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Treatment of non-specific inflammation mimicking oral sarcoma: A rare case report

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Abstract---Sarcoma occurring in oral and maxillofacial is a rare case. Sarcoma of the oral cavity is rare, accounting for around 1% of all malignant oral tumors. These tumors usually proliferate and are aggressive. The report aimed to show the treatment of non-specific inflammation mimicking oral sarcoma. We herein report a patient with a history of a lump in the maxilla. Initially, the size of a pea, painless and gum-colored, and does not bleed easily. It grew up rapidly for four months. The patient planned to be performed existential biopsy by the collaborative Pediatric Surgery and Oral and Maxillofacial Surgery for obturator fitting. The pathology anatomy results from the biopsy show that the lesion was surprisingly a non-specific inflammatory process. After two months, there was bleeding at the maxilla after removing sutures at the obturator, and there was granulation tissue under the obturator. After that, the patient planned to receive excision of granulation tissue and osteotomy with general anesthesia. Because the defect was so vast, reconstruction with a vestibular flap for excellent defect closure was needed. After three weeks from the treatment that had been given to the patient, the control showed a satisfactory result.

Keywords---health outcomes, inflammation, sarcoma, mimicking, pediatric.

Introduction

Sarcoma of the oral cavity is rare, accounting for around 1% of all malignant oral tumors. Sarcomas are a diverse group of cancers that typically develop from mesenchyme-derived tissue. Only around 12% of all juvenile head and neck malignancies are soft tissue sarcomas, with a tiny minority occurring in the oral maxillofacial region. These tumors usually proliferate and are aggressive (Argyris et al., 2012; Thompson et al., 2016).

Sarcomas can arise from a variety of tissues, including bone, cartilage, muscle, fibrous, vascular, fatty, and neural tissue, so it has many types, including fibrosarcoma and osteosarcoma. Clinically, fibrosarcomas usually appear as slow-growing lumps that might grow to be quite large before causing pain. There are several differential diagnoses of soft tissue masses of the oral and maxillofacial, one of which is odontogenic inflammation (Shrivastava et al., 2016; Yamaguchi et al., 2004).

The treatment of choice for fibrosarcoma is wide resection with a clear margin. Inoperable situations or as a palliative treatment, radiation therapy and chemotherapy may be employed. The prognosis of the tumor is determined by its histological grade, size, and surgical therapy with disease-free margins. We were herein presenting a case of Non-Specific Inflammation Mimicking Oral Sarcoma (Swain et al., 2013; Wadhwan et al., 2010).

Case Description

A 5-years-old male child with a lump history in the maxilla from the patient's parents was known to have appeared four months ago. Initially, the size of a pea, painless, and the color of the gums. It does not bleed easily. Because the mass proliferated and bled easily since about one month ago, the patient was referred to the Universitas Airlangga Hospital. There are not blood clotting disorders, allergies, and asthma.

From the general examination, the general condition was good. Vital signs were stable, and general status was within normal limits. From the examination of the maxillofacial region, the extra-oral examination (Figure 1) did not reveal edema, hyperemia, and tenderness. From intra-oral (Figure 2), it was found that the mass on the anterior palate extends to the maxillary gingiva of teeth 52, 51, 61 to the extent of the mucolabial fold, clear boundaries with a size of $\pm 2.5 \times 2 \times 1$ cm, palpable solid hard on the palate and chewy in the maxilla, looks bummed, hyperemic with areas of fibrosis, and no active bleeding. The orthopantomography (OPG) revealed a radiolucent border surrounding the 11 seed with radiopaque calcification of the maxillary bone below the crown 11. A chest radiograph was advised to rule out the metastasis and for surgery with general anesthesia preparation, apparent without any significant and relevant findings (Figure 3).



Figure 1. Extra-oral examination did not reveal edema, hyperemia and tenderness



Figure 2. Intra-oral examination. Labial view (a), Oocclusal view (b), Palatal view (c)

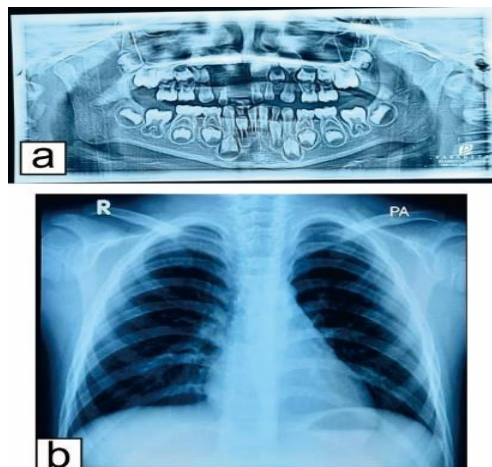


Figure 3. The orthopantomograph (OPG) revealed a radiolucent border surrounding the 11 seed with radiopaque calcification of the maxillary bone below the crown 11 (a), Chest radiograph was found to be clear without any significant and relevant finding (b)

Case Management

The treatment plan, in this case, was an excisional biopsy by a pediatric surgeon and installation of a surgical obturator by an oral and maxillofacial surgeon. Excisional biopsy and surgical obturator placement were performed under general anesthesia. Before surgery under general anesthesia, a blood test was performed, and the results were expected. During the surgery, a solid mass measuring 2.5 x 2 x 1 mm was found on the anterior palate to the labialis oris vestibule in the region of teeth 53-63. When the mass was excised, it appeared that the mass had destroyed the hard palate. So an osteotomy was performed to prevent the residual mass from forming. During the surgery, no cystic lesion was seen. It was eliminating the possibility that this mass was an adenomatoid odontogenic cyst like the orthopantomography showed. After the excision, a surgical obturator was installed to protect the defect from debris and prevent infection (Figure 4).



Figure 4. Postoperative defects

Stereolithographic is the process of turning a liquid polymer media into a solid resin, which includes both the 3D model and the support pillars, using a laser. These models have a 0.1 mm accuracy, and the duration of construction is established by the structure's complexities being printed.

The results of anatomical pathology examination of the mass showed surprising results, pieces of tissue lined with squamous epithelium, which is partially ulcerated underneath. It showed fibro collagenous stroma with necrotic areas, few myxoid areas, calcifications, vascular proliferation, and bleeding areas with inflammatory cell infiltration of lymphocytes, histiocytes, and neutrophils. There were no signs of malignancy, concluding that it was only a non-specific inflammatory process so that no further action was planned. At week 4, the sutures on the obturator were removed, and granulation tissue with hyperemia was still visible in the postoperative wound, so the obturator was still used to avoid infection. At week 6, the patient had bleeding in the maxilla when removing the obturator before. The bleeding did not stop however the gauze was pressed. So the patient was brought to the ER and given treatment until the bleeding stopped (Figure 5).

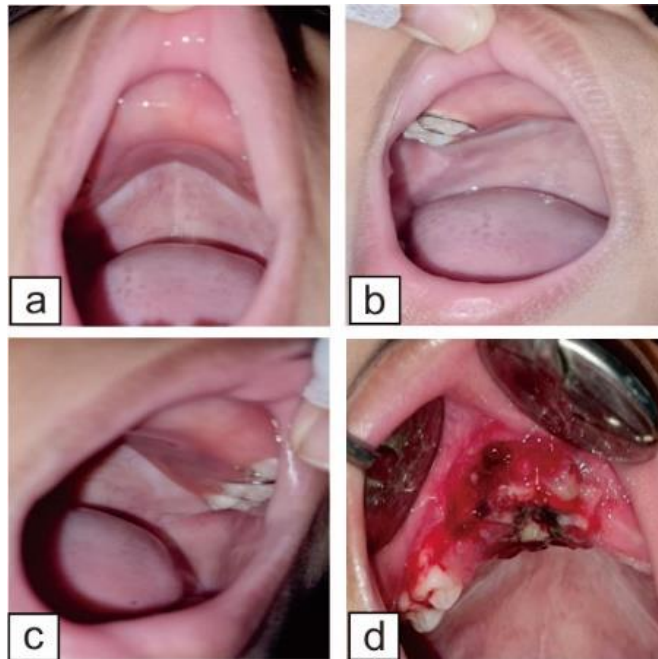
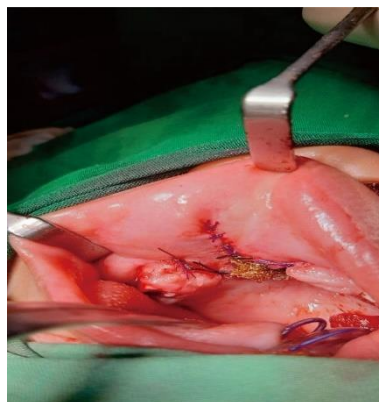


Figure 5. 6th week postoperative intra-oral with obturator (a,b,c), 6th week postoperative intra-oral without obturator (d)

The general condition was good, and vital signs were stable; general status was within normal limits from the general examination. From the examination of the maxillofacial region, the extra-oral examination did not reveal edema, hyperemia, and tenderness. From intra-oral, it was found that the obturator was attached well to the maxilla, the palatal mucosa showed granulation tissue with hyperemia, with no active bleeding. The patient then planned to granulation tissue excision, osteotomy, and reconstruction with vestibular flap. A blood test was performed before surgery under general anesthesia, and the hemoglobin was low (7 g/dL), maybe because of the bleeding before. After that, hemoglobin improved by blood transfusion until it reaches 10 g/dL.

During the surgery, the surgical wound in the region of tooth 53 had active bleeding and a wound consisting of fragile granulation tissue. Then excision of the granulation tissue and osteotomy was performed to create an excellent new tissue on the edge of the bone that has been done osteotomy, coated with bone wax to stop bleeding after osteotomy. When approximating wound closure, it was found that the surrounding mucosal tissue was not sufficient to cover the postoperative wound adequately, so the incision was widened with the vestibular flap technique. Then the surgical was applied, and the wound was sutured with 3.0 silk thread, then an obturator was applied again to hold the surgical at the wound.



(Fig. 6)

After three weeks of control (Figure 7), the postoperative wound healed satisfactorily. After suture removal, intra-oral examination showed no granulation tissue, and the gingiva closed well. There appears to be a slight palate depression may be due to the absence of a bone graft. The patient's permanent anterior teeth also started to erupt, and there was slight hyperemia of the gingiva around the erupting teeth.

Discussion

Fibrosarcoma is a malignant tumor that arises from fibroblasts. This type of sarcoma is mostly found around bones or in soft tissue. This determination has been regularly connected to luxury cells, spindle cell that produces collagen tumors count harmful fibrous histiocytoma, malignant peripheral nerve sheath tumors, and have other sarcoma and pseudosarcomatous injuries (Shrivastava et al., 2016; Thompson et al., 2016).

Of all the fibrosarcomas occurring in people, as it were 0.05% happen within the location of the head and neck. Of this, nearly 23% is seen within verbal depression. Clinically, within the verbal depression, the primary indications are torment, swelling, and in some cases extricating of the teeth and paresthesia. Secondary ulceration may be seen as the injury enlarges. Radiographically, an osteolytic injury is more often than not show, with ill-defined borders, but few authors also reported the lesion's good circumscription or cyst-like appearance (Shrivastava et al., 2016; Swain et al., 2013).

In the case of odontogenic infection, pulpal and periapical injuries have more or less the same course in primary and permanent teeth. Unaltered teeth may be affected by disease of the overlying primary teeth due to physiological situations. Pulpitis in primary teeth if cleaned without treatment can progress to periapical injury. The development of these lesions does not depend on the destruction of microorganisms, the susceptibility status of the person, local but also late predisposing components, and the most important vital factor - the duration of infection (Gupta et al., 2016).

The treatment of choice for bone fibrosarcoma is wide resection with clear margins without prophylactic lymph node dissection. In fibrosarcomas that cannot be extracted from the area, postoperative radiotherapy of 6000-7000 cGy is appropriate. In grade III fibrosarcoma, postoperative adjunct chemotherapy is prescribed to treat potential subclinical or small metastases. Maxillary submucosal vestibulopathies are used to increase surgical site exposure, reduce vertical midline scars, and deepen the anterior vestibular sulcus (Swain et al., 2013; Wessberg et al., 1980).

Histopathologically, bone fibrosarcoma has spindle-shaped cells that are relatively uniform and arranged in fascicles, often forming a herringbone pattern. In contrast to this case, the histopathological findings were pieces of tissue lined with squamous epithelium, some of which was ulcerated underneath. It shows a fibrocollagenous stroma with necrotic areas, few myxoid areas, calcifications, vascular proliferation, and areas of hemorrhage. with inflammatory cell infiltration of lymphocytes, histiocytes, and neutrophils. No signs of violence. It can be concluded that it is just a non-specific inflammatory process. This proves that the clinical state of cases resembling malignancy can only be in the form of an inflammatory process. Therefore, the biopsy results should be examined histopathologically to make an adequate treatment plan (Shrivastava et al., 2016; Thompson et al., 2016).

Figure 1. This is a figure example

Conclusion

Sarcoma of the oral cavity is a rare tumor that usually proliferates and is aggressive. The biopsy's pathological anatomy of the patient revealed that the lesion was a non-specific inflammatory disease, which was unexpected. After releasing the sutures at the obturator, there was bleeding at the maxilla and granulation tissue beneath the obturator after two months. Excisional biopsy and surgical obturator placement were performed under general anesthesia. After three weeks from the treatment that had been given to the patient, the control showed a satisfactory result.

Acknowledgments

There is no acknowledgment.

Conflict of Interest

There is no conflict of interest to declare.

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