# Journal of Oral Diagnosis ABSTRACTS OF VII MEETING OF DIGITAL ORAL PATHOLOGY 8-9 november 2017 - Belém. Pará. Brazil

#### Case 1

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# Osteosarcoma of the alveolar ridge: case report

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Osteosarcomas of the jaw are rare and represent only 2-10% of all osteosarcomas. We herein present a rare case of an osteosarcoma exhibiting histological characteristics of fibro-osseous lesions in the lower alveolar ridge. A 58-year-old female patient presented with the complaint of gradual swelling of the right alveolar ridge of the mandible over 3 months. Radiography revealed an illdefined radiolucency, without a periosteal reaction. Microscopical analysis revealed areas of fibro-osseous lesion containing a highly cellular fibroblastic stroma with spherical, concentric lamellated ossicles and bone. In addition, the lesion exhibited typical characteristics of chondroblastic osteosarcoma with osteoid and cartilage intimately associated with anaplastic tumor cells. The diagnosis was primary osteosarcoma. The patient was submitted to surgery with wide margins and follow-up of 1 year revealed no signs of recurrence.

### Case 2

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# Mammary analog secretory carcinoma of the buccal mucosa: case report

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Mammary analog secretory carcinoma (MASC) is a carcinoma of the salivary glands characterized by morphologic and immunohistochemical features that strongly resembles a secretory carcinoma of the breast, occurring more commonly in parotid gland of male patients. A 61-year-old female patient was referred because she noticed a mass of approximately 3 cm on the right buccal mucosa. Microscopically, cells with vesicular round-to-oval nuclei with finely granular chromatin and distinctive centrally located nucleoli organized in microcystic/solid and tubular structures with abundant eosinophilic homogeneous secretory material were observed. The secretory material stained positive for periodic acid-Schiff (PAS). Tumor cells were positive for S-100 protein, mamaglobin and gross cystic disease fluid protein 15 (GCDFP-15). The final diagnosis was MASC. Local excision with wide margins was made and post-operative follow-up of 2 years revealed no signs of recurrence.

#### Case 3

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# Ameloblastic carcinoma: case report

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Malignant odontogenic tumors are rare and represent approximately 1% of all oral malignancies. Ameloblastic carcinoma is a rare odontogenic tumor, which is aggressive in nature with extensive local bone destruction that has retained the features of ameloblastic differentiation and also exhibits cytological features of malignancy. It occurs primarily in the mandible in a wide range of age groups. It may arise de-novo or in pre-existing ameloblastoma or odontogenic cyst. A 36-year-old male patient was referred complaining of an asymptomatic growth on the left side of posterior mandible since 1,5 years. Clinical examination disclosed a mild swelling on the left mandibular region, whereas the intraoral exam did not reveal any significant expansion of the vestibular aspect of the posterior region of the mandible. Panoramic radiograph showed a welldefined unilocular radiolucent image in the posterior area of the body and ascending ramus on the left side of the mandible. An incisional biopsy revealed odontogenic epithelium arranged in the form of islands with peripherally arranged columnar cells with central stellate reticulum like cells. Loss of polarization, hyperchromatic nuclei, and increased mitotic activity was seen. The final diagnosis was ameloblastic carcinoma. Local excision with 2 centimeters of wide margins was made and post-operative follow-up of 2 years revealed no signs of recurrence.

#### Case 4

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# Anaplastic large cell lymphoma with oral involvement: case report

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Anaplastic large cell lymphoma (ALCL) is characterized by proliferation of anaplastic large lymphoid cells with abundant cytoplasm that strongly express CD30 antigen, being uncommon in the oral cavity. An 18-year-old male patient was referred for evaluation of a painful swelling in the face and ulcer in the oral cavity lasting 30 days, which was interfering in the mastication. The patient was a nonsmoker and his medical history did not reveal other relevant information. The extraoral examination showed a swelling in the left submandibular region and absence of cervical lymphadenopathy. Intraoral examination revealed an ulcerated mass showing reddish and yellowish areas, located at the floor of the mouth. Image exams revealed hypodenses areas in the submandibular region compatible with necrosis. Microscopically, it was identified a uniform proliferation of medium to large-sized atypical cells with abundant amphophilic to eosinophilic cytoplasm and eccentric nuclei. Mitotic figures and reactive macrophages were also observed. By immunohistochemistry, tumor cells were positive for CD45, EMA, CD30 and ALK. Ki-67 index was 90%. The tumor cells were negative for cytokeratin AE1/AE3, CK20, desmin, MyoD1, CD34, CD56, TdT, myeloperoxidase, CD15, CD20, CD3, CD10, CD1a, CD68, CD79a, CD138, Kappa, Lambda and EBV. The final diagnosis was primary ALK-positive anaplastic large cell lymphoma. The patient was submitted to surgery and chemotherapy and no evidence of disease was detected after 7 months of follow-up.

### Case 5

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# Prostate adenocarcinoma with metastasis to the posterior mandible: case report

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Metastatic tumors to the oral cavity are uncommon, accounting for 1% of all neoplasms, and the most common primary sites are the breast and lung. Oral metastasis usually involve the bone, being the mandible the most common site, while one third are located in the soft tissue, mostly in the gingiva and tongue. A 69 year-old male was referred for evaluation of a swelling lasting 4 weeks. The patient's main complaint was pain in the left mandibular region and paresthesia. His medical history revealed that he is under treatment for prostate adenocarcinoma. Intraoral examination revealed a mass covered by a smooth surface, located in the left mandibular gingiva. Radiography exam showed a diffuse radiolucency in the left mandibular body with osteolysis and sunray appearance. A biopsy was performed and histopathologically, the lesion consisted of invasive malignant epithelial cells nests in a ductal arrangement. The tumor cells were negative for cytokeratin 7 and 20 and positive for PSA, and the final diagnosis was prostate metastatic adenocarcinoma. The patient was submitted to surgery and chemotherapy. Although rare, metastatic tumors should be included in the differential diagnosis of an intraoral malignant neoplasm, above all if there is a primary known tumor and the histological features are similar, because in some patients this could be the first indication of the presence of a primary tumor.

# Case 6

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# Vascularized tumor in the parotid gland

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Solitary Fibrous Tumor (SFT) is benign mesenchymal tumor associated with the fusion gene (NAB2-STAT6). This tumor present fibroblastic phenotype and branching vasculature. SFT was first described in the pleura in 1931, and more recently has been reported in several extrapleural sites, including the head and neck, which represent an uncommon region for this tumor. Among head and neck SFT's, the oral cavity and major salivary glands are the third and fourth more frequent sites respectively, after sinonasal tract and orbit. SFT of major salivary glands affects mainly the parotid gland (about 80%). Herein we report a case of 45 year-old male with a slow-growing

swelling in the parotid region. At surgery, the tumor was well-delimited of the surrounding glandular tissue. It was treated by partial parotidectomy. Microscopically the tumor was well-defined and partially encapsulated, composed of spindle-shaped cells and vascular spaces. The spindle cells were arranged in storiform and haphazard pattern, presenting hypercellular zones intermingled with hypocellular, myxoid, collagenized and focally hyalinizing areas. Vessels varied in diameter some with perivascular hyalinization and the characteristic staghorn pattern described in SFT. Focal hyperchromatism and occasional mitosis were present in the hypercellular areas, however, atypia or necrosis were absent. The diagnosis was SFT which was confirmed immunohistochemically by the positivity of CD34 and BCL-2. Ki-67 index ranged from 9,5% to <1% in the hypercellular and hypocellular areas respectively, however, this was not considered as a sign of malignancy, since commonly STF presents hypercellular areas with these features. The patient is free from recurrence after two years of follow-up.

### Case 7

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# Plasmacytoma/multiple myeloma with amyloidosis

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Multiple myeloma is a clonal malignancy of terminally differentiated B lymphocytes characterized by the expansion of clonal plasma cells in the bone marrow. These findings often result from direct injury or accumulation of immunoglobulins in various organs. The toxic effects and organ dysfunction caused by immunoglobulin deposition, however, differ in severity, clinical presentation and prognosis from that caused by 'amyloidogenic' light chain deposition as seen in amyloidosis. Nests of myeloma cells can cause osteolytic lesions across the entire skeleton. Radiologically, these lesions appear as sharply defined unilocular or multilocular radiolucencies without sclerotic borders or as a general reduction in bone density. The radiologic differential diagnosis involves intraosseous malignancies, various odontogenic tumors, acute osteomyelitis or metastases. A 60-year-old man was referred to Batista Memorial Hospital of Fortaleza City for evaluation of generalized swelling in the jaws. The patient denied pain and discomfort. Intraoral examination revealed two soft tissue masses on maxilla. The panaromic radiography unveiled multiple radiolucencies in maxilla and mandible, and the roots of the mandibular incisors showed

signs of resorption. A biopsy in three different affected areas was performed. Histopathological examination discovered a diffuse infiltration by clonal plasma cells of a mature appearance, compatible with plasmacytoma. It could also be observed the presence of eosinophilic amorphous and fibrillar material consistent with amyloid. The eosinophilic amorphous material showed apple green birefringence with congo red stain when under polarized light. The immunohistochemistry showed focal positivity for Ki67 and diffuse positivity for CD168, also showed intense presence of lambda light chains and a restricted kappa light chain population, confirming a diagnosis of plasmacytoma/MM.

#### Case 8

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# Pleomorphic leiomyosarcoma: first report in the oral cavity

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Leiomyosarcoma (LMS) is an exceptionally rare tumor in oral cavity due the paucity of smooth muscle tissue in the mouth. The classical LMS can present focal areas of pleomorphic cells, however, the coexistence of large pleomorphic component lets the tumor to be designated as pleomorphic leiomyosarcoma (PLMS). This variant of LMS is a high grade sarcoma with an aggressive course. Until this moment, PLMS was not described in mouth. This report describes a case in a18 years-old man complaining of a painful mass in the retromolar area lasting for 15 days. He had a previous history of third molar extraction in this area. Under clinical exam there was a granulomatous, ulcerated tumoral mass extending in the retromolar area. This hyperplastic tissue was removed by incisional biopsy and sent to histopathological exam. The microscopic analysis revealed an extremely atypical tumoral proliferation, with scarce areas of cells poorly arranged in fascicles and mixture of polygonal, rabhdoid, epithelioid, spindle cells and multinucleated bizarre cells. Atypical mitoses were frequent. Immunohistochemistry was performed including Desmin, SMA, h-caldesmon, S-100, Vimentin, Ki-67, AE1-AE3, HHF-35, HMB45. The pleomorphic areas were strongly positive for h-caldesmon and SMA, and were negative for the others markers. Proliferative index by Ki-67 was higher than 90%. The final diagnosis was PLMS. The treatment was a partial mandibulectomy. The patient died few months after the surgery.

### Case 9

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# Diagnosis of nodular lesion in the infraauricular region: a case report

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Acinar cell carcinoma (ACC) is an uncommon malignant salivary gland neoplasm that usually exhibits indolent clinical behavior. It represents 1-3% of all salivary gland neoplasms, often affecting the parotid gland. The diagnosis is microscopic, especially with the aid of histochemical staining by Hematoxylin and Eosin (HE) and Periodic Acid-Schiff (PAS). A 59-year-old female presented an asymptomatic nodular lesion in the right infra auricular region with an evolution time of 2 years. Ultrasonography revealed a heterogeneous, hypoechogenic nodular lesion of 0.7 cm in diameter, suggestive of sebaceous cyst. Considering the image and clinical findings of the lesion, an incisional biopsy was performed and the specimen was referred for histologic examination. Microscopic findings showed a malignant neoplasm with solid and microcystic areas presenting hyaline material inside, permeated by a stroma densely collagenated. The neoplasia consisted of polygonal acinar cells with abundant basophilic granular cytoplasm (similar to zymogen granules) and eccentric hyperchromatic nuclei, as well as, cubic cells with eosinophilic cytoplasm and small dark nuclei. The acinar cells were strongly positive for PAS staining and weakly positive for Mucicarmin. The final diagnosis was ACC and the patient was referred for treatment at a specialized cancer center.

### Case 10

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# Secretory carcinoma on buccal mucosa: a case report

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Secretory carcinoma (SC) of salivary glands is a lowgrade adenocarcinoma that harbor the translocation, t(12;15) (p13;q25), leading to an ETV6-NTRK3 fusion oncogene. SC demonstrates a wide morphological spectrum, so immunohistochemistry is very important for its diagnosis. A 28-years-old female patient presented a nodule in the buccal mucosa, with approximately 5 months of evolution. A presumptive clinical diagnosis of fibroma was established and the lesion was completely excised. Histopathologic analysis of the lesion disclosed a well-circumscribed tumor with duct-forming structures disposed in a microcystic pattern. An abundant bubbly or homogeneous secretion in the cystic spaces was also observed, which was PAS-positive. The tumor cells were positive for CK-7, S-100 and mammaglobin, whereas CK-14 and p63 demonstrated focal expression. ETV6 fluorescence in-situ hybridization showed one signal per cell and confirmed the final diagnosis of SC. The patient received chemotherapy and after follow-up of 12 months remains alive. In conclusion, SC of the minor salivary glands must be considered as differential diagnosis of nodules in the buccal mucosa, and the correlation of the mammaglobin positive expression with confirmation of ETV6-NTRK3 fusion is essential for a proper diagnosis of SC.

### Case 11

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# Primary syphilis of the oral mucosa

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A previously healthy 56 years old man presented in an Oral Care for further examination of a large bleeding painful ulcer that extended from left lip commissure to central region of buccal mucosa. At clinical examination a vegetative ulcer with the borders covered by a fibrinopurulent membrane was noted. The lesion measured 3.0 cm in extension and had been present for an unknown time; however, according to the patient it increased in size in the past 10-days, when the patient decided to seek for professional care. Besides the lesion the patient presented with constitutional signs suggestive of systemic infection such as lymphadenopathy, loss of appetite and low fever. He denied any previous STD, as well as oral sexual contact. An incisional biopsy was performed and histopathological analysis showed an ulcerated mucosa, lined by a fibrin purulent membrane, and presenting in the lamina propria a dense mononuclear infiltrate predominated by plasma

cells. Granulation tissue, characterized by multiple capillaries surrounded by endothelial cells was also present especially below the ulcerated area. Due to the massive presence of plasma cells, especially surrounding vessels, an immunohistochemical reaction against T. pallidum was performed. A large number of positively stained spirochetal microorganisms, compatible with spirochetes were present both, scattered in the connective tissue and located perivascularly. The patient was submitted to serological exams such as VDRL and FTA-ABS, being positive for both. The patient was treated by an intramuscular injection of G-penicillin (2.400.000 UI) and complete remission of the lesion occurred in a few days.

#### Case 12

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# Soft tissue neoplasm of the lip

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An 18-year-old woman was seen by a dentist from a Public Health Dental Unit with a complaint of an ulcerated nodule in her upper lip. According to the patient the lesion had been present for 3 years with an increase in size in the past three months. The clinical suspicion was that of a malignant salivary gland neoplasm. An incisional biopsy was taken and the material was sent for histopathological diagnosis. Microscopic examination showed a mucosal tissue, partially lined by squamous epithelium and showing in the lamina propria a diffuse proliferation of spindle cells, in storiform, whorled or with a nodular growth pattern. The possibilities of dermatofibrosarcoma protuberans, myofibroma, solitary fibrous tumor, fibrohistiocytic tumor and schwannoma was considered. Immunohistochemical reactions were performed and the cells stained positively only for vimentin amongst the tested antibodies. A few dendritic S100 positive cells were seen, as well as CD68 positive cells. The tumor was not encapsulated. A diagnosis of Soft tissue neoplasm was rendered and a suggestion of removal of the whole lesion was done. However, the patient was lost to follow up.

#### Case 13

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# Leiomyomatous hamartoma

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Hamartoma is a tumour-like malformation presenting as a mass of disorganized tissue. Leiomyomatous hamartomas (LM) are extremely rare in oral cavity. This report describes a case of oral LM. An 8-year-old female patient was referred for diagnosis of a nodule on the tongue, of unknown duration. Intraoral examination revealed a painless nodule located on the middle-dorsum of the tongue, measuring approximately 1.0 cm. The main hypothesis of diagnosis was granular cell tumor. Under local anesthesia, the lesion was excised. Microscopically, the lesion consisted of randomly orientated bundles of spindle-shaped cells located subepithelially, non-encapsulated, and surrounded by fibrous tissue stroma. These bundles of spindle-shaped cells were intermixed with blood vessels. The cells were positive for alpha-smooth muscle actin and H-caldesmon. Additionally, the cells appeared negative for S-100 and CD-34. The diagnosis of LM was established. Although LM may develop at any oral site, it mainly occurs on the dorsum of the tongue in patients during the first years of life. Thus, similar to other developmental disorders, LM should be considered in the differential diagnosis of nodules on the dorsum of the tongue of pediatric patients.

# Case 14

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# Patient with bilateral generalized lymphadenopathy

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A 76 year-old male presented with bilateral generalized lymphadenopathy, involving preauricular, submandibular, cervical, supraclavicular and axillar lymph nodes. The patient stated three months duration. The affected nodes were firm and fixed. Excisional biopsy of a laterocervical node, level IB was performed. Microscopically, the loss of normal lymph node architecture was evident, substituted by a diffuse and monotonous lymphoid cell proliferation. The infiltrate consisted of small to medium-sized lymphocytes with hyperchromatic and

slightly irregular nuclei. Mitotic figures were frequent. This homogeneous proliferation was intermingled with many epithelioid macrophages and few plasma cells. Occasional perivascular hyalinization was observed. Immunohistochemistry was performed, and the tumor showed positive results for CD20, PAX5, CD5, CyclinD1, and negative for CD3, CD15 and CD30. Ki-67 index was >40%. The diagnosis was mantle cell lymphoma, which represents a mature B cell lymphoma, more commonly affecting lymph nodes and characterized by a CCND1 translocation leading to CyclinD1 overexpression. Histologically it may present a slight nodular pattern that resemble the mantle zone. This is an aggressive lymphoma, which is frequently diagnosed at late stages, with a median overall survival <4 years. However, features observed in the present case as diffuse growth pattern and high proliferation rate is associated with even worse prognosis. On the other hand, primary extranodal disease, has better prognosis.

#### Case 15

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# Gingival tumor in a child

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An otherwise healthy 5-year-old Mexican boy presented with an elevated, smooth surfaced, yellowish, and asymptomatic swelling located in the mandibular lingual gingiva, at the level of the right central and lateral incisors. The lesion had a rubbery consistency, showing sessile base, measuring 0.8 cm in diameter No other lesions were present elsewhere, and no alterations were observed in the periapical radiograph of the involved region. An excisional biopsy was performed under local anesthesia and submitted to histopathologic examination. Tissue sections revealed a poorly circumscribed proliferation of spindle cells with oval nuclei, also presenting an infiltrate of macrophages with a variable degree of lipidization (foamy macrophages), and scattered Touton type giant cells. Cellular atypia was not observed and mitotic figures were rare. The histopathologic diagnosis was Juvenile Xanthogranuloma (JXG) and to confirm it and illustrate the case, immunohistochemistry was performed.

Negative reactions were obtained with the antibodies pancytokeratin AE1/AE3, α-smooth muscle actin, desmin and CD1a; focal cells were weakly positive for LCA, CD34 and S-100; while all mononuclear and multinucleated cells were positive for vimentin, CD68, CD68Kp1, CD63, FXIIIa, CD163 and CD10. Ki-67 index was 10% and 3% in superficial and deep areas respectively, and negative for all multinucleated giant cells. The immunohistochemical results confirmed the diagnosis of JXG. No recurrence was observed at two years follow-up and no other cutaneous or mucosal lesions appeared during the same period. Juvenile Xanthogranuloma (JXG) is a non-Langerhans cell histiocytosis (non-LCH) affecting normolipemic infants and children most frequently in the first year of life, often showing spontaneous regression within 3 to 6 years. Because of the rarity of oral lesions and possible variations in the clinical and histological presentation, the correct diagnosis can be challenging, requiring a careful clinical and histopathological evaluation with adjuvant immunohistochemical studies.

#### Case 16

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# Parotid gland neoplasm with extension to the stensen duct: a case report

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The first description of epithelial-myoepithelial carcinoma (EMC) was in 1972 by Donath et al., and in 1991 WHO recognized it as a subtype of salivary gland adenocarcinoma. It is a rare biphasic tumor that accounts for less than 1% of salivary gland tumors. The objective of this work is to report a case of EMC with atypical presentation with extension through the Stensen duct. Male patient, 64 years old, from Feira de Santana - BA, complaining of bulging in left parotid topography one year previously, progressive growth,

without local pain. The locoregional examination showed normal oroscopy and nodule in the left parotid region measuring 8 cm, hardened, fixed to the deep planes. The patient underwent magnetic resonance imaging (MRI) of the face. A fine needle aspiration (FNA) was requested and, after a 42-days follow-up, the patient had a vegetative lesion on the left cheek mucosa, measuring 1x1cm, at the ostium of the Stensen duct. The FNA result returned findings consistent with salivary gland basaloid cell neoplasia, with surgery being indicated. The patient underwent total parotidectomy with partial preservation of the facial nerve, resection of the entire tract of the Stensen duct, and cervical emptying Ib and II on the same side. The results of the histopathological examination revealed EMC with extension to the salivary duct, measuring 5.3 x 4.1 cm, free margins and without lymph node metastasis. The patient underwent adjuvant radiotherapy (RT), remaining in clinical follow-up for 43 months and showed local recurrence after 27 months, treated with surgical removal without complications.

# **Cases 18 and 52**

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# Tongue metastasis of cutaneous melanoma: two cases report

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Melanoma of the skin is characterized by a high metastatic potential, but metastases to the tongue are very rare. However, late diagnosis is often associated with advanced disease with dissemination to regional lymph nodes and distant metastases. Two female patients in the 7th decade of life presented to our dental service with nodules in the tongue. Both patients had multiple metastases at the time of oral diagnosis and primary melanoma originated on the skin. An intra-oral incisional biopsy was performed under local anesthesia and the histopathologic analysis was characterized by the proliferation of atypical epithelioid cells displaying a poorly delimited cytoplasm and hyperchromatic

nucleus which contained eosinophilic macronucleoli. Immunohistochemistry was performed in both cases to confirm the clinical hypothesis of metastatic melanoma. After the diagnosis of oral metastatic melanoma, the patients were maintained under palliative care and close medical follow-up. One of the patients died four and a half months after the diagnosis of tongue metastasis and the other patient is still alive after 15 months. The present findings highlight the importance of a complete medical evaluation of the patient by anamnesis to identify possible oral repercussions of primary diseases in other organs and/or systems.

## Case 19

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### Leishmaniasis

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A 56-year-old female patient was referred to the dental office for oral examination. Under clinical evaluation the patient presented multiple extraoral nodular lesions of different sizes and scabs on the face and lips. During the intraoral analysis the patient presented an asymptomatic, soft nodule in the hard palate of the same color of the mucosa. In the rest of the skin of the chest, back, arms and legs the patient also presented many nodular lesions and scabs. The time of evolution was 52 years, since she reported that she began to develop after 4 years of age and has not been treated medically. There was any additional relevant clinical data in her past medical history. The differential diagnoses were nodular leprosy and neurofibromatosis. An excisional biopsy was performed on the palate and, histologically, the formation of granulation tissue was observed with multiple small particles of eosinophilic aspect forming spaces in the chronic inflammatory infiltrate. Grocott and PAS stainings were negative and the diagnosis of Leishmaniasis was rendered.

### Case 20

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# Hyperpigmentation by chloroquine

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A 60-year-old female patient with pigmented blue-gray lesion on the hard palate of 6 months of evolution, asymptomatic, presented to our Department. She reported having rheumatoid arthritis and been given 150 mg per day of chloroquine diphosphate as part of the treatment for one year. The differential diagnoses were hyperpigmentation due to drugs, Addison's disease and melanoma, among others. An incisonal biopsy was performed and histologically, subepithelial pigment was found between the fibroblasts and some macrophages that corresponded to melanin and hemosiderin respectively. A final diagnosis of chloroquine pigmentation was established.

#### Case 23

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# Metachronic ameloblastic fibro-odontoma and dentygerous cyst in posterior mandible

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Ameloblastic fibro-odontoma (AFO) is a rare mixed odontogenic tumor with histological features of ameloblastic fibroma in conjunction with the presence of dentin and enamel. It usually appears as a wellcircumscribed radiolucency with radiopaque foci, slow growth and commonly seen in children and young adults. A 13-years-old Caucasian male presented an asymptomatic swelling in the posterior right mandible and ascending ramus. The clinical, imaginological and histopathological findings confirmed the diagnosis of AFO. Interestingly, after 8 months, a radiolucent lesion was observed involving unerupted third molar in the left mandible with the final diagnosis of dentigerous cyst (DC). In this report, we presented a case of a young patient, which exhibited an AFO, and, during the follow-up, a DC was also diagnosed. Although a coincidentally event, metachronic odontogenic lesions suggest a possible common genetic origin, since both can be caused by related cellular signaling pathways. A complete enucleation is recommended for both conditions with low recurrence rates.

### Case 24

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# Palate lymphoid infiltrate: reactive or neoplastic?

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Lymphoepithelial sialadenitis (LESA) mainly affects parotid glands of women with Sjögren's syndrome (SS). Affection of intraoral minor salivary glands has been rarely reported, 90% in palate, and half of them associated with SS. Distinguishing this entity from an initial stage of a mucosa-associated lymphoid tissue (MALT) lymphoma is challenging. Woman, 56 years-old, presented a painless soft tissue swelling in the hard palate. The covering mucosa was slightly granular with focal erythematous areas. Salivary gland tumor was suspected and an incisional biopsy was performed under local anesthesia. Histopathological analysis revealed a fragment of palatal mucosa covered by an intact squamous epithelium and a dense and diffuse proliferation of small lymphocytes replacing the entire lamina propria, numerous lymphoepithelial lesions were identified. Also, acinar destruction was evident, only ducts persisted towards the base. No "halos" of clear monocytoid centrocyte-like cells or lymphoid follicles were noted. Immunohistochemical analysis with pan-cytokeratins (AE1/AE3) highlighted the lymphoepithelial islands and the remaining ducts, the lymphocytic infiltrate were mainly B-cells (CD20), Kappa and Lambda showed no light-chain restriction, and proliferation index was less than 1% as measured with Ki67. A diagnosis of LESA was made and an excisional biopsy was performed showing same features. These findings led to the work-up for Sjögren's syndrome. Patient referred fatigue, eye, mouth and vaginal dryness. Laboratory studies revealed positivity to Anti-SSA, Anti-SSB, and rheumatoid factor. The patient was referred to a rheumatologist for further evaluation, and close long-term follow-up was suggested.

### Case 25

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# Oral manifestation of secondary tuberculosis: a case report

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A 31-year-old men presented at a private dental clinical in October/2014, complaining about an symptomatic wound on left buccal mucosa, lasting around 2 months. His medical history included persistent cough and use of illicit drugs. No other contributing information was collected from the anamnesis. The intra-oral examination revealed an ulcerated sessile nodule measuring about 2.5 cm. The extra-oral examination revealed no abnormalities. An incisional biopsy was performed. The specimen was submitted for histopathological examination at Center of Diagnosis of Oral Diseases (Federal University of Pelotas - Brazil). Microscopic evaluation of sections stained with hematoxylin-eosin (H&E) showed a chronic granulomatous inflammatory process, with participation of epithelioid and multinucleated giant cells, as well as areas of deposition of eosinophilic material interpreted as caseous necrosis. Periodic Acid-Schiff (PAS) and Ziehl-Neelsen were done. The later showed mycobacteria, usually isolated, present on giant cell and inflammatory infiltrate. The patient was referred to the Infectious Diseases Department of Medical School because histopathological exam was suggestive of tuberculosis. The chest radiograph presented nodules and micronodules in the upper pulmonary lobes, compatible with active tuberculosis. The serological tests revealed negativity for HIV and hepatitis C and positivity for syphilis. The patient started the antimicrobial treatment and 6 months after the diagnosis the chest X-ray showed no signs of lung disease.

### Case 26

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# High grade transformation of acinic cell carcinoma of the parotid gland

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Acinic cell carcinoma (ACC) is a low grade malignant salivary gland neoplasm with favorable prognosis. High-grade transformation in this tumor is rare and it is characterized by a low- and high-grade component juxtaposed with one another. The outcome is a more aggressive clinical course than the classic ACC. We hereby report a case of ACC in a 56-year-old woman with a history of a right parotid nodule. After surgery the surgical specimen was histopathologically evaluated and showed classic areas of acinic cell carcinoma together with a undifferentiated component. The patient is under follow-up without disease. Given the high incidence of metastases and morbidity, recognition and description this entity is necessary due treatment implications and prognosis.

### Case 27

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# Aggressive tumor in a 11 year-old child

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A Peruvian 11 year-old child, presented with a tissue neoformation in the left maxilla. Microscopic analysis revealed a diffuse proliferation of large and highly pleomorphic cells in an hemorrhagic background, with occasional lymphocytes and blood vessels. The tumoral cells presented a large eosinophilic cytoplasm and marked nuclear and cellular atypia. Some of the pleomorphic cells presented kidneyshaped nuclei (hallmark cells) and other cells were multinucleated in a wreath-like pattern. Numerous mitoses and atypical mitoses were present. Due to the undifferentiated morphology of the tumoral cells, a large immunohistochemical panel was performed. Desmin was negative decreasing the possibility of rhabdomyosarcoma, while LCA was focally positive. Then a immunohistochemical panel for lymphoid neoplasms was performed, the reactions were negative for CD20, CD79a, CD138, VS38c and CD3. Positivity was observed for CD30 with membrane and perinuclear dot pattern, CD43, Granzyme, EMA, ALK, and Ki-67 with a rate about 100%. Based on these results, the diagnosis was anaplastic large cell lymphoma, ALKpositive (ALK+ ALCL). This is an aggressive CD30positive T-cell lymphoma that presents a chromosomal translocation in the ALK gene. It represents 10% to 20% of the lymphomas in the childhood, mainly in the range age of 10 to 14 years old. The diagnosis of this lymphoma may be difficult, since some cases appear as a "null phenotype" with loss of the pan T antigens, about 75% of the cases are negative for CD3. Alternatively can be used CD2, CD5 or CD4. Also CD43 positivity is observed in two thirds of the cases without being specific for the cell lineage. Also positivity for cytotoxic granules as granzyme and perforin may be seen. However the most important markers for the diagnosis of ALK+ ALCL are ALK, CD30 and EMA.

#### Case 28

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# Sporotricosis manifesting in the submandibular region

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A 21-year-old female was referred for evaluation of a swelling in the right submandibular region causing local discomfort lasting 2 months. Clinical examination showed a diffuse fibrous partially fixed swelling measuring 3,0 x 3,0 cm covered by normal skin in the right submandibular region. Provisional clinical diagnosis was a Salivary gland tumor and an incisional biopsy was performed under general anesthesia. The HE-stained sections showed a granulomatous reaction with well-organized granulomas and focal areas of neutrophilic abscesses. PAS, Grocott and Ziehl-Nielsen stainings were negative. Patient was referred to a infectologist and culture from the lesion content was positive for Sporotrix, rendering a diagnosis of Sporotricosis. The patient was prescribed daily doses of 100 mg oral intraconazol and responded well to therapy. In conclusion, sporotricois should be included in the differential diagnosis of submandibular and cervical swellings.

#### Case 29

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# Uncommon variant of a well-known lesion: diagnostic discussion

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A 59-year-old woman presented with a 2cm submucosal painless nodule in the middle third of the dorsal tongue, with a clinical evolution of one year. On magnetic resonance imaging examination, a 14 x 26x 13 mm lesion, with an intermediate signal, was noted. Based on the clinical diagnostic hypothesis of leiomyoma and solitary fibrous tumor, an incisional biopsy was performed. Histologically, the muscle fibers were diffusely infiltrated by numerous blood vessels, mainly capillaries, of varied size, and adipose tissue. Sometimes, perineural invasion by tumor cells was seen. Endothelial cells were positive to CD34, and pericytes and smooth muscle in blood vessels walls were positive to smooth muscle actin antibody. Several lymphatic vessels present in the lesion were highlighted by D2-40 antibody. The final diagnosis was intramuscular hemangioma and the excision of the lesion was performed. Intramuscular hemangioma represents less than 1% of all hemangiomas. In the head and neck region, it occurs mostly in the masseter, temporalis and sternocleidomastoid muscles. Rare cases are located in the tongue. Besides its infiltrative growth pattern and several worrisome histological features, as increased mitotic activity, plumpness of the nuclei, intraluminal papillary projections or perineural infiltraction (as in the present case), the lesion is benign, and the complete surgical excision is the preferred treatment of oral lesions.

### Case 30

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# Maxillary adenoid cystic carcinoma

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A 36-year-old female patient presenting a painless swelling at the right maxilla, resulting in a marked facial asymmetry; intraorally it was observed a swelling with erythematous and focally ulcerated surface, affecting gingiva and crossing the midline of the hard palate. Panoramic radiography showed an ill-defined radiolucency, involving the right maxilla at the level of alveolar ridge and opacifying the maxillary sinus. Incisional biopsy was performed and revealed basophilic cribriform islands and solid sheets of neoplastic cells invading bone trabeculae. Most of the tumoral cells presented myoepithelial characteristics and few true luminal/epithelial cells were observed. To demonstrate the epithelial-myoepithelial pattern, immunohistochemical reactions were performed: AE1/AE3, CK5, CK7, CK14, CK18, CK19, CK20, p63, α-SMA, calponin, CD117, S-100 and Ki-67; as well as double immunohistochemical staining for AE1/AE3 and α-SMA. According to the histopathological an immunohistochemical features, the tumor was diagnosed as adenoid cystic carcinoma.

# Case 31

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# Mandibular swelling as first multiple myeloma clinical manifestation

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Multiple myeloma (MM) usually manifests as a tumor involving the marrow of several bones, and develops more commonly in the skull, vertebrae, and pelvis. The occurrence of a mandibular lesion as the first sign of MM is uncommon. This report describes a case of MM diagnosed because of a mandibular lesion. Patient R.S.A., 69 years old, female, presented alveolar ridge expansion, asymptomatic, with two months of evolution, in posterior region of the mandible. Imaging exams showed lytic,

irregular, destructive lesion, with poorly delimited margins and containing mineralized material with sunburst appearance. Histopathological analysis revealed cords and large nests containing monotonous proliferation of round and hyperchromatic cells of plasmacytoid morphology, besides typical and atypical mitoses, cellular pleomorphism and collagenized stroma. After immunohistochemical evaluation, these neoplastic cells were positive for CD38, CD138, MUM1, LAMBDA, negative for CD20, KAPPA, LMP-1 and with cell proliferation index (Ki-67) of less than 5%. The anatomopathological diagnosis of plasmacytoma/ MM was performed. Other complementary exams were performed and presented the following results: Negative anti-HIV (1 and 2) serology, lesions in skull and mandible during the evaluation by bone scintigraphy and detection of monoclonal gammopathy in the electrophoresis of serum proteins. The diagnosis of MM was confirmed and the patient underwent chemotherapy (dexamethasone, cyclophosphamide and thalidomide) and symptomatic/ preventive treatment with acetylsalicylic acid, omeprazole, sulfamethoxazole, trimethoprim and ondansetron. During the follow-up of the patient after 3 months of treatment, clinical and imaging regression of the lesion was observed, in addition to the reduction of the monoclonal gammopathy. This case illustrates the contribution of oral evaluation in the diagnosis of systemic diseases with generalized involvement, such as MM.

#### Case 32

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# Desmoplastic fibroblastoma: case report

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The desmoplastic fibroblastoma (DF), also known as collagenous fibroma, is a rare soft tissue lesion with an uncertain etiology. DF is commonly found in the subcutaneous and intramuscular tissues of the upper extremities, having a wide anatomic distribution. Its occurrence in the oral cavity is rare, and only ten cases have

been reported so far. This report presents a case of a 59-yearold woman, who presented a well-circumscribed, lobulated, firm nodule located on the right mandibular ridge in the region of tooth 44, covered by normal mucosa, measuring 10mm in diameter. Radiographic examination did not demonstrate any bone involvement. The histopathological examination revealed a non-encapsulated, well-defined lesion characterized by proliferation of cells in highly collagenous matrix. At high power, fibroblasts presented spindle to stellate shapes, with rounded nuclei. The connective tissue consisted of dense collagen bundles with inconspicuous blood vessels. Mitotic figures and necrosis were not seen. Tumor cells were strongly and diffusely immunopositive for vimentin, and some cells were focally positive for alpha-smooth muscle actin. The cells were negative for desmin, S-100, CD34, and CD68. Histochemical analysis with Masson's Trichrome stain confirmed the collagenous nature of the stroma. The histological and histochemical findings combined with the immunohistochemical profile are consistent with desmoplastic fibroblastoma. The patient is currently under follow-up and is free of disease for 18 months. The findings observed in the present case highlight the importance of a careful microscopic analysis of these rare lesions.

#### Case 33

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# Myofibroblastic Sarcoma

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Myofibroblastic sarcoma is a malignant tumor with myofibroblastic differentiation and variable morphological characteristics. Myofibroblastic sarcomas are uncommon but have been described in different sites such as breast, thyroid, skin, tongue, and jaw. We described a 27-years-old male with complaint of painful nodule in the left soft palate with 1-month of evolution. The microscopic examination revealed a mesenchymal neoplasia composed by spindled to stellate-shaped cells arranged into intersecting fascicles. Immunohistochemical reactions showed negativity for S100, laminin, CD34, ALK1, CK pool and desmin. The positivity was obtained for SMA, calponin, collagen IV and Ki-67. The diagnosis of myofibroblastic sarcoma was confirmed, and the patient was referred for surgical treatment. At the 10-month follow-up evaluation the patient was free from this tumor.

### Case 35

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# Melanotic neuroectodermal tumor of infancy in maxilla: case report

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Melanotic neuroectodermal tumor of infancy (MNTI) is a very rare, melanin-containing neoplasm, and it is usually diagnosed during the first year of life. We report the case of a 6-month-old baby girl who presented a swelling in the anterior maxilla, without pain and no bleeding. Extraoral examination showed superior displacement of the paranasal region and upper lip on the right side of the face. Tomographic reconstruction revealed a unilocular and expansive area associated with the upper right central primary incisor. An incisional biopsy was performed based on the initial clinical diagnosis of dentigerous cyst or adenomatoid odontogenic tumor. Microscopic examination revealed a biphasic cell population, consisting of small rounded neuroblastic-like cells and epithelioid cells containing melanin. The melanocyte-like component was strongly positive for HMB-45 and Melan-A, but weakly positive for S100. Based on these findings, definitive diagnosis of MNTI was established. Enucleation of the lesion was performed by careful curettage. After 2 years of follow-up, no evidence of recurrence was observed.

# **Case 37**

**DOI:** 10.5935/2525-5711.20170078

# Ameloblastic carcinoma of mandible

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Ameloblastic carcinoma is the malignant counterpart of ameloblastoma. It is rare and microscopically characterized by a combination of both cytologic malignancy and ameloblastoma. Herein, it was described a case of ameloblastic carcinoma of mandible. A 74 year-old female was referred to the stomatologist for evaluating an asymptomatic oral swelling lasting 6 months. Clinical exam revealed a reddish nodule extending from the posterior region of right alveolar mucosa to right buccal mucosa. There was an associated ulcer. Radiographic exam revealed two relatively well-delimited radiolucencies located in the posterior segment of the right mandible with no teeth association. It was performed an incisional biopsy under clinical diagnosis of giant cell lesion. At macroscopic analysis, there was a single white soft tissue measuring 1.5 cm in diameter. At microscopy analysis, there was an odontogenic epithelium proliferation the lining epithelium. They characterized by cellular nests and trabeculae having peripheral palisading and reverse nuclear polarity, some areas of stellate reticulum in the center and common clear cells. There were basaloid areas, pleomorfism and hypercromatism. Ameloblastic carcinoma diagnosis was established. Patient was referred for an oncologic center for suitable management.

#### Case 38

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# Benign lymphoepithelial lesion affecting minor salivary gland

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Benign lymphoepithelial lesion represents enlargement of salivary glands especially parotid glands affecting more women, being infrequent in the minor salivary glands. We described a 62-year-old female patient diagnosed with Sjogren's syndrome that presented an enlargement in palate. Intra-oral examination revealed an asymptomatic sessile nodule in the transition of soft and hard palate with reddish color, fibrous consistency, oval shape, smooth surface, measuring 1 cm in diameter. Clinical differential diagnosis included reactive lesion as fibrous hyperplasia. An excisional biopsy was performed. Histopathological aspects revealed an intense inflammatory infiltrate associated with the destruction of the salivary acinar

structures and presence of mioepithelial islands. Immunohistochemical analysis including CD3,CD20, Kappa, Lambda,CD136 and Ki-67 confirmed the diagnosis of benign lymphoepithelial lesion. No recurrence was observed after one-year follow up.

### Case 40

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# Radiolucent lesion associated to an unerupted deciduous tooth

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A Japanese 5 year-old boy presented with complain regarding the lack of eruption of the right mandibular deciduous second molar. Intraoral examination revealed mobility of the first deciduous molar and an asymptomatic vestibular swelling at the affected site, the surface mucosa was apparently normal. Panoramic radiography showed a well-defined radiolucent lesion involving the crown and one root of the unerupted tooth, it was also observed root resorption of the deciduous molar with mobility. By cone beam computerized tomography it was observed buccal cortical expansion, extending from the unerupted molar to the deciduous canine. A biopsy was performed and the histopathological diagnosis was primordial odontogenic tumor. Later, the excision of the lesion was performed under general anesthesia, with extraction of the retained tooth. The gross specimen consisted of a smooth surfaced whitish spherical solid mass, measuring 0.9 cm in the major diameter. Histologically the tumor presented a cell-rich mesenchymal tissue with stellate and fusiform fibroblasts in a myxoid background, resembling the dental papilla of the tooth germ. The periphery of the tumor was completely coated by a heterogeneous non-keratinized epithelial tissue with columnar to cuboidal morphology at the basal layer and squamous in the surface. In some areas of the covering epithelium, enamel organ-like structures composed by basal columnar cells and suprabasal cells resembling stellate reticulum. Evidence of mitotic activity, cellular atypia, odontoblastic differentiation or induction of dental hard tissues was not observed. The microscopic features of the surgical specimen confirmed the diagnosis of primordial odontogenic tumor, which is a benign mixed odontogenic tumor, recently recognized in the World Health Organization (WHO) classification of Head and Neck tumours in 2017, and only eight cases have been reported worldwide. Further reports and series studies are required to know better the clinicopathologic features and clarify the pathogenesis of this tumor.

#### Case 41

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# Fusocellular proliferation in face: differential diagnosis and immunohistochemistry analysis

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A 64-years old male patient, farmer, complaining of a painful swelling in the right side of the face for 3 months, which appeared after a trauma. The patient also reported to have undergone surgical procedure to extract teeth 14 and 15, with the use of antibiotics and anti-inflammatory, but showing no improvement. The lesion grew in size with the presence of throbbing pain. On extra-oral examination it was observed a nodular lesion, normal color, smooth surface, sessile, with well defined limits, fibroelastic consistency and measuring 4.5 x 4.0 x 3,0 cm. Chest X-ray was normal excluding the possibility of bone fracture. Incisional biopsy was done which revealed benign neoplasm of mesenchymal origin characterized by the fusocellular proliferation. Patient underwent wide excision under local anesthesia. Histopathological examination showed a well circumscribed, encapsulated spindle cell lesion with hyaline and mixomatous areas. The cells were arranged in irregular bundles and fascicles. These cells appeared to be immature myofibroblasts and fibroblasts. Immunohistochemistry showed that the tumour cells were positive for Vimentin, focally positive for Smooth Muscle Actin (SMA) and negative for S100 and CD34. Ki67 labelling index was less than 1%. Due to the clinical, histopathological and immunohistochemical findings, the diagnosis of nodular fasciitis was established. Post-operative period was uneventful, and after 6 months of follow-up there were no signs of relapse.

#### Case 42

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# Rhabdomyosarcoma affecting the maxilla

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Rhabdomyosarcoma (RHS) is classified as a skeletal muscle tumor arising from undifferentiated skeletal tissue, affecting the head and neck region in approximately 40% of the cases. RHS is the most common primary soft tissue malignancy in children. It often occurs as an unilateral and solid mass during the first decades of life. The treatment and prognosis are related to the stage of the tumor, and therefore, accurate and prompt diagnosis is crucial. A 35-year-old man was referred to our service with a progressive propotosis and loss of vision in the left eye. The patient was previously seen by an ophthalmologist 3 months earlier for a reddish swelling in the lower eyelid with no conclusive diagnosis. The lesion rapidly increased and caused facial asymmetry. Intraorally, the patient presented a normal colored lesion on the left maxillary molars region. A CT scan obtained after the first visit revealed an illdefined hypodense lesion extending from the cranium basis to the maxilla. An incisional biopsy was performed on the maxillary lesion. Histologic examination disclosed small, round malignant tumoral cells with hyperchromatic nuclei. It was observed spaces containing small, round and poorly differentiated skeletal muscle cells: fibrovascular stroma is lined by undifferentiated round cells and differentiating cells with abundant eosinophilic cytoplasm consistent with rhabdomyoblasts. Immunohistochemistry revealed positive staining for vimentin, desmin, myo-D1, miogenin and Ki-67 (>90%). A diagnosis of RHS was made. The patient was referred for the head and neck oncologic treatment.

#### Case 44

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# Familial gigantiform cementoma

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A 17-year-old male was referred for evaluation of a painless swelling in the left mandibular region lasting 6 months. Clinical examination showed a hard buccal swelling in the area adjacent to teeth 34, 35, 36 and 37 covered by normal oral mucosa. Panoramic radiograph and computed tomograph showed a well-defined radiolucent area with radiopaque foci in the area measuring 4,0 cm in its greatest diameter. Some additional discrete diffuse radiolucencies were observed in the right mandible. Provisional diagnosis included Ossifying fibroma and Calcifying epithelial odontogenic tumor and an incisional biopsy was performed under local anesthesia. The HE-stained sections showed a benign fibroosseous lesion, compatible with Ossifying fibroma. After receiving the report, the patient reported that his father (a 53-year-old male) was diagnosed with a similar entity some years before. Clinical examination of his father showed the presence of diffuse swellings in both posterior maxilla and mandible. Panoramic radiographs and computed tomograph showed multiple large radiopaque condensations in all four quadrants and in the skull. An incisional biopsy was performed under local anesthesia in the mandible and the HE-stained sections showed a benign fibroosseous lesion. Both the patient and his father were systemically investigated and no other bone lesions were found; serum markers of bone metabolism, including Calcium, phosphorus, alkaline phosphatase and parathyroid hormone were all within normal limits. Final diagnosis was benign fibroosseous lesions compatible with Familial gigantiform cementoma. The patient was managed with local conservative surgical removal of the lesion in the left mandible. Both the patient and his father remain in clinical and radiological follow-up. In conclusion, familial gigantiform cementoma should be considered in the differential diagnosis of multiple benign fibroosseous lesions.

#### Case 45

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# Large cell (undifferentiated) carcinoma ex pleomorphic adenoma

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Carcinoma ex pleomorphic adenoma (CXPA) is a rare malignant tumour of the salivary gland. The tumour

histopathology comprises a variable proportion of pleomorphic adenoma and carcinoma. The malignant component can be a specific carcinoma type, including the several histopathological types. The aim of this case is to report a large cell (undifferentiated) CXPA parotid region in 54-year-old woman. The patient underwent surgical ressection and the microscopic findings in malignant component were poorly formed duct-like structures with lack features of acinar, ductal, or myoepithelial differentiation. The patient is under follow-up and without disease after 24 months from surgery. Undifferentiated salivary gland tumors are highly aggressive neoplasms with frequent metastases and a poor prognosis. The lack of histomorphologic features of either glandular or epithelial differentiation may be misleading, so the recognition and knowledge this entity is important to avoid mistakes in diagnosis.

### Case 46

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# Glomus tumor of the upper lip

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The glomus tumor (GT) occurs most commonly on the distal extremities. GT affecting the oral cavity is extremely rare. The aim of this report is to describe a GT in the upper lip. A 51-year-old female patient presented with a painful nodule located in the upper lip, with 6 months of duration. Intraoral examination revealed a submucosal, smooth-surfaced, normochromic, and well-circumscribed nodule, measuring about 1.0 cm of diameter. The main clinical diagnosis was pleomorphic adenoma and canalicular adenoma. Under local anesthesia, the lesion was excised. The histopathological examination revealed an encapsulated tumor. The tumor cells showed a granular eosinophilic cytoplasm with prominent basophilic nuclei, ranging from rounded epithelioid to spindle-shaped morphology. Spindle cells nodules protruding into vascular lumens were observed, besides hemangiopericytomatous areas. The tumors cells formed two patterns of arrangement, one loose surrounding the larger vessels, and another solid, with dense sheets of tumor cells permeated. The perivascular concentric layers of rounded cleareosinophilic cells around larger vessels resembled the glomus body. The tumor cells were positive for alphasmooth muscle actin, HHF35, and H-caldesmon. Basement membrane material around tumoral cells was positive for type IV collagen in a chicken wire pattern. Desmin was negative. According to these features, the diagnosis of GT was established. No recurrence was observed after 18 months of follow-up. Oral GT is very rare and it appears as submucosal nodule, with good prognosis.

### Case 47

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# (Hemangio)ameloblastoma of mandible: a case report

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A 66-year-old male patient was referred to a Surgery Ambulatory, presenting an extensive lesion on the body-angle-ramus of the right side of mandible for four years. Computed tomography revealed unilocular hypodense mandibular lesion associated with extensive bone destruction. Incisional biopsy revealed a cystic lesion and a subsequent marsupialization, caused an intraosseous communication with the mucosa. Intra-oral examination revealed a normocromic diffuse swelling, with pain at palpation and a hole draining foul-smelling secretion. Radiographically, the lesion was radiolucent and multiloculated. Excisional biopsy was performed and microscopic examination revealed odontogenic neoplasm lesion, with high cell proliferation, delimiting the looser central area similar to the starry reticulum of the enamel organ. Some central cells are spindle-shaped, opening into large blood-filled or thrombosed. Sometimes this epithelium opens into large cystic spaces with epithelium lining the lumen. Some cells are oval and discreetly pleomorphic, forming denser layers. The histopathologic diagnosis of atypical hemangioameloblastoma was established. Patient is under clinical follow-up with no recurrence after 2 years.

#### Case 48

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# Massive extrusion of calcium hydroxide paste including barium sulfate during endodontic treatment: a case report

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A 31-year-old female patient was referred to the Piracicaba Dental School, University of Campinas, presenting paresthesia on the left-sided mandible, with one month of evolution. Her previous medical data was unremarkable. In terms of dental history, the patient reported acute pain during the endodontic treatment of the lower left first molar, which led to the extraction of the tooth and the curettege of the periapical bone. The material was submitted to histopathological analysis. Hematoxylin and eosin staining showed particles of crystalloid foreign bodies, with greenish coloration and fragments of a necrotic pulp in demineralized portion of the root canal and adjacent hard periapical tissue. An inflammatory infiltrate was also found on the apical adjacent soft tissue. Polarized light microscopy identified abundant birefringent granulated material in demineralized fragments of the tooth root canal and adjacent soft and hard periapical tissues. Analysis by energy-dispersive X-ray spectroscopy detected barium and sulfur energy peaks. The digital panoramic radiographic view 4 weeks after the clinical intervention showed overspread radiopaque material in left mandibular area and diffuse bone resorption with epicenter in the area of the extracted tooth. Despite surgical management, the paresthesia of the left-sided mandible is stable and the digital panoramic radiography showed a bone resorption healing, after 18 months of follow-up. Accidental extrusion of injectable calcium hydroxide paste with barium sulphate opacifier agent was confirmed after contact with the dentist responsible for endodontic treatment. Clinicians should be aware of the possibility of extrusion of intracanal medicaments to adjacent root areas and subsequent damage to the bone and inferior alveolar nerve and, for this reason, injectable calcium hydroxide should be applied with care during clinical endodontic procedures.

### Case 49

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# Monostotic fibrous dysplasia

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A 63-year-old male was referred to the OROCENTRO Clinic - Piracicaba Dental School - due to a swelling at the right side of the face. Patient stated that first noted the swelling for over 20 years. The patient was submitted to multiple tooth extractions on the region due to pain. After extractions pain ceased, nevertheless the swelling kept growing. Extraoral examination revealed a hard swelling on the right side of the face causing elevation of the nose wing and deletion of nasolabial groove. Intraoral examination revealed a hard, normochromic swelling on the right maxilla affecting both palatine and vestibular surfaces with deletion of maxillary vestibule. Digital panoramic radiography showed a poorly defined radiopaque image with "ground glass" aspect. Computed tomography revealed a mixed density image affecting the floor of the maxillary sinus and vestibular and palatine cortical bone of the right maxilla. Incisional biopsy was performed under local anaesthesia on the vestibular surface of the right maxilla using a Trephina drill (7 mm X 180mm). Histopathological analysis presented fine branching curvilinear/irregular trabeculae of woven bone among a fibrous cellular stroma and was compatible with Monostotic Fibrous Dysplasia. Patient did not present signs of post-operatory infection or dehiscence and was referred to an appropriate treatment service for surgical planning.

### Case 50

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# Central ossifying fibroma

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A 22-year-old male was referred to the OROCENTRO Clinic - Piracicaba Dental School - due to a small and slightly painful swelling at the right side of the maxilla. Patient stated that first noted the swelling for about one week. Extraoral examination revealed preserved facial symmetry. Intraoral examination revealed a hard, normochromic swelling on the right maxilla affecting the palatine surface and a small swelling with a slightly deletion of the maxillary vestibule. Digital panoramic radiography showed a round radiopaque image at the root area of the posterior maxillary teeth. The panoramic reconstruction of the computed tomography revealed a mixed density nodular image surrounded by a hypodense halo affecting the rootadjacent area of the posterior maxillary teeth with extension to the maxillary sinus. The sagittal and axial planes of the computed tomography revealed a wellcircumscribed image with mixed density at the epicentre and hypodensity surrounding and preservation of the cortical lining. Incisional biopsy was performed under local anaesthesia on the right maxillary vestibule using a Trephina drill (7 mm X 180mm). Histopathological analysis presented a well-demarcated lesion separated from the overlying cortical bone, the lesion was composed by a fibrous cellular stroma and the presence of bone trabeculae of varying sizes and small rounded and basophilic osteoid structures, compatible with Central Ossifying Fibroma. Patient did not present signs of post-operatory infection or dehiscence and was referred to an appropriate treatment service for surgical planning.

#### Case 51

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### Spindle cell carcinoma

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Sarcomatoid carcinoma may affect different parts of the body, being considered as a rare neoplasm. The most affected sites are the oral cavity, larynx, esophagus and skin. An 82-year-old female patient, edentulous, presenting volume increase, asymptomatic, ulcerated, erythematous with a size of 5 x 3 cm in the of upper left molars region

with an evolution time of approximately 6 months. The computerized tomography study revealed a tumor mass that invades the entire maxillary sinus, expanding the bony cortices. an incisional biopsy was carried out and histopathological analysis showing anaplasic spindle epithelial cells and some sarcomatoid-like cells in deeper parts of the tumor and numerous abnormal mitoses, immunohistochemistry study was conducted with AE1/AE3, vimentine, p63, CK18, EMA and Ki-67 antibody found positivity in all of them, ratifying the diagnosis of spindle cell carcinoma. The diagnosis of this kind of infrequent malignant neoplasms requires clinical, radiographic and histopathological correlation, the morphological cell characteristics are important for diagnosis due to biphasic pattern that presents the lesion, hence the immuhistochemistry studies are valuable for diagnosis.

#### **Cases 18 and 52**

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# Tongue metastasis of cutaneous melanoma: two cases reports

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Melanoma of the skin is characterized by a high metastatic potential but metastases to the tongue are very rare. However, late diagnosis is often associated with advanced disease with dissemination to regional lymph nodes and distant metastases. Two female patients in the 7th decade of life presented to our dental service with nodules in the tongue. Both patients had multiple metastases at the time of oral diagnosis and primary melanoma originated on the skin. An intra-oral incisional biopsy was performed under local anesthesia and the histopathologic analysis was characterized by the proliferation of atypical epithelioid cells displaying a poorly delimited cytoplasm and hyperchromatic nucleus which contained eosinophilic macronucleoli. Immunohistochemistry was performed in both cases to confirm the clinical hypothesis of metastatic melanoma. After the diagnosis of oral metastatic

melanoma, the patients were maintained under palliative care and close medical follow-up. One of the patients died four and a half months after the diagnosis of tongue metastasis and the other patient is still alive after 15 months. The present findings highlight the importance of a complete medical evaluation of the patient by anamnesis to identify possible oral repercussions of primary diseases in other organs and/or systems.

#### Case 53

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# Erythematous and diffuse lesion on upper alveolar ridge

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We present a case of an 83-year-old female with a granular, erythematous and diffuse lesion on the right maxillary edentulous alveolar ridge, without any apparent sign of infiltration and with three-month of evolution. The previous medical history was not relevant. Panoramic and periapical radiographs revealed irregular bone loss in the alveolar ridge. Microscopically, incisional biopsy revealed areas of conventional squamous cell carcinoma originating from the mucosal epithelial surface associated with deeper nests which morphology resembled an ameloblastomatous pattern, with peripheral palisading columnar cells showing inverse polarization and central cells in a loose arrangement. However, these ameloblastic nests presented marked cellular pleomorphism and atypia (bizarre cells with variable size, nuclear hyperchromatism, evident nucleoli and atypical mitoses) resembling an ameloblastic carcinoma. Nests of ameloblastic epithelium were surrounded by a collagenous stroma. Moreover, we observed dysplasia of the surface epithelium presenting interruption of the basement membrane and tumoral invasion of the underlying connective tissue. Immunohistochemical analysis of the biopsy specimen reveled intense and homogenous positivity for AE1/AE3 and CK5, in all cells of tumor and surfaceepithelium. GLUT-1 presented cytoplasmic and membrane positivity in most areas

of tumor. The Ki-67 index was about 30% of the tumoral cells. CK18, CK19 and D2-40 showed weak expression in the central cells and in the peripheral columnar cells of the islands.CK7 and CK14 were negative negative in the tumor but the latter, positive in the surface epithelium. Based on this inconclusive immunostaining profile we propose the diagnosis of squamous cell carcinoma with areas of ameloblastic carcinoma. The patient was referred to the oncologist for appropriate treatment.

#### Case 54

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# Asymptomatic gingival lesion

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Rhabdomyosarcoma (RMS) is a rare malignant soft tissue neoplasm composed of neoplastic mesenchymal cells, with varying degrees of striated muscle cell differentiation. With most cases occurring in children younger than 10 years, it is remarkably rare in adults. Further in adults, the typical pediatric rhabdomyosarcoma variants (embryonal and alveolar sub-types) occur less frequently and exhibit predilection for viscera followed by the head and neck region. A rare case rhabdomyosarcoma arising from the gingival mucosa in a 30-year old female patient is herewith reported, with clinical diagnosis of pyogenic granuloma. The correct diagnosis is of critical importance in the therapy of this disease, and immunostains are very helpful, since the treatment is not uniform in the literature because of the rarity of this neoplasm in the adult population.

#### Case 55

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# Peripheral squamous odontogenic tumor

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Odontogenic tumors constitute a group of heterogeneous diseases that range from malignant and benign neoplasms to malformations of dental tissues of self-limited growth. They are principally jaw lesions but some may present as localized gingival swellings, so-called peripheral odontogenic tumors. Squamous Odontogenic Tumor (SOT) is a rare benign odontogenic neoplasm. Most of these tumors are located within the bone, although a few peripheral cases have been reported. This case report provides clinical and microscopic features about a rare peripheral SOT. A 59-year-old female patient, complained of lesion in gingiva that had developed over 14 years. During intraoral examination, it was found a sessile, welldemarcated, dome-shaped nodule with approximately three millimeters in diameter. It was located in the attached gingiva in the mandibular left incisor region. The asymptomatic lesion was covered by normal pink mucosa with a smooth nonulcerated surface. No local traumatic injury to the oral region was recorded, nor were noted abnormalities on periapical radiographs. The presumptive clinical diagnosis included benign mesenchymal neoplasm, gingival cyst of the adult and peripheral odontogenic tumor. Excisional biopsy was performed and histopathological examination revealed a fragment lined by a parakeratinized stratified squamous epithelium. Underlying, there was fibrous connective tissue with islands of squamouslike epithelium of varying sizes displaying flattened layer of cells at the periphery and absence of capsule. Thus, the diagnosis of peripheral SOT was established. The microscopic features this rare odontogenic tumor can be confused with other lesions, such as squamous cell carcinoma and ameloblastoma. Therefore a careful evaluation is essential to avoid misdiagnosis, as a malignant neoplasm, and incorrect treatments.

# Case 56

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# Collagen-rich mandibular lesion: case report and differential diagnosis

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Central odontogenic fibroma (COF) is a very rare and poorly understood odontogenic neoplasm. We report a case of COF with unusual histological features. A 51 year-old male patient presented to our service with an

asymptomatic swelling in anterior region of mandible, with a 3-year evolution history. Imaging studies showed a well-defined multilocular radiolucency with corticated margins, rupture of vestibular cortical bone and divergence of mandibular left canine and incisors. Under the clinical hypotheses of desmoplastic ameloblastoma, squamous odontogenic tumor and calcifying epithelial odontogenic tumor, an incisional biopsy was performed. Histological analysis showed a dense collagenous connective tissue with pleomorphic fibroblasts and scant islands of inactivelooking odontogenic epithelium. The histological diagnosis was COF and a conservative removal of the tumor was performed. The lesion was easily shelled out from the bone. In the surgical specimen, besides the features found in the incisional biopsy, mineralized cementum-like calcifications were found. Immunohistochemical staining using the AE1/AE3 antibody highlighted the epithelial islands, and the initial histological diagnosis was confirmed. Desmoplastic fibroma, sclerosing odontogenic carcinoma and fibro-osseous lesions associated to tuberous sclerosis should be considered in the histological differential diagnosis. The patient is being followed up for 2 years, with no sign of recurrence. Pleomorphic fibroblasts are rarely found in COF, and, to the best of our knowledge, just two cases with this uncommon feature were previously described in the literature.

#### **Case 57**

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# Gingival nodule in a 81-year-old male

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An 81-years-old white man, diagnosed with Myelofibrosis, was referred by his hematologist to the Hospital Dentistry Service for evaluation of a bleeding gingival nodule. The patient was symptomatic and did not present facial asymmetry. On intra-oral evaluation, it was observed an erythematous, smooth and wavy surface, firm consistency, sessile, nodular lesion, measuring 2 x 2 x 1 cm localized at vestibular gingiva between first and second maxillary right premolars. There was bone loss and significant teeth mobility. Extraction of premolars and the surgical excision of nodule was performed. Microscopic analysis revealed sheets of mature and immature myeloid cells positive for CD68, Ki-67 (50%) and myeloperoxidase.

### Case 58

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# Solitary fibrous tumor of the buccal mucosa

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Solitary fibrous tumor is a benign mesenchymal neoplasm of uncertain origin that is microscopically characterized by a spindle cell proliferation. It was primarily described in pleura, but extrapleural sites may also be involved in approximately 25% of the cases, in which 6% of these include the head and neck region. Oral cavity is rarely involved. Herein, it was described a case of a solitary fibrous tumor of buccal mucosa. A 49-year old white-skinned female had a clinical finding of an asymptomatic well-delimited soft submucosal nodule at right buccal mucosa. It was performed an excisional biopsy under clinical diagnosis of fibrous hyperplasia and mucocele. At macroscopic analysis, there was an spherical single soft tissue fragment brown colored measuring 1 cm of diameter. Cleavage revealed a solid lesion easily detachable from the surface. At microscopic analysis, there was a submucosal encapsulated lesion that at low power was characterized by a mixed of hyper and hypo cellular areas. At high power, there was a disorganized spindle cell proliferation associated with delicate blood vessels and inflammatory cells, including lymphocytes and mast cells. The tumor cells were positive for vimentine, CD34 and bcl-2 in the immunohistochemical reactions. rendering the diagnosis of solitary fibrous tumor. Patient is well without signs of recidive in the on year follow-up.

### Case 60

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# Papular lesion in the dorsum of the tongue

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A 35 years old male, complaining of burning and hardening on the dorsum of the tongue, which appeared about 4 months ago. He reported using Nistatin mouthwash for 2 weeks with improvement only from burning, but the hardened area persisted and it was slowly increasing of size. Medical history was not contributory. Intra-oral examination revealed papular lesion, reddish, surrounded by whitish areas, smooth surface, fibroelastic consistency, located in the posterior dorsum of the tongue, and measuring 2.0 x 0.8 cm. Due the main diagnostic hypothesis of pyogenic granuloma an excisional biopsy of the lesion was performed. Histopathological analysis showed epithelial hyperplasia associated with intense and diffuse mononuclear inflammatory cell infiltration predominantly of plasma cells in a perivascular and perineural distribution. Serologic tests were requested and showed the following results: reactive VDRL, reactive FTA-ABS, negative for both HIV-1and HIV-2. After these findings, the patient was asked about his sexual behavior. He informed us that he was unmarried and denied unprotected sexual intercourse, but reported multiple partners. So, the clinical pathologic findings and serologic tests confirmed the diagnosis of primary syphilis. The patient was referred to an medical service and treated with 1 weekly injection of penicillin, 2.4 million units, for 2 weeks, and showed no sign of relapse of the disease with 12 months of follow-up.

#### Case 61

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# Central ossifying fibroma

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A 20-year-old male was referred for evaluation of a painless swelling in the left maxilla lasting 4 years. Clinical examination showed a hard buccal swelling in the left maxilla extending from the anteriro to the posterior region, covered by normal oral mucosa. Panoramic radiograph and computed tomograph showed a well-defined mixed radiolucent-radiopaque area in the area extending to

the maxillary sinus but showing no invasion of adjacent structures. Provisional diagnosis was Ossifying fibroma and an excisional biopsy was performed under general anesthesia. During surgical removal the lesion easily dettached from the adjacent healthy bone. The HE-stained sections showed a proliferation of benign mesenchymal fusiform and focal areas of calification. In some areas there was a concentric disposal around blood vessels and myxoid zones with low cellularity. Final diagnosis was Ossifying fibroma. The patient remains in clinical and radiological follow-up with no signs of recurrence. In conclusion, Ossifying fibroma can show different histological patterns that should be known by oral pathologists.

#### Case 62

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# Adenoid ameloblastoma

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Adenoid ameloblastoma (AA) is a rare odontogenic tumor, as only less than 20 cases have been reported to date. This type of lesion present distinct histopathologic features reminiscent of AME and AOT, and in some cases it produces variable amounts of dentinoid. According to previous reports, AA has a high tendency for recurrence (71% of reported cases) and a locally aggressive biologic behavior. The present case belong to a 62 year-old woman and presented as a slowly growing (several months of apparent evolution) asymptomatic lesion that produced slight vestibular expansion of the right maxillary tuberosity. The histopathological aspects are compatible with the diagnosis of AA, and immunohistochemical markers showed positivity for CK14, CK19, CD138 and was negative for Calretinin and EMA. Ki-67 was expressed in around 2% of basal (peripheral) cuboidal cells. PAS and Alcian blue stains failed to detect mucous-producing cells, and there was no evidence of dentinoid or other mineralized tissue in the tumor. According to our review of the literature, including the present case, there are 17 well recognized examples of this tumor, most of which (7/17) have occurred in the posterior maxilla, without gender predilection (9 male and 8 female patients) in a wide age range (19-79 years).