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Case Presentation

An Inclusion Epidermoid Cyst, a Rare Complication of Versatile Naso-Labial Flap

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Abstract

We are reporting a case of 47 years old gentleman who was diagnosed with left lower alveolus carcinoma and later on he presented with epidermal inclusion cyst post oncological resection with nasolabial flap reconstruction. Compared to microsurgical free flaps, nasolabial flaps are simple, easy to learn, one staged procedure and provides good cosmetic results. It can be used bilaterally for coverage of both, intra-oral and extra-oral defects. It is highly vascular flap with blood supply from the facial, transverse facial, angular, infraorbital, infratrochlear artery and the free anastomosis between the terminal branches overlying the nasolabial skin. The important thing in harvesting this flap is to avoid implantation of epidermal tissue like hair follicles, sebaceous glands and sweat glands into the deep tissues. Inadequate de-epitheliaslisation results in formation of inclusion cyst during transposition of flap through trans-buccal tunnel into the oral cavity. Formation of inclusion epidermoid cyst is a rarely encountered phenomenon in head and neck cancers.

Keywords: Naso-labial flap; Epidermoid cyst; Inclusion cyst; De-epithelialisation; Oro-facial defects; Reconstruction

Introduction

Lip and oral cavity cancers are complex cancers and after resection, reconstruction procedure is required for covering of the defect. Depending on the complex nature and size of the defect, regional flaps and microsurgical free flaps can be used to cover it. Microsurgical free flaps are complex, lengthy, costly and require expertise. Nasolabial Flaps (NLF) can be used for oro-facial defects as it is a small procedure, learning curve is small and achieves desired cosmesis. NLF was first described by Dupuytren and it was popularised by Diffenbach [1]. NLF is a versatile flap with rich vascular supply and it is tunnelled into oral cavity based on superiorly based pedicle or inferiorly based pedicle as per location of the defect. Thiersch used a superiorly based flap in 1868 and he tunnelled it through the cheek for closure of a palatal fistula [2]. Esser had described an inferiorly based flap [3]. Wallace had described the method of first one-stage, de-epithelialized nasolabial flap in 1966 for the closure of a palatal defect [4]. Rose in 1981 described a one-step arterialized island flap to avoid the bulk of the de-epithelialized pedicle in the tunnel and to provide more mobility [4]. NLF is a pliable, simple, versatile reliable alternative flap and it has been used as a subcutaneous pedicled "random flap" [5]. For the last 20 years, several case series has been reported where NLF was used to cover the defects upto 5cm in diameter of floor of mouth and ventral tongue with good functional outcome [6,7]. De-epithelialisation of the NLF is the foremost and important step. It is very important to avoid implanting epidermal tissue like hair follicles, sebaceous glands and sweat glands into the deep tissues. If it is inadequate de-epithelialisation, it may result in the formation of an epidermal inclusion cyst as the covering epithelium is 'implanted' into the deeper mesenchymal tissues [6]. Inclusion cysts are very rarely encountered entity in head and neck region. We are reporting a case of 47 years old gentleman who was diagnosed with left lower alveolus carcinoma and post oncological resection with NLF reconstruction presented with epidermal inclusion cyst.

Case Presentation

A 47 year old gentleman with no co-morbidity presented to our clinic with ulcero-proliferative lesion over left lower alveolus. Patient had history of tobacco chewing for the last 27 years and he used to consume 5-6 times per day. Family history, past history, surgical and medical history were not significant. Systemic examination was unremarkable. Local examination was suggestive of ulero-proliferative lesion over left lower alveolus in relation to 2nd molar teeth measuring 2.5 x 2.5 cm. No other positive findings were noted in the oral cavity and no neck lymph nodes were palpable. We had advised him to undergo biopsy and it was suggestive of squamous cell carcinoma. For staging purpose, we had advised him to do CECT (Contrast Enhanced Computed Tomography) of oral cavity, neck and thorax. CECT was suggestive of same lesion with size 2.3 x 2.2 cm with sub-centimetre lymph nodes at level IA and IB and both lungs were free from disease. Case was discussed in our multidisciplinary institutional tumor board and patient had been advised upfront surgery. He underwent wide local excision of the left lower alveolus lesion with marginal mandibulectomy with left naso-labial flap reconstruction with left selective neck dissection (Level I to IV). Naso-labial flap was marked and harvested from left cheek about 1 -1.5cm lateral to medial canthus based on inferiorly oriented pedicle (Figure 1 and 2). Flap was transposed into the oral cavity through trans-buccal tunnel near left oral commissure (Figure 3). Postoperative course was uneventful. Oral feeding was started from 3rd postoperative day and he was discharged on 5th postoperative day. All sutures were removed on 12th postoperative day (Figure 4). Final histopathology report demonstrated a well differentiated keratinizing squamous cell carcinoma with size 2.1 x 1.2 cm with absent lymphovascular

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Figure 1: Marking of inferiorly based NLF.

Figure 2: Nasolabial flap harvesting.

emboli and perineural invasion with all mucosal , soft tissue and bony cut margins were free from tumor without lymph node metastasis (0/40 lymph nodes). AJCC 8th edition staging was p T2 p N 0 p M0. Case was re-discussed in our multidisciplinary board and plan was to keep him under observation as per our institutional follow up protocol. After one month of removal of facial sutures, he revisited our clinic with complain of discharge from the facial wound where naso-labial flap was tunnelled into oral cavity. Clinical examination was indicative of multiple nodular swellings over left buccal mucosa with cystic swelling over the left cheek (Figure 5). We did punch biopsy from the nodular area of buccal mucosa which were looking suspicious recurrent lesion. Cyst was located over the left cheek at the junction of insertion of flap into oral cavity (Figure 6). There was no intra-oral communication. After squeezing the cyst, serosanguinous fluid drained. Biopsy from the nodule demonstrated a hypertrophic squamous epithelium without atypia or malignancy. We had managed it with conservative treatment for next 3 weeks with weekly visits at our clinic with the support of antibiotics and analgesics. It was responded to conservative treatment however, cyst was persistent. After 3 weeks, we had excised the superficial cystic

Figure 3: Nasolabial fold closure with point of flap transposition into oral cavity (marked by yellow arrow).

Figure 4: During suture removal, 1st visit.

Figure 5: Multiple nodules over left buccal mucosa (Marked by red arrow).

swelling along with surrounding skin. Wound edges were refreshed and resutured. Secondary sutures were removed after 14 days and wound healed completely with no residual cyst portion. Patient is in periodic follow up with us as per our institutional follow up protocol and after one year of completion of treatment, he is disease free.

Discussion

Reconstruction of defects, post oncological resection is the toughest job for the treating surgeon. Over the decades, reconstructive techniques has changed and in the current era, microsurgical free flaps becomes an alternative aesthetic option with commendable outcome. Nasolabial Flap (NLF) is a simple with short learning curve, less technique sensitive with minimum number of complications and provides a good cosmetic results. NLF is based on superiorly based

Figure 6: Epidermoid Inclusion Cyst (Marked by yellow arrow).

pedicle, inferiorly based pedicle and random blood supply. Defects of the palate, upper alveolus, cheek, side or dorsum of nose, ala, columella, lower eyelid and upper lip are closed by superiorly based flap as this avoids twisting of the pedicle [4]. Defects of the floor of the mouth, lower alveolus, lower lip and buccal mucosa are closed by inferiorly based flap [5]. The color of the flap matches with the other facial tissues hence, desired cosmesis can be achieved. NLF is marked and harvested lateral to the nasolabial fold with medial limit of the flap 2-3mm lateral to this fold [8]. In the superiorly based NLF, the base of the flap is near the ala and the apex is in line with the oral commissure [9]. When there is requirement of extra length (10-12 cm) in case of superiorly based flap, the incision is extended to the skin over the mandibular border. It is called extended NLF, a variant of superiorly based flap [10]. In the inferiorly based NLF the apex of the flap is 5-7 mm lateral to the medial canthus [11].In the index case, an inferiorly based flap was harvested and passed through the trans-buccal tunnel into the oral cavity near the left commissure, NLF is supported by vascular supply from the facial, transverse facial, angular, infraorbital, infratrochlear artery and the free anastomosis between the terminal branches overlying the nasolabial skin [12]. Depending on the defect size, the thickness of the flap is determined. The thickness of the flap can be moulded into as small as subdermal plexus or as thick as facial musculature with maintaining its nerve supply intact. It is transposed into the oral cavity through the trans-buccal canal which is located near the commissure of the mouth for inferiorly based flap or near ala in nasolabial fold for superiorly based flap. In the index case, full thickness flap was harvested and transposed into oral cavity for covering of left lower alveolus defect. After a period of 2 years, NLFs are replaced by mucosa when used for intraoral defects. The postoperative scars are hidden in the nasolabial folds which is more acceptable in elderly population [7]. The advantage of NLF is that it can be used bilaterally for covering of both, intraoral and extraoral defects and it's role in correcting oral submucus fibrosis is well known [13].

De-epithelialisation is an important factor in achieving successful NLF. Meticulous de-epithelialisation of the pedicle is very important because it helps in avoiding implantation of epidermal tissue including hair follicles, sebaceous glands and sweat glands into the deep tissues. 1 to 1.5cm de-epithelialization of the skin flap near the base is necessary to prevent iatrogenic epidermoid inclusion cyst when the flap is passed through the trans-buccal tunnel into the oral cavity [14]. It is very rare phenomenon in head and neck cancer surgeries post oncological resection and reconstruction. In the index case, de-epithelialisation was not adequate hence, it may be the reason for development of inclusion epidermoid cyst. In the second setting, we had excised the cyst completely with surrounding normal tissue. Wound edges were refreshed with adequate de-epithelialisation. During follow up, till one year of completion treatment, there was no recurrent disease or recurrent cyst.

Conclusion

Nasolabial flap is a highly vascular, easy to learn, versatile and safe flap. It can be used for immediate one stage reconstruction of intra-oral and extra-oral, small to medium sized defects with adequate de-epithelialisation with minimum postoperative complications.

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