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CLINICAL ARTICLE

A RARE CLINICAL CASE FROM THE PRACTICE OF A MAXILLOFACIAL SURGEON: INFRAORBITAL HAMARTOMA

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Abstract

Hamartomas are very rare in the practice of a maxillofacial surgeon and dentist. It is necessary to know the distinctive features of hamartosal lesions in order to understand how to plan the necessary treatment for a patient and determine the tactics of patient management.

A small number of cases may reflect the true rarity of the lesion or may be due to its undetected nature and / or underreporting. On the one hand, this will allow for a competent verification of the diagnosis. On the other hand, it will lead to the fact that the fact of malignant proliferation will not be missed, which significantly changes the tactics of a maxillofacial surgeon or dentist.

A 60-year-old woman came to the maxillofacial surgery department of the Yerevan Medical Center, complaining of facial asymmetry, swelling of the right half of the upper jaw, the presence of a tumor, and a feeling of heaviness. For the purpose of final diagnosis and treatment, an operation was performed: removal of the neoplasm, extensive biopsy. Pathohistological diagnosis: hamartoma of the infraorbital region. The postoperative period was calm and without complications.

Based on the results of the operation, it can be concluded that the surgical method used is low-traumatic for such a diagnosis, provides a sufficient surgical field of vision, increases the possibility of radical tumor removal, and reduces trauma around important anatomical structures.

Key-words: *hamartomas; infraorbital lesions; dentistry; maxillofacial surgery*

Introduction

Hamartomas commonly occur in the lungs, liver, spleen, pancreas, and kidneys. Hamartomas are rare in the head and neck area, and even more rare in the oral cavity. Most often, hamartomas are benign lesions.[1] However, there are described episodes when malignancy occurs and hamartoblastomas develop from them, so the maxillofacial surgeon and dentist should be oncological alert in this case.

Some hamartomas can also lead to infection, infarction, fracture, haemorrhage, or rarely malignant transformation, this risk does not exceed 1%.[2]

Hamartomas of the maxillofacial region are divided into non-odontogenic and odontogenic. Clinically, most of them are asymptomatic and rarely cause any complications, except in cases of localization at the base of the tongue.

Based on the medical literature, malformations may be present at birth but appear later. Usually it has self-limiting growth, consistent with the growth of surrounding tissues. There can be both single and multiple lesions. Also, spontaneously regress may occur. These tumors usually are not encapsulated with undefined fields. The peculiarity of a hamartoma is that it consists of the same tissue elements as the localization organ, but is characterized by an abnormal structure. There is a connection with chromosomal abnormalities and syndromes.[3] Regarding the non-tumor nature of hamartomas, surgical removal is the treatment of choice.[4]

Hamartomas usually showing minimal growth, except as part of normal growth of the body or under hormonal influence. They may potentially cause cosmetic issues depending on their location.[5]

Case Report

A 60 years old female patient was admitted to the maxillofacial surgery department of "Yerevan" Medical Center. She was complaining of facial asymmetry, swelling of the right half of the upper jaw, the presence of a tumor and a feeling of heaviness. The patient noted, the presence of the tumor, the feeling of the heaviness during 10 years. During the last 1-2 years the swelling has increased in size, most noticeable during the last 6 months.

During extraoral examination, the patient's face was asymmetric due to the swelling of the right infraorbital region. The overlaying skin is of normal color, gathers into a fold, the surface is smooth, with solid-elastic consistency (Figure 1a). The lymph nodes were not enlarged. Intraorally there is tumor growth at 12-17 teeth, covered with normal mucosa (Figure 2).

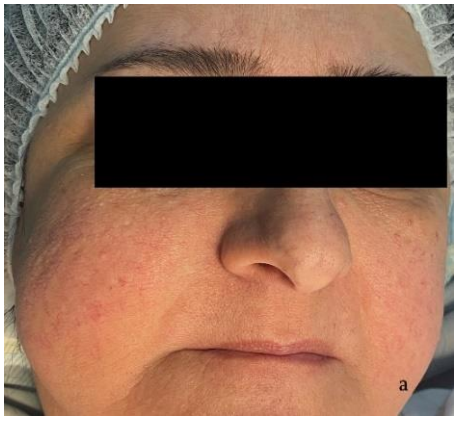

Figure 1. *A patient with infraorbital hamartoma, external view of the patient*

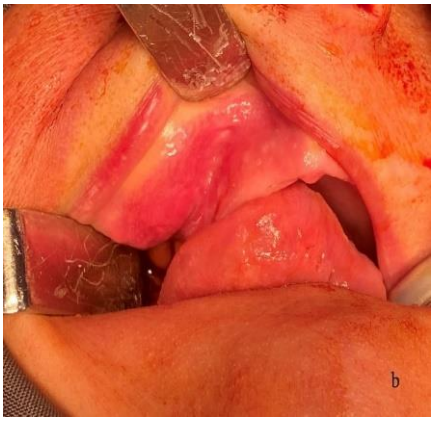

Figure 2. *Intraoral view of the patient*

According to ultrasound examination of the face and neck there was a round encapsulated hypoechogenic tumor in the right infraorbital region, the size of which is 4-4.5-5cm. Also, there was growth of the tumor into the right maxillary sinus.

Surgical operation

Surgery was performed under general anesthesia. An intraoral incision was made in the right half of the vestibule, the mucosa and submucosa were cut with the projection of the 12th-17th teeth (Figure. 3). The neoplasm was detected, it was separated from the adjacent tissues in a blunt and sharp manner, avoiding to the border of healthy tissues by about 2 mm (Figure 3). The tumor was removed in its entirety (Figure 4).

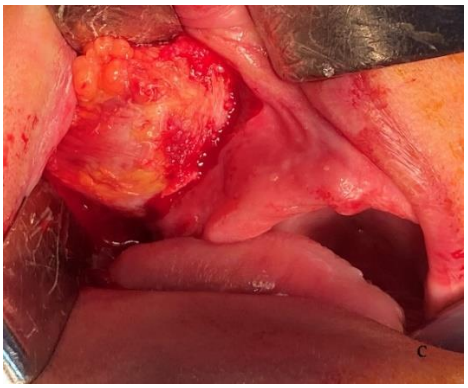

Figure 3. *Intraoperative view of the exposed tumor*

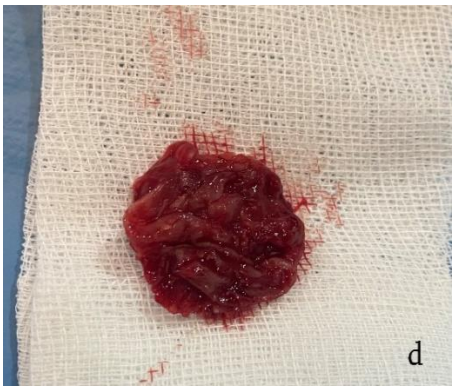

Figure 4. *Removed tumor*

A resorption of the anterior wall of the right maxillary sinus with a projection of about 2-2.5 cm was detected (Figure 5). A revision of the sinus was performed. Maxillary sinus defect was closed with buccal fat pad (Figure 6). For its mobilization small incision was done over the periosteum at 17 teeth. Then the buccal fad pad was mobilized with blunt dissection. After it was transposed over the defect zone and fixed resorbable sutures (Figure 7). The buccal fat pad was closed by local mucoperiosteal flaps (Figure 7).

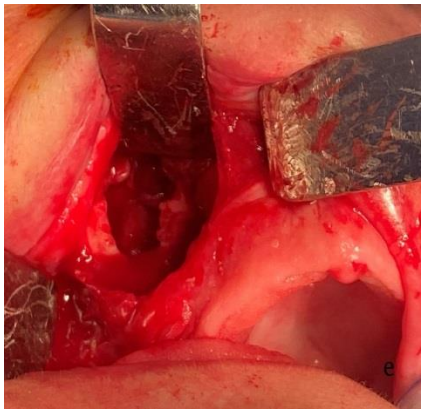

Figure 5. *Maxillary sinus floor defect after tumor removal*

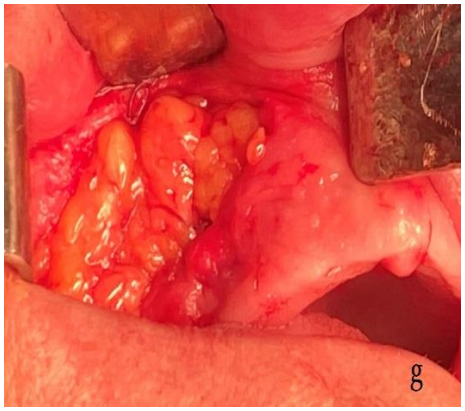

Figure 6. *Maxillary sins defect is closed with buccal fat pad*

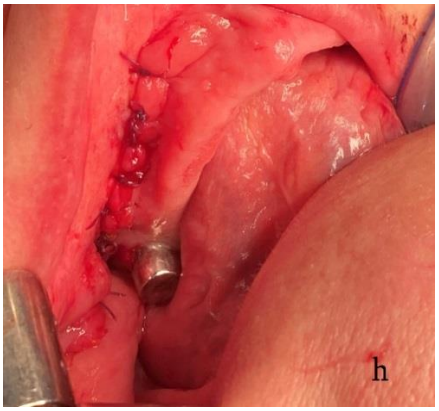

Figure 7. *View of the intraoral wound*

Sutures were removed on 10th day. Postoperative follow-up period was 2 months. No any complications were found during that period.

Morphological Study

The materials for morphological studies were the samples of lesion tissues. According to the standard histological scheme, the pieces of the tissue were fixed in 10% neutral formalin, dehydrated and embedded in paraffin. A series of sections 4 μm in thickness were stained with hematoxylin–eosin for a general assessment of the condition of the examined tissues. Histological micropreparations were studied with a ZEISS Primo Star trinocular microscope (ZEISS Microscopy, Jena, Germany) under 100, 400- and 1000-times magnification. Microphotographs were taken with a ZEISS Axiocam ERc 5 s (Carl ZEISS Microscopy, Jena, Germany).

Results

Grossly, it was no distinctly demarcated mass, infiltrative with varying amounts of tan-yellow adipose tissue and gray-tan fibrous tissue, with sizes 3x2,5 cm, with soft-elastic consistency.

It was diagnosed with a combination of laboratory testing, imaging (e.g, ultrasound) and biopsy. In

microscopical examination it was diagnosed a dysontogenetic hamartomatous lesion with a bronchogenic cyst, fibrolipomatous and skeletal muscle tissue with multiple blood vessels, e.g. it is composed of aberrant disorganized growth of mature tissues (Figure 8A, B, C, D).

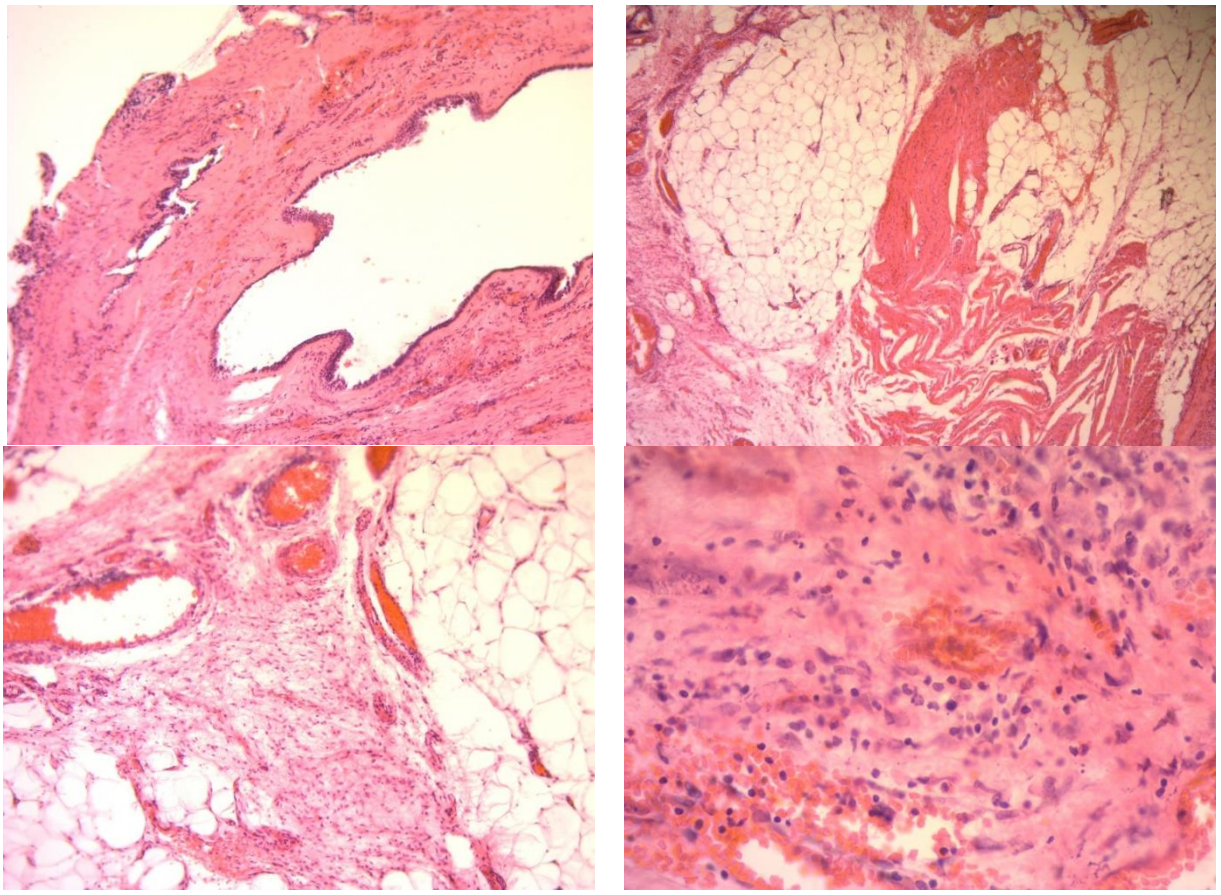

Figure 8. *Dysontogenetic hamartomatous lesion.* ***A.*** *Bronchogenic cyst.* ***B.*** *Fibrolipomatous and skeletal muscle tissues.* ***C.*** *Numerous blood vessels.* ***D.*** *Chronic inflammatory reaction (cellular infiltrate). Stained with hematoxylin-eosin, A, B, C x 100, D x 400*

The fibrous areas usually show haphazardly arranged intersecting fascicles of collagen fibers. The sceletal muscle bundles are haphazardly arranged and often form a horizontal band.

An increased number of well-formed but disorganized capillaries, veins, arteries/arterioles, and/or lymphatics was seen. The cells cytologically resemble the local tissues.

Differential diagnoses of a hamartoma included different types of benign or malignant tumours. Some of these may cause diagnostic difficulty and potentially be mistaken for malignant neoplasms.

It had not got a capsule and a clear boundary with surrounding tissues, which can cause difficulties for surgeons during the primary operation in determining the borders of tumor removal. We consider it advisable to use express biopsy during surgery for this purpose.

The general condition is satisfactory, the symmetry of the face has been restored, the patient has no complains. After 1 month, an CT examination was performed, according to which no recurrence was observed after 1 and 3 months.

Conclusion

Hamartomas are rare in the practice of the maxillofacial surgeon and dentist. Studying the specific features of hamartomatous lesions is essential to understand how to plan the necessary treatment and determine the patient management strategy for a patient with such pathology.

The small number of cases may reflect the true

rarity of the lesion or may be because of underrecognition and/or underreporting, which can lead to improper patient management. The information about hamartomas, on the one hand, will allow to correctly verify the diagnosis. On the other hand, it will lead to that the fact of malignant proliferation will not be missed, which significantly changes the tactics of the maxillofacial surgeon or dentist.

Declarations

Conflicts of interest and financial disclosures

The author declares that he has no conflict percent and there was no external source of funding for the research in question.

Ethical approval

The study was approved by the University ethics committee and was conducted in accordance with the Declaration of the World Medical Association.

Informed consent

Informed consent was obtained from all individual participants included in the study.

Source of funding

The work was not funded.

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