored to the tarsal plate using a tarsal strip. This involved performing a lateral canthotomy and transecting the appropriate crus of the lateral canthal tendon. The height and tension of the lateral cantus were adjusted by suturing it to the periosteum at the lateral orbital wall.

The combination of these techniques complements each other, working synergistically to rectify ectropion by strengthening both the muscles and tarsal support. Although the long-term evaluation of ectropion recurrence remains unconfirmed, the results at the three-month follow-up were highly satisfactory. Where, the initial issue with constant tearing gradually improved to normal conditions, there are no signs of recurrent ectropion. Overall, there has been a significant improvement in aesthetic appearance.

The strength of this study lies in the innovative combination of the midface lifting and tarsal strip techniques as an alternative approach to reconstructing a lower eyelid in patients with a facial cleft. This study comprehensive overview of Tessier craniofacial clefts, encompassing various classifications and distinct features of different cleft types. It presents a detailed description of Tessier cleft number 5, highlighting the complexities and dual phenotypic presentations associated with this condition. Additionally, it delves into the specifics of Tessier cleft type 3, elucidating the tissue deficiencies and excesses along different cleft margins, offering insights into the surgical approach.

This novel combination has not been previously utilized for addressing soft tissue problems associated with facial clefts. The proposed technique offers potential advantages, particularly in simplifying the complex marking process and improving surgical outcomes. The midface lifting technique, involving dissection from the preseptal space to the premaxillary space, provides sturdy support to the lower eyelid by allowing the midface area to move anti-gravity when the muscle flap is pulled and advanced towards a superolateral vector. This not only addresses the primary goal of strengthening muscle and tarsal support but also offers an innovative anti-gravity effect, enhancing the overall effectiveness

of the procedure. The use of a tarsal strip further contributes to the restoration of tension and strength in the tarsal plate. The combination of techniques, including wedge excision to remove scar tissue and tie over the remnant cartilage graft to the tarsal, provides a comprehensive solution. The lateral canthotomy and adjustment of the lateral cantus height and tension contribute to the overall success of the procedure. the comprehensive exploration of rare craniofacial cleft types, providing a nuanced understanding of the unique characteristics and challenges associated with each. The discussion of Tessier cleft number 5 and the nuances of its distinct features, as well as the detailed soft tissue descriptions and surgical implications for Tessier cleft type 3, add depth to the understanding of these conditions. Furthermore, it acknowledges the absence of established standard care guidelines, emphasizing the complexity of managing these anomalies.

However, it is essential to acknowledge the limitations of the study. The long-term evaluation of ectropion recurrence remains uncertain, and the proposed technique's effectiveness may vary among different patients. Additionally, the study mentions the limited availability of tissue expanders in Indonesia, which prompted the exploration of alternative approaches. Continued research and long-term evaluations could further validate its effectiveness and broaden its applicability.

CONCLUSION

This combined technique shows promise in effectively reconstructing an ectropion eye, particularly within the context of this uncommon facial cleft condition, by enhancing both tarsal and muscular support. However, further evaluation is necessary to ascertain its effectiveness in preventing ectropion recurrence. The three-month follow-up detailed in this article demonstrates outstanding outcomes, leading to patient satisfaction.

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CONFLICT OF INTEREST

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AUTHOR CONTRIBUTION

The authors confirm contribution to the paper as follows: Study conception and Design: TA; Drafting of the article : HTH, AIG; Analysis and interpretation of results: HTH; Critical revision of the article for important intellectual content TA, HTH Final Approval: TA; Administrative, technical, or logistic support: AIG; Collection and Assembly of data: AIG. All authors reviewed the results and approved the final version of the manuscript.

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