Polymorphous Low-Grade Adenocarcinoma—Management and Reconstruction with Temporalis Myofacial Flap

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Abstract Polymorphous low-grade adenocarcinoma is a rare, malignant salivary gland tumor, which is found almost exclusively in minor salivary glands. It is more frequent in the third–seventh decade of life, with a clear female predilection in a 2:1 ratio. It is usually located in the hard or soft palate, although it may be found in the rest of the oral cavity too. It is rare in major salivary glands. In general it has good prognosis, with recurrence rates in the range of 17–24%. Although rare, metastasis to regional lymph nodes has been reported.

We report a case of 38 year old female patient who reported with recurrent low grade polymorphous adenocarcinoma in the hard palate, who has been treated with wide excision of the lesion, with reconstruction of the defect using temporalis myofacial flap.

Introduction

Minor salivary gland malignant neoplasm accounts for 2–4% of head and neck malignant neoplasms, 10% of all oral cavity malignant neoplasm and 15–23% of all salivary gland malignant neoplasms [1]. In 1984 Evan HL and Batsakis JG identified a different clinicopathologic entity from adenocarcinoma and designated it as ‘polymorphous low grade adenocarcinoma (PLGA) [2, 3]. Earlier it was named as pleomorphic adenoma, unspecific carcinoma, lobular carcinoma [4] or even sometimes as adenoid cystic carcinoma [3]. It is a malignant neoplasm of salivary

glands, more frequently detected in minor salivary glands [5]. It is a rare pathology that affects the people in the third to seventh decade of life, with a female predilection in a 2:1 ratio [6].

Case Report

A 38 year old female reported to the outpatient clinic of Department of Oral and Maxillofacial Surgery, Mar Baselios Dental College Kothamangalam, Kerala having complaint of recurrent swelling in the palate. She first noticed the swelling at the left side of hard palate 20 years back which slowly increased in size. Patient had no difficulty in deglutition and speech. She underwent an excisional biopsy with extraction of upper left second molar 10 years back via a Weber Ferguson approach at a local hospital. Histopathologic examination report was low grade polymorphous adenocarcinoma of minor salivary gland.

Five year back patient started to notice the swelling again which increased in size gradually.

Clinical examination revealed a swelling of 2 × 2 cm in the region of retromolar area of left side of maxilla extending from the distal aspect of 26 up to the maxillary tuberosity and medially in the hard palate (Fig. 1). The swelling was firm in consistency, non tender and not fixed to the underlying periosteum. Occlusal radiograph showed radiolucent lesion 2.5 cm in diameter (Fig. 2). Axial and Coronal CT did not show any bony erosion.

Incisional biopsy was done. Histopathologic examination revealed low grade polymorphous adenocarcinoma (Fig. 3).

A wide excision along with the posterior part of the maxilla (partial maxillectomy) keeping 1 cm margins and

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Fig. 1 Swelling in the hard palate

Fig. 2 Maxillary occlusal view

Fig. 3 Histological examination revealed polymorphous low grade adenoma

reconstruction of the defect with temporalis myofascial flap was planned considering the high recurrence rate of the lesion.

Patient came after a month for the planned surgical procedure.

Management

Treatment modality—partial maxillectomy with reconstruction using temporalis myofascial flap.

Intraoral incision is placed from buccal sulcus distal to 25, extended towards the midline, encircling the lesion keeping safe margin of 1 cm. Osteotomy was done with chisel and mallet, upper first molar was also extracted. Preauricular incision with temporal extension was placed just anterior to the tragus and extended superiorly towards vertex and ends above the superior temporal line. The incision is carried down to the deep temporal fascia, thus placing it posterior to the superficial temporal artery and its branches. Blunt dissection is done to elevate the scalp involving skin, subcutaneous cellular tissue, galea, temporo-parietal fascia and innominate fascia. Temporalis muscle is identified and detached from the temporal bone and sectioned into two parts with a vertical incision (Fig. 4). Anterior portion is rotated and brought to the defect in the oral cavity, by tunneling through below the zygomatic arch. Flap is sutured to palatal and buccal mucosa (Fig. 5). At the donor site posterior segment of the temporalis muscle is brought anteriorly and sutured to the temporalis fascia to reduce the temporal hollowing. Incision is closed with 3-0 vicryl and 4-0 nylon.

Excised specimen was sent for histopathologic examination and it revealed polymorphous low grade adenocarcinoma and specimen margins were clear for malignancy. Patient is kept under regular follow up. Good take of the flap was seen. Patient did not have any problems with speech and deglutition following surgery. Temporal hollowing was minimal. Normal mucosalisation of the flap was also seen (Fig. 6).

Fig. 4 Temporalis flap sectioned

Fig. 5 Flap sutured to defect

Fig. 6 Three months post op- mucosalisation of flap

Discussion

Polymorphous low grade adenocarcinoma is a malignant epithelial tumor of salivary glands [7], found almost exclusively in minor salivary glands [3], and is rare in extra oral locations, including major salivary glands. It is the second most common tumor of minor salivary glands [3]. 60% of the cases occur on the hard or soft palate, followed by 13% of the cases occurring in the buccal mucosa, 10% in the upper lip, 6% in the retromolar area, and 9% in the rest of the oral cavity [6]. The lesion is normally described as a painless, slow growing mass, covered by non-ulcerated mucosa. In some cases it may be adhered to deep planes and it can reach sizes between 1 and 4 cm [6, 8]. Diagnosis is late and can take weeks or even years because of its slow growth. This lesion can erode or infiltrate bony tissue [8]. Histology shows a non-encapsulated lesion with infiltrative margins. It is named as polymorphous due to its different growth patterns: tubular, solid, papillary, microcystic,

cribriform, fascicular, and cords. PLGA morphologic diversity and cytologic uniformity may cause a diagnostic dilemma especially with adeno cystic carcinoma and pleomorphic adenoma [9]. It can infiltrate bony tissue and even present perivascular and perineural invasion. The best treatment is surgical excision including the subjacent bone, if necessary [6, 8]. This surgery is frequently followed by radiotherapy. The prognosis is good and recurrence rate ranges between 17 and 24% [5, 10]. Metastasis is rare [7] and unusual (9%) but in case it occurs, it mainly affects regional lymph nodes [9]. PLGA require a long follow up period for good prognosis results [11].

Polymorphous low grade adenocarcinoma is a rare lesion. In a research study carried out by González Lagunas [1], a sample of 59 malignant salivary gland tumors were assessed, and no PLGA was found. This lesion is located almost exclusively in minor salivary glands within the oral cavity, mostly hard palate, its extra oral presentation extremely rare. Nasal fossa and nasopharynx locations have been described in less than 0.5–1% of the cases [12]. There is a clear female predilection, particularly affecting those in their 40 s and 50 s.

The maxillectomy defect creates a significant rehabilitative issue, as it creates speech, deglutition, cosmetic, and possible orbital problems. Micro vascular surgical techniques have revolutionized surgical reconstruction but have not eliminated the need for prosthetic rehabilitation. Surgical reconstruction should meet the objectives outlined by Hoopes and Edgerton: 1. consistently obtain a healing wound, 2. restore palatal competence and function, 3. support the orbit or fill the orbital cavity in exenteration, 4. obliterate the maxillectomy defect, and 5. re-store facial contour [13].

According to Corderio maxillary defects can be classified into 4 types, type 1 is termed as limited maxillectomy [14]. Limited term is used when one wall of maxillary antrum is removed [15], temporalis myofascial flap is the first line of reconstruction option for limited resection of maxilla [16].

Temporalis myofascial flap for reconstruction of ablative defects within maxillofacial region with the discretion by Lentz using it in reconstruction of defect after resection of ankylosed temporomandibular joints [17].

Temporalis myofascial flap is used by Golovine in reconstruction of orbital defect in 1898 [18].

Reconstruction of the palatal defect after partially maxillectomy with temporalis flap have the advantages of ease of elevation, reliable blood supply, proximity to the maxillofacial region, camouflage of the incision in the hair line. Fascia in contact with oral cavity re-epithelialise in 3 weeks [19]. Disadvantages may be sensory disturbance, facial nerve injury, and temporal hollowing [20]. Post surgical radiotherapy can lead to fibrosis of flap. Tumor

resection involving internal maxillary artery, temporalis myofascial flap is contraindicated [21].

The temporal hollowing is not considered a major problem because it can be camouflaged by the hair line especially in non bald men and women. Falconer and Phillips [22] have suggested reconstruction of the defect at the donor site primarily by use of allograft like acrylic or silastic. If, only the anterior part of the temporalis muscle is used, the posterior part can be repositioned into the anterior region to fill the dent, since the hollowing is more pronounced in the anterior region.

We have used a similar technique and found that the temporal hollowing in the anterior region was minimal. We could use this technique as our defect was small. After three months of follow up, patient complained of traumatic bite on the surgical site which was managed by selective grinding of the lower molars lingual cusps. Good take of the flap was noticed, with normal mucosalisation (Fig. 6), temporal hollowing was minimal.

As a treatment modality for low grade polymorphous adenocarcinoma surgical excision is the main stay treatment. Use of radiotherapy is still not clear as metastases are rare and reoccurrence can be successfully salvaged in 50% of the cases [7]. Long follow up period is required due to its recurrence rate [11]. Our patient was on regular follow up at monthly intervals for the first year and advised long term follow up. Reconstruction of defect with temporalis myofascial flap in the maxillofacial region is very reliable due to its blood supply and ease of harvest [20].

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