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Polymorphous low grade adenocarcinoma of the buccal mucosa

PLGA is a rare tumor of the salivary gland that is limited, to a great extent, to the minor salivary glands. However, the most common subsite being the hard or soft palate. This study presents clinical report of PLGA located very rarely on the buccal mucosa, with brief overview on the importance of adequate surgical therapy, PH analysis and cooperation with a pathologist.

KEY WORDS: Buccal Mucosa, Multidisciplinary Management PLGA

Introduction

Polymorphous low-grade adenocarcinoma (PLGA) is a rare tumor of the salivary glands that is limited, to a great extent, to the minor salivary glands. Based on population studies report an annual incidence is 0,051 cases per 100000 individuals [1].
PLGA most commonly arising in the region of the junction of the hard and soft palates, followed by the lip[1]. Female to male preponderance is 2:1, with mean age of presentation in the sixth decade of life. The typical presentation is that of an indolent or slowly growing lump in the oral cavity, which may occasionally be painful or even ulcerated. Ulceration is usually second to biopsy or pressure from a denture or other trauma. The patients who presented with pain, ulceration, or bleeding were not considered to have a more aggressive disease [2].

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Tumor size is usually between 2 and 2,5 cm^3. These small specimens can be challenging and in some instances the pathologist may be unable to offer a definitive diagnosis. In these circumstances, close clinicopathological correlation in the context of a multidisciplinary team meeting is essential for treatment planning. Removal of the tumor with judicious sampling and analysis of the surgical specimen will help to refine the diagnosis and guide further management[3]. Definitive treatment for PLGA is surgical resection with or without radiotherapy [4].
This study presents clinical report of PLGA located very rarely on the buccal mucosa, with a brief overview on the importance of adequate surgical therapy, PH analysis and cooperation with a pathologist.

Clinical Report

At the beginning of February 2021, female patient age 73 visited our Clinic for evaluation of tumor of the right buccal mucosa. Before she visited us she was treated by general dentist with various conservative treatments.
Anamnesis and clinical examination indicate a painless, slow-growing tumor on the right buccal mucosa that has been present for few years. Further examination showed exophytic lump clearly demarcated from the environment, 2x2 cm in diameter, with central ulceration (Fig. 1, left). The patient's medical history showed that she do not suffer from serious general disease.
Clinical examination and ultrasound (US) of the neck

Fig. 1: PLGA on the buccal mucosa (left), The deeper structure of the tumor (middle), The same patient postoperative (right).

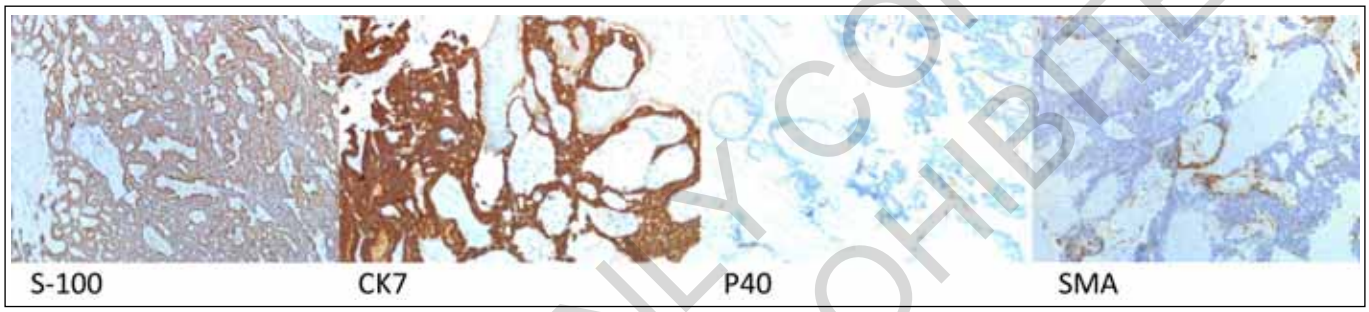

Fig. 2: IHH analysis S-100, CK7, P40, SMA.

were conducted as part of the examination process. They did not indicate pathologically enlarged regional lymph nodes.

The preoperative US of the neck showed that thyroid gland as well as parotid and submandibular glands had symmetrical lobes without focal changes. A lymph node with a diameter around 12mm was found in the jugulodigastric region on the right side of the neck.

A wedge biopsy was performed under local anesthesia LA. A fragment of sample taken from the center of the tumor to the part of the healthy mucosa was sent to University Hospital Banja Luka for patohystological (PH) analysis. After a few days, the biopsy result confirmed a pleomorphic adenoma PA of the small salivary glands.

Taking all this and the clinical examination results into consideration, a surgical procedure in LA was planned. Articain-chlorhydrate with epinephrine 1:100,000 was used in the course of the surgery. First step was to carefully and effectively apply LA. Afterwards, the tumor was radically removed, with the excision line 1 cm from the edges of the tumor. Special attention was made during the extirpation of deeper tumor structures of it, in order to radically remove the tumor and preserve the buccal branch of the facial nerve. An interesting observation was that the deeper structures were encapsulated and did not show an exophytic form like the one on the mucosal surface (Fig. 1, middle).

The resulting defect was reconstructed with local transposition flap where arch of rotation was 90 degrees. During the surgical procedure, care was taken not to lead the parotid salivary gland duct. The edges of the wound were sutured with 4.0 PDS suture. One hour after the surgery, the patient was released from the clinic with a prescription to take Xiclav 1gr. 2x1, per os and painkillers as needed. The postoperative recovery was uneventful (Fig. 1, right). The excised sample was sent for PH analysis. This time the definitive PH finding of the material indicates that it is a PLGA with clear surgical margins.

The most striking feature of the tumor is the diversity of its architecture. Microscopically, tumor cells form, solid, tubular, cribriform, and foci of papillary structures. The stroma in the tumor is partly myxohyaline, partly dense and sclerotic. Rare atypical mitosis were found. Transversely striated muscle were tumor free. There was no evidence of distant metastasis. Immunohistochemical IHH analysis showed S-100 protein +, SMA + (positive in myoepithelial cells) and p 40 - (Fig. 2).

During the analysis, the diagnosis was validated by a fellow pathologists from bigger analysis centers. However, the final diagnosis certainly would not change the previously set surgical concept. The tumor was radically removed, and it was decided to monitor it in the future. The patient came for a check-up every 3 months. Local

postoperative control and clinical examination of the neck were conducted each time. Six months after the operation, an ultrasound of the neck were also conducted showing no signs of enlarged lymph nodes. So far, a follow up over a period of two years did not show any signs of the loco regional recurrence.

Discussion

PLGA are slowly growing malignant neoplasms that primarily occur predominantly in the minor salivary glands of the oral cavity. However, the most common subsite being the hard or soft palate [5,6].

Primary these tumours rarely spread [7]. Despite this low rate of neck metastases, a diagnosis of PLGA requires further imaging to assess the status of the regional nodes. PLGA of the buccal mucosa is rare malignancy and its pathological diagnosis is also quite challenging. The differential diagnosis with adenoid cystic carcinoma ACC, PA, and other salivary gland tumors is important because it does have an overall excellent prognosis. Although they share many clinical and pathological similarities PLGA have a better prognosis than ACC [8].

An excellent coordination between surgeons and pathologists is vital to the previously described procedure. A multidisciplinary approach provides a satisfactory surgical approach and follow-up of the patient. This clinical report highlights the above. In this case, the first step was wedge biopsy. An insufficient amount of tissue was the cause of the misdiagnosis as a PA of the minor salivary gland. The question arises whether in such and similar cases when the tumor shows an ulcerated exophytic form on the surface, a radical surgical approach from the start is the option in order to create conditions for the pathologist to obtain an accurate diagnosis.

After radical surgical removal of the tumor, the pathologist makes the final diagnosis microscopically as well as by using IHH. Microscopically both PA and PLGA show the diversity of tumor architecture. However, the key diagnosis is made by IHH analysis.

Where PLGA is strongly positive for s100 protein and CK. Typically, PLGA is diffusely and strongly positive for S100 and CK7 [9]. In addition p63 positive and negative p40.

Recently, several studies have reported that PLGA usually has a p63 positive/p40 negative immunoprofile [10,11]. Distant metastases and death from disease are exceptionally rare [11]. Taking that into consideration, the importance of accurate diagnosis and radical surgery in such cases is reflected in excluding radiation as a therapeutic method. Side effects such as mucositis, salivary gland dysfunction, radiation caries, osteoradionecrosis, trismus can be excluded if the multidisciplinary management is used as the best alternative to minimize or even prevent such reactions [12].

Buccal region and its structure compared to the hard palate as the most common location of PLGA gives the surgeon more creativity to primarily close the defect with one of the reconstructive methods. In this case defect was reconstructed with local transposition flap where arch of rotation was 90 degrees providing a fast treatment and healing process, and same-day discharge from the Clinic. However, considering PLGA follow up by the surgeon so far was conducted within a short period of time, but it is promising since the surgery was used as a main medical method.

Lastly, PLGA of the buccal mucosa is a rare entity. For the form presented in this case, exophytic and ulcerated on the surface excisional biopsy as a radical surgical maneuver is as method of choice.

It gave pathologist enough tissue sample for accurate and fast diagnosis which is the fundamental basis for further therapy planning and patient follow-up.

Riassunto

L'adenocarcinoma polimorfo di basso grado è un tumore raro della ghiandola salivare che interessa per lo più ghiandole salivari minori. Tuttavia la sua sede più comune è il palato duro o molle. Questo studio presenta un referto clinico di PLGA localizzato molto raramente sulla mucosa buccale, con una breve panoramica sull'importanza di un'adeguata terapia chirurgica, analisi del PH e collaborazione con un patologo.

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