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Rhomboid Flap versus Linear Closure for Odontogenic Oro-Cutaneous Fistula: A Case Series

[1]Aashish Deshmukh, Professor, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

[2]Khushal Desai, Assistant Professor, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

[3]Ayeshwarya Chaudhary, Assistant Professor, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

[4]Manasi Bavaskar, Resident, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

[5]Rhea Khanna, Resident, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

[6]Mehul Bhoye[6], Resident, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

Corresponding Author: Rhea Khanna, Resident, Department of Oral and Maxillofacial Surgery, Y.M.T. Dental College and Hospital, Kharghar, Navi Mumbai, Maharashtra, India.

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Abstract

Introduction: Oro-cutaneous fistula (OCF) is rare conditions, characterised by a pathologic communication between the oral cavity and the skin. The literature currently available, talks extensively about linear closure of the fistulas. However, no report till date has been presented on the use of a local flap for its closure. The aim of this novel case series was to evaluate and compare the results of a local rhomboid flap with the conventional linear technique for the closure of OCF.

Method: Two cases of oro-cutaneous fistulas were surgically treated – one with a local rhomboid flap and one with conventional linear closure.

Results: Parameters considered for the evaluation and comparison of the two treatment modalities were - time taken for the surgery, and post-operative scar. Both the techniques ensured their own merits and demerits; nonetheless, time taken for the surgery was more, and a conspicuous post-operative scar was observed with the rhomboid flap.

Conclusion: Linear closure is the treatment of choice for closure of oro-cutaneous fistula, when compared to the local rhomboid flap.

Keywords: Closure, Case-report, Local flap, Oro-cutaneous Fistula

Introduction

Odontogenic oro-cutaneous fistulas are manifestations of chronic dental infections, which provide a pathway for drainage of pus; and act as a communication between the oral cavity and the cutaneous surface of the face. [1,2] The extra oral opening usually arises as a sequel to bacterial invasion from the offending carious tooth; causing spread of inflammatory process into the peri-radicular space, and leading to bone resorption that subsequently dissects along the path of least resistance; thereby erupting through the skin as an odontogenic cutaneous fistula.[1]

Long standing odontogenic infections and dental implant complications are the most common cause of oro-cutaneous fistulas followed by maxillofacial trauma, salivary gland lesions, and head neck neoplasms. [3] 80% of the cases arise from mandibular teeth; and the most common site for these fistulas are the chin and the posterior mandible near its lower border. [4] Few OCF arise from infected maxillary teeth, and open at the lateral aspect of the nose and upper cheek. [4]

Patients with oro-cutaneous fistulas often report to physicians or dermatologists; as the presentation of fistula is far from the dental cause of origin; and therefore, often misdiagnosed as lesions of non-odontogenic aetiology. [5] Hence, the diagnosis of these lesions require a high degree of suspicion; else can lead to unnecessary procedures such as mycological studies, biopsy, treatment with antibiotics or radiation, and skin surgeries. [1,5] A permanent cure of such long standing OCF is obtained only with proper dental treatment; either endodontic therapy or extraction, followed by fistulectomy and aesthetically pleasing closure of the extra oral defect. [1,6]

Literature search on oro-cutaneous fistula invariably reveals about case reports and series aiming to understand the epidemiology, clinical presentation, and differential diagnosis of this lesion. Limited convincing results focussing on the use of different treatment options for OCF closure have been presented to date. Erol Cansiz et al. [2] employed a submandibular gland flap for closure of a resistant oro-cutaneous fistula below left side of mandible. Likewise, Balakrishnan et al. [7] used pedicle expanded deltopectoral flap to repair a large pharyngocutanoeous fistula in a patient with previous neck dissection and radiotherapy. However, no study till now has utilised a local flap; such as a transposition, a rotation, or an advancement flap for the closure of an odontogenic origin oro-cutaneous fistula.

Since there is a lacunae in the literature regarding the above mentioned subject; the aim of this report is to evaluate the use of a local transposition rhomboid flap for closure of oro-cutaneous fistula on face; and compare it to the conventional linear closure technique used hitherto.

Case Report 1

A 23-year-old otherwise healthy male patient reported to the Department of Oral and Maxillofacial Surgery, with the chief complaint of a scar on the right side of the face for the past 1-month. On taking the detailed history of the presenting illness, the patient reported that he first experienced pain in the right posterior mandibular region since 2-months; which was mild, dull aching in nature along with slight swelling. Hence, he consulted a general physician who prescribed him antibiotics and analgesics which brought symptomatic relief; but the pain and swelling re-appeared after 4 weeks. However, this time he also noticed extra oral pus discharge from the right side of the cheek; which later subsided, but a scar persisted.

On extra oral examination, a visible scar measuring approximately 1 cm x 1 cm was located at a position where an imaginary line from the right outer canthus of the eye met the lower border of mandible (Figure1). The scar appeared retracted and dimpled. On bimanual palpation, the region was firm in texture; and a cord-like extension was felt communicating from the extra oral skin to intraoral mucosa.

Figure 1: Right profile photograph.

Intra-oral examination revealed mild erythema associated with the mucosa surrounding the tooth 46, with no obliteration of vestibule. Palpation of adjacent vestibule caused no tenderness, but pain on percussion w.r.t. 46 was elicited. There was no presence of any sinus opening or pus discharge.

A panoramic radiograph was taken, which revealed a disto-proximal radiolucency extending up to the distal pulp horn and a small, solitary, well-defined radiolucency involving the mesial root of 46 (Figure 2). Correlating the history and clinico-radiological examination, a provisional diagnosis of Oro-cutaneous fistula w.r.t. 46 was proposed.

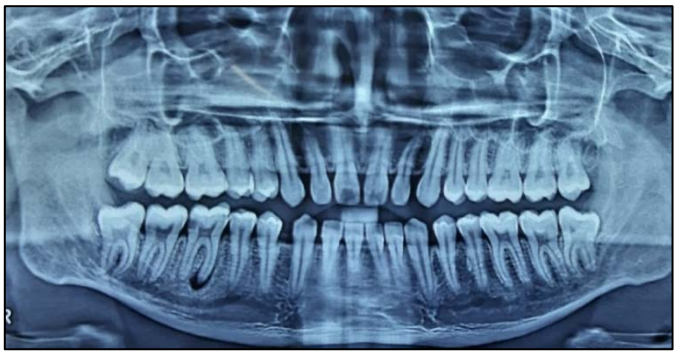

Figure 2: Orthopantomograph showing carious 46.

After due consent, the patient was taken into the clinical operatory. A rhomboid flap design was opted for this patient. Following all aseptic precautions, intraoral 2% lignocaine with 1:80,000 adrenaline local anaesthesia using right inferior alveolar and long buccal nerve block technique was administered; and extraction of 46 was carried out.

A marking of rhombus ‘ABCD’ with equal opposing sides; and two opposing acute and two obtuse interior angles were constructed around the defect (Figure 3). Further, the configuration was designed by extending a limb CE from the edge C; equivalent to the length of the diagonal AC. Additionally, a second limb EF was drawn upwards and of equal-length, parallel to BC.

Extra orally, local infiltration was given around the scar. An incision was made along the markings around the fistulous tract and subcutaneous dissection was carried out. The fistula was traced intraorally till the lateral border of the mandible and subsequently excised from the surrounding tissue (Figure 4).

Figure 3: Rhomboid transposition flap markings.

Figure 4: Fistulous tract excised.

Then the flap CEF was raised in a subcutaneous plane, adequate undermining was done to ensure passive transposition, and eventually was rotated over the defect (Figure 5). Tension free closure was achieved by placing 5-0 Ethilon sutures over the rhombic defect, and a primary closure was achieved at the area from where the flap was transposed (Figure 6).

Figure 5: Flap transposed over the defect.

Figure 6- Sutures placed.

Case Report 2

A 35-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of pus discharge on the right side of the cheek for 15 days. Further history taking unveiled that, in the last two months the patient had undergone extraction with 46 and simultaneously was undergoing root canal treatment of 47 as well. During the course of treatment, she experienced dull, diffuse pain along with minor swelling in the lower right cheek region; for which she took antibiotics and analgesics prescribed to her by a local physician. Following which the pus drainage and pain reduced, in addition, the swelling also decreased in size. However, there was a recurrence of draining pus and blood from the opening on the right cheek; along with an increase in swelling which alarmed the patient; hence she desired treatment for the same.

Extra oral inspection exhibited 1cm x 1 cm red, erythematous, and a dimpled lesion with granulation; present midway between the angle of mandible and corner of the mouth on the right side (Figure 7). On palpation, the encircling region around the lesion was tender and exuded pus along with blood from the opening. An attempt to introduce a gutta percha point from the extra oral opening to trace the path and cause of the lesion was done. Though

it could not be inserted entirely probably because of kink in the fistulous tract; consequently, posing a diagnostic dilemma.

Figure 7:Right profile photograph.

On intraoral examination, mild swelling surrounding the mucosa w.r.t. tooth 47 was seen, along with minor obliteration of the buccal vestibule in the same region (Figure 8). Restoration and mesio-proximal caries were also noted on 47. On palpation, pain on percussion was elicited w.r.t. 47, with absence of any sinus opening or pus discharge. A panoramic radiograph was obtained; which manifested a small, solitary, diffuse radiolucency involving the mesial root of 47 (Figure 9). Associating the clinical and radiological investigation, a provisional diagnosis of Oro-cutaneous fistula w.r.t. 47 was established.

Figure 8: Intraoral photograph showing obliteration of buccal vestibule w.r.t. 47.

Figure 9: Orthopantomograph showing peri-apical radiolucency w.r.t. 47.

The patient was brought into the clinical operatory after obtaining the duly signed informed consent. The conventional linear closure technique was chosen for this case. The patient was scrubbed and draped under all aseptic precautions; administration of right inferior alveolar and long buccal nerve block technique using 2% lignocaine with 1:80,000 adrenaline was done; and extraction of 47 was carried out.

Next, an elliptical design was created extra orally, encompassing the fistulous tract and the surrounding erythematous area (Figure 10). Local infiltration using ring block was given around the lesion, following which an incision was placed around the marking; and the external retracted fistulous opening was excised in a subcutaneous plane (Figure 11).

Figure 10: Elliptical marking drawn.

Figure 11: Fistulous tract seen after skin excision.

Following this, a complete dissection was carried out to separate the fistula tract from the extra-oral tissue till the intraoral mucosa (Figure 12); and the tract was excised in toto. Layer wise closure was achieved using 4-0 Vicryl subcutaneously and 5-0 Prolene over the skin (Figure 13).

Figure 12: Fistula tract being excised.

Figure 13: Linear closure achieved.

Result

The results were computed on the parameters of time taken for the surgery and the post-operative scar in the follow up period. Both the cases were performed by the same operating surgeon. The time taken was measured using a stop watch from the administration of anaesthesia extra orally till the placement of last suture. Intra oral procedure of extraction of offending tooth was not included for evaluation of this parameter. The time noted for the rhomboid flap was relatively higher than the linear closure technique (Table 1). This finding can easily be attributed to the fact that the intricate design construction and careful reflection of the rhomboid flap is largely experience dependant; and inadvertently consumes additional time compared to the conventional method.

Table 1: Time taken for surgery

Case	Time Taken
1- Local Rhomboid Flap	48 minutes
2- Linear Closure	32 minutes

The parameter of gauging the post-operative scar was carried out by the same investigator. Both the patients underwent alternate suture removal on the 7^{th} and 11^{th} post-operative days; with a regular follow, up to 3 months. (Figure 14, 15) During this course, a sunscreen with sun protecting factor of 50 was advised to be applied topically. At the third month follow up, the scar was evaluated using

the Manchester Scar Scale proposed by Beausang et al. [8] A proposed 5- point clinical assessment sheet, along with the grading on Visual Analog Scale from 0 to 10 decimal point (excellent to poor) was adopted [8] (Table 1).

Figure 14: Case 1- Three month follow up

Figure 15: Case 2- Three month follow up

Visual Analog Scale		
Excellent ←		→ Poor
Color	Perfect	1
	Slight mismatch	2
	Obvious mismatch	3
	Gross mismatch	4
Matte vs shiny	Matte	1
	Shiny	2
Contour	Flush with surrounding skin	1
	Slightly proud/Indented	2
	Hypertrophic	3
	Keloid	4
Distortion	None	1
	Mild	2
	Moderate	3
	Severe	4
Texture	Normal	1
	Just palpable	2
	Firm	3
	Hard	4

Figure 16: Manchester Scar Scale

Score on the Manchester Scar Scale ranged from 5 to 28, low scores representing clinically well healed scar of good cosmetic appearance; and high scores representing clinically poor scar. [8] Accordingly, the scores of both the patients were charted (Table 2). A total score of 18 and 12 was observed respectively for Case 1 of local rhomboid flap and Case 2 of linear closure; indicating a better, acceptable scar in favour of linear closure for OCF. This element of drawback for the rhomboid flap can be explained by the increased height of the scar owing to its geometry; subsequent enhancement of distortion, and thereby imparting a visually conspicuous outcome over the linear closure technique.

Table 2: Scores charted for post-operative scar

Criteria	Score	
	Case 1: Local Rhomboid Flap	Case 2: Linear Closure
Colour	2	2
Matte vs Shiny	1	1

Contour	3	2
Distortion	3	2
Texture	2	1
Visual Analog Scale	7	4
Total	18	12

Discussion

Cutaneous fistula of dental origin begins at the apex of an infected tooth and acts as a pathway to drain infected material through the skin.[9] The most frequent reason for the occurrence of OCF is the diagnostic delay, which results in a fundamental elimination of the lesion as an odontogenic fistula; there by leading to chronicity of the underlying infection.[10] These lesions present as soft, erythematous, and depressed lesion extra orally; and results in pus discharge on palpation.[11] The key objective in the treatment of an OCF is removal of the cutaneous fistulous scar, in conjunction to appropriate closure; as well as eradication of the source of infection.

A well-planned and executed local flap can lead to excellent cosmetic results with minimal distortion of the surrounding facial landmarks, preclude jeopardising of tissue vascularity and prevent chances of wound contracture. [12] Albeit these numerous advantages, the literature so far has no mention regarding the utilisation of local flap as a treatment modality for the closure of an odontogenic orocutaneous fistula on face. This case report therefore, focussed on evaluating a local transposition flap for the closure of oro-cutaneous fistula; and compare it with the conventional linear closure technique.

Among the local flaps obtainable in maxillofacial region; the rhomboid flap was introduced by Alexander A. Limberg in 1946; incorporating configuration of a classic rhombus with two opposing 60° and two opposing 120° interior angles, along with equal sides; for closure of a similarly shaped defect. [13] It is a geometrically designed, random-pattern, simple transposition flap; which follows the principles of flap design, retaining its vascularity and providing tissue composition similar to the excised region. [14] This flap in particular is exceptionally flexible in terms of altering the internal angles, so as to cover the defect more efficiently. [15] Additionally, in contrast to other advancement or rotation flaps; the rhomboid flap exclusively has a rare incidence of pin cushioning effect due to its straight lines and angles. [15]

However, various surgical factors such as excessive undermining, unsuitable tight closure, and manifestation of standing cutaneous deformities; colloquially termed as dog-ear can dictate the prognosis of rhomboid flap. [14] Furthermore, integration of the specifically engineered vectors can occasionally result in the failure of incorporating the multiple limbs of the rhomboid within relaxed skin tension lines. [12] Therefore, it is prudent to keep these factors in mind when using this flap.

Linear closure on the contrary, is the simplest advancement flap for approximating the incised wound; in which the adjacent tissue is advanced from both sides to close the defect primarily.[16] This has been extensively used as the treatment for OCF so far. Nonetheless, this method relies heavily on skin elasticity and its ability to stretch into the defect.[16] Crucial factors such as consideration of facial subunits, satisfactory advancing and adequate tension free closure along the linear path to minimise scarring have to be taken into deliberation when using this method of closure.[16]

With afore mentioned detailed insight of both the techniques for closure of an odontogenic oro-cutaneous fistula, and the experience gained from this case report; few valuable thoughts can be put forward. The conventional linear closure technique is widely accepted for the reason of being a relatively simple, faster and easy to learn modality. But, chances of scar contracture with

subsequent depression in the operated site can remain as a major concern when using the technique. On the contrary, local rhomboid flap can be considered an ideal method for closure of larger OCF; eliminating the possibility of contracture as it replaces tissue with tissue. But, it is sensible to highlight and dredge the few pitfalls of rhomboid flap; such as being an expertise oriented procedure, time consuming, and resultant outcome of a conspicuous scar due to multiple limbs of the geometry of its design.

Conclusion

Linear closure is the mainstay treatment modality for odontogenic oro-cutaneous fistula. Larger studies on considering local rhomboid flap for the closure of OCF needs to be undertaken to validate its use.

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