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# #1 CLINICOPATHOLOGIC FEATURES OF ORAL CAVITY VESICULOBULLOUS DISEASES SEEN IN THE UF ORAL MEDICINE AND DERMATOLOGY CLINICS

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Tuesday, 25th May - 08:00: Poster Presentations - Oral to Poster - Abstract ID: 57

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INTRODUCTION: Vesiculobullous disorders encompass several rare conditions with varying pathology and morbidity, but similar signs and symptoms. MATERIALS AND METHODS: With IRB approval, the record systems of the UF Oral Medicine and UF Dermatology Clinics were searched for cases diagnosed between 2011 and 2020 of the following disorders: pemphigus vulgaris (PV), mucous membrane pemphigoid (MMP), bullous pemphigoid (BP), linear IgA disease, lichen planus pemphigoides, paraneoplastic pemphigus, epidermolysis bullosa acquisita, and angina bullosa hemorrhagica. Exclusion criteria included lack of oral involvement or lack of clinical information and outcome data. Location of lesions within the oral cavity, clinical signs and symptoms, treatment provided, and outcome of treatment were recorded. RESULTS: A total of 38 cases were included in the study consisting of 20 (52.6%) cases of MMP, 9 (23.7%) cases of PV, 6 (15.8%) cases of BP, and 3 (7.9%) cases of EBA. Twenty-two patients were male (57.9%) and 16 were female (42.1%). Age range was 34 to 92 years, with a mean of 69.7 years. EBA was seen in patients with the highest average age (74.7 years) while patients with PV were the youngest on average (59.8 years). About 90% of MMP cases affected the gingiva, while only one case (16.7%) of BP involved the gingiva. Pain was the most common feature associated with both types of pemphigoid. Ulcerations and erosions were the most common features seen in PV and EBA. All (100%) of EBA cases, 88.9% of PV cases, 66.7% of BP cases, and 25.0% of MMP cases required treatment beyond topical steroids. These therapies included topical tacrolimus, prednisone, methotrexate, ritux- imab, mycophenolate, and dapson. PV exhibited the highest percentage (22.2%) of cases refractory to treatment. CONCLUSION: Most MMP cases were effectively managed with topical steroids. EBV, PV, and BP may require more potent therapy.

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## #2 CENTRAL ODONTOGENIC FIBROMA IN ASSOCIATION WITH BROWN TUMOR OF HYPERPARATHYROIDISM IN A PATIENT WITH NF1 AND RENAL FAILURE

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 58

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**Objective:** Neurofibromatosis type 1 is an autosomal dominant disorder caused by mutations in the NF1 gene at chromosomal location 17q11.2. We present a patient with bone abnormalities and a myriad of lesions secondary to his redeveloping renal failure and NF1. **Clinical Presentation:** A 21-year-old male renal transplant recipient with NF1 presented with painless masses and large, irregular radiolucent lesions in the maxilla and mandible. The bone was generally “moth-eaten” with a sparse trabeculation pattern. **Intervention and Diagnosis:** Histologic examination of an incisional biopsy of the left mandibular lesion revealed two types of tissue with distinct histologic features. Approximately 3/4ths of the specimen was made up of variably cellular dense fibrous connective tissue interspersed with numerous inactive islands and cords of odontogenic epithelium. The remaining specimen consisted of abundant multinucleated giant cells embedded within a highly cellular stroma containing extravasated erythrocytes and hemosiderin. Throughout the specimen, seams of lace-like osteoid and irregular trabeculae of woven bone were noted. The lesion was diagnosed as a central odontogenic fibroma (COdF) in association with a central giant cell lesion, most consistent with brown tumor of hyperparathyroidism. The bone changes were interpreted to be highly suggestive of renal osteodystrophy. **Conclusion:** Around 30 cases of hybrid central giant cell granuloma-like lesion in association with central odontogenic fibroma have been reported. This, to the best of our knowledge, is the first reported case of brown tumor in association with COdF. It has been proposed that a primary COdF induces a reactive giant cell response, however, this is unlikely here, given that the primary tumor was probably a ‘brown tumor’. Our case illustrates the point that the giant cell component may be the initiating entity in these hybrid lesions.

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## #3 Primary intraosseous squamous cell carcinoma arising in odontogenic cysts: a series of three cases with immunohistochemical analysis

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Tuesday, 25th May - 08:00: Poster Presentations - Oral to Poster - Abstract ID: 56

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*Dr. Sumita Sam (University of Florida), Dr. Sarah Fitzpatrick (University of Florida), Dr. Mohammed N. Islam (University of Florida), Dr. Indraneel Bhattacharyya (University of Florida), Dr. Donald Cohen (University of Florida)*

### Introduction:

Carcinomatous transformation in odontogenic cysts is rare, with few cases described in the literature. Squamous cell carcinoma (SCC) is the most common form of malignant transformation in odontogenic cysts. Radicular and dentigerous cysts are the most common odontogenic cysts to undergo malignant transformation. We present three cases of primary intraosseous carcinoma (PIOC) arising in odontogenic cysts.

### Method and Materials:

Following IRB approval, cases were identified from the University of Florida Biopsy Service archives (1994-2020). The clinical and demographic data was reviewed, along with radiographs and original slides. An immunohistochemistry (IHC) panel including CK19, p53, and Ki-67 was performed on all cases.

### Results:

The first case occurred in a 67-year-old male with a radiolucency around impacted #17. Microscopically, a verrucous carcinoma was diagnosed arising within a developmental odontogenic cyst, most likely an orthokeratinized odontogenic cyst. The second case involved a 59-year-old male with a lesion adjacent to tooth #15. Histologic examination revealed a well differentiated SCC arising from an odontogenic keratocyst (OKC). The final case involved a 13-year-old male with a radiolucency near teeth #6-7. A well differentiated SCC was diagnosed arising from an OKC. Further investigation to assess for Nevoid Basal Cell Carcinoma Syndrome was recommended for this case, but the outcome was unavailable. IHC analysis revealed P53 positivity for all cases. Ki67 showed 80-90% positivity in the basal layer of the odontogenic epithelium for cases 1 and 3, and 50% positivity for case two. Ck19 was only focal positivity in all cases.

### Conclusion:

We present three cases of squamous cell carcinoma arising within odontogenic cysts. P53 and Ki-67 positivity aid in confirmation of the aggressive nature of each tumor. CK19 staining was weak, despite the obvious odontogenic nature of the epithelium in all cases. Careful follow-up for odontogenic cysts demonstrating recurrence or progressive growth is imperative.

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## #4 Significance of Oral Cavity Biopsy in the Diagnosis of Extranodal NK/T-cell Lymphoma, Nasal Type: A Case Report

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 19

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*Dr. Jamie White (Moun), Dr. Suchie Chawla (Mount Sinai Hospital , New york), Dr. Andrew Ferraro (Mount Sinai Hospital , New york), Dr. Naomi Ramer (Mount Sinai Hospital , New york)*

### Introduction:

Extranodal NK/T-cell lymphoma, nasal type (ENKTL) is a rare, but highly aggressive entity. It occurs most commonly in Asia and native populations of Mexico and Central and South America. This case report details the clinical course of a patient who initially presented for work-up of pancytopenia, and was ultimately found to have a diagnosis of Extranodal NK/T-cell Lymphoma. The patient underwent biopsies of multiple sites including the bone marrow, lymph node, tonsil, and buccal gingiva, with the latter biopsy giving the most definitive, unequivocal result.

### Clinical Presentation:

A 38 year old Asian male presented to the emergency department for pancytopenia and fevers after he was previously found to be anemic with low hemoglobin levels. His physical examination was notable for right tonsillar enlargement with exudate, a white mass on the right palate, sinusitis, and lymphadenopathy. A bone marrow biopsy was performed and diagnosed as EBV lymphoproliferative disorder with T/cytotoxic cell proliferation, and the differential diagnosis included a T/NK cell neoplasm. Subsequent biopsies of a cervical lymph node and the right tonsil were performed with the lymph node being diagnosed as idiopathic multicentric Castleman's disease; EBV-positive, HHV8-negative. As treatment, the patient was started on siltuximab. The necrotic mass on the palate continued to worsen, extending to the maxillary anterior buccal gingiva. Fortunately, after some time, the patient was referred to oral surgery and a biopsy of the right buccal gingiva was performed, revealing the final diagnosis of ENKTL. The patient was diagnosed with stage IV disease, received treatment with the SMILE regimen, underwent a stem cell transplant, and was overall stable 17 months post-transplant.

### Conclusion:

This case highlights the importance of thorough oral examinations and timely sampling of any lesions present, as these practices can aid in arriving at an accurate diagnosis.

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## #5 Oral Lipomatous Neurofibroma: A report of two cases and a review of the literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 29

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*Dr. Shraddha Kamat (New York Presbyterian Queens), Dr. Rachelle Wolk (New York Presbyterian Queens), Dr. Renee Reich (New York Presbyterian Queens), Dr. Paul Freedman (New York Presbyterian Queens)*

Neurofibroma is a benign tumor that originates from the peripheral nerve sheath. This lesion may assume multiple growth patterns and various histologic variants have been described in the literature. We present two cases of lipomatous neurofibroma, a rare variant in the oral cavity. This variant is typically seen as a cutaneous lesion predominately located in the head and neck.

Case 1 was a 62-year-old female who presented with a mass of her left posterior palate. Case 2 presented as a submucosal dorsal tongue mass in an 18-year-old male. Both lesions appeared clinically benign. Neither patient had a known history of neurofibromatosis type 1.

The biopsies from case 1 and case 2 showed similar histopathological features. These two lesions were well delineated submucosal tumors characterized by an admixture of spindled neural cells and mature adipocytes. The mature adipocytes were separate from the regional adipose tissue. The spindle cells contained serpentine nuclei set in a wavy well-vascularized fibrillar stroma. The immunohistochemical staining for both lesions revealed S100 positivity of the tumor cells and negative staining for CD99, CD34, and BCL2. Differential diagnosis for these tumors included solitary fibrous tumor and spindle cell lipoma. However, based on the lack of a hemangiopericytoma-like vascular pattern, wire-like collagen and the pattern of IHC staining, these entities were excluded.

While the diagnosis of a conventional neurofibroma is straightforward, the diagnosis of a lipomatous neurofibroma may be more challenging. Neither of our patients had a known history of Neurofibromatosis type 1 (NF1); however, cutaneous lipomatous neurofibromas have been associated with NF1. Therefore, a patient with a diagnosis of lipomatous neurofibroma should have a medical work-up to exclude this syndrome. With the presentation of two cases of lipomatous neurofibroma arising intraorally, we hope to raise awareness of this distinct and rare morphologic subtype of neurofibroma.

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## #6 Distinct H3F3A Driver Mutation Defines Giant Cell Tumor of Bone: A Necessary Adjunct in the Diagnosis of Such a Lesion in the Hyoid Bone, Presenting as a Base of Tongue Mass

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 33

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**Introduction:** Giant cell tumor of bone (GCTB) is a rare benign tumor mostly affecting the epiphyses of long bones in skeletally mature patients that is rarely described in the head and neck. Conventionally, diagnosis is based on microscopic features, exclusion of mimics, and clinico-radiologic correlation. Morphologic overlap with numerous giant cell-containing lesions of bone and soft tissue complicates the diagnosis. GCTB is characterized by the *H3F3A* mutation, which can be detected immunohistochemically using H3.3 G34W, a monoclonal antibody specific for this mutation.

**Case Findings:** A 57-year-old male presented with dysphagia, dysarthria, and right base of tongue (BOT) mass suspicious for carcinoma. Imaging revealed a heterogeneous parapharyngeal lesion extending from BOT to the hyoid bone. A BOT biopsy demonstrated giant cell-rich granulation tissue and non-endothelial lined bloodlakes, suggestive of aneurysmal bone cyst (ABC). Fluorescent in situ hybridization (FISH) analysis did not identify *USP6* rearrangement, effectively eliminating primary ABC and favoring secondary ABC. Base of tongue resection, partial pharyngectomy and hyoid bone resection followed.

**Results:** Microscopically, the resection revealed a giant cell-rich lesion emanating from the hyoid bone, suggestive of GCTB and thus unlikely soft tissue giant cell tumor. Cellular pleomorphism and atypical mitoses were absent. Stromal cells expressed H3.3 G34W, confirming the diagnosis of GCTB. Zonal features of secondary ABC were juxtaposed to the dominant GCTB.

**Conclusions:** Hyoid GCTB is exceedingly rare, and such a diagnosis presents a conundrum. H3.3 G34W nuclear staining is an indicator of *H3F3A* mutation in GCTB that can help distinguish GCTB from other mimics. Prior reports of hyoid GCTB did not employ ancillary studies to identify *H3F3A* mutation or *USP6* rearrangements, as these have only been recently available. Our case fulfills current diagnostic criteria for GCTB, and excludes other giant cell-rich lesions. Accurate diagnosis of GCTB in this atypical location allows for appropriate surveillance algorithms.

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## #7 Sulfamethoxazole-trimethoprim Induced Thrombocytopenia: A report of two cases and a literature review

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 35

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*Dr. Leigh Griffin (New York Presbyterian Queens), Dr. Jenna Marcinczyk (New York Presbyterian Queens), Dr. Paul Freedman (New York Presbyterian Queens), Dr. Renee Reich (New York Presbyterian Queens)*

Sulfamethoxazole- trimethoprim (ST) is a sulfonamide derivative antibiotic that interferes with bacterial folic acid synthesis and growth by blocking dihydrofolic acid formation from para-aminobenzoic acid. One of the more common and concerning side effects of sulfamethoxazole-trimethoprim is thrombocytopenia. Thrombocytopenia is a blood disorder characterized by a decreased number of circulating platelets. A normal platelet count is generally considered between 200,000-400,000 cells/mm<sup>3</sup>.

Spontaneous oral bleeding, purpura and petechiae of the oral mucosa may be seen when the platelet count drops below 100,000 cells/mm<sup>3</sup>. Here we present two cases of ST induced thrombocytopenia where the oral manifestations were the first indication of the low platelet count subsequently leading to life saving interventions. It is our aim to educate dentists about both the oral manifestations of thrombocytopenia and its association with ST as their examination may be instrumental in directing treatment.

Case 1 is a 69 year old male who was given ST 10 days post gynecomastia surgery and presented to an oral surgeon with the chief complaint of "blisters". Case 2 is a 75 year old female who presented to an oral surgeon with a chief complaint of "blood blisters" after having a difficult time swallowing her antibiotic pill the night before. Clinically both patients had intraoral purpura and petechiae. Both patients were admitted to the hospital with platelet counts less than 5,000 cells/mm<sup>3</sup>. Due to the very low platelet counts, both were given IV immunoglobulins and steroids after discontinuing ST. After several days they recovered and demonstrated a normal platelet count.

The best treatment for ST induced thrombocytopenia is discontinuation of the causative agent. Blood transfusions are not usually indicated due to continued platelet destruction in the presence of the drug. However, transfusions can be helpful in patients with a platelet count of <20,000 cells/mm<sup>3</sup> after discontinuation of the drug.

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## #8 Combined Adenomatoid Odontogenic Tumor and Calcifying Epithelial Odontogenic Tumor: A report of three cases and a review of the literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 37

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Odontogenic tumors present considerable histopathologic diversity. Lesions with combined features of more than one entity present a diagnostic challenge. One such lesion described in the literature is the combined lesion of adenomatoid odontogenic tumor (AOT) and calcifying epithelial odontogenic tumor (CEOT). Due to its rarity, there is limited data regarding its histopathologic features and clinical behaviors. Here, we present three cases of odontogenic tumors with features of both AOT and CEOT.

AOT usually occurs in female teenagers with predilection for the anterior maxilla and is often associated with an impacted tooth. In contrast, CEOT is often found in patients at the age of 30-50 years and has a predilection for the posterior mandible. CEOT is also often associated with impacted teeth. We searched the cases of Oral Pathology Lab at NYPQ from 1986-2021 and identified 63 AOTs, 24 CEOTs, and 3 AOT/CEOTs. All 3 AOT/CEOT cases were from females between ages 13-19 and were described as cystic lesions associated with impacted teeth in the anterior maxilla. Radiographic findings were described as mixed or radiolucent. The demographics and sites of our AOT/CEOT cases resemble those of AOT as opposed to CEOT.

The histology revealed odontogenic epithelial tumors composed of polygonal and spindle-shaped cells arranged in whorls, rosettes, and gland-like structures. Admixed throughout the tumors were sheets and strands of polyhedral epithelial cells with variably shaped nuclei set in abundant eosinophilic cytoplasm. Amorphous eosinophilic material and concentric calcifications were noted throughout. A Congo Red stain was positive in the one case it was performed. While both collision and hybrid theories have been suggested etiologies for AOT/CEOT, the admixture of the two components supports the latter. The similarity of our cases to the demographics of AOT, and the difference in sites and demographics of AOT and CEOT speaks against the collision theory.

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## #9 Two cases of oral discoid lupus erythematosus in previously undiagnosed patients

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 40

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Lupus erythematosus is a chronic inflammatory autoimmune disease with a wide range of clinical manifestations. The severity of disease presentation ranges from extensive systemic, multiorgan involvement to a more benign course limited to the skin. In the dental setting, undiagnosed patients may present with asymptomatic oral lesions that signify the first clinical manifestation of lupus erythematosus. While mucous membrane involvement without cutaneous lesions is rare, it should not eliminate lupus from the differential diagnosis. Here we describe two cases of oral discoid lupus erythematosus (DLE) with a previously unknown history.

Both patients were referred for evaluation of asymptomatic oral lesions. Patient 1 was a 50-year-old healthy female with a recurrent lesion of the left buccal mucosa and a second lesion of the right buccal mucosa. The lesion of the left buccal mucosa was initially diagnosed as epithelial dysplasia, but the recurrence and the contralateral lesion were subsequently diagnosed as interface mucositis suggestive of lupus erythematosus. Patient 2 was a 33-year-old African American female who presented with a painless, long standing ulceration with slightly elevated red and white borders, encompassing the length of her hard palate. Biopsy of the lesion revealed interface mucositis consistent with lupus.

Histologic exam of both biopsies revealed a lichenoid infiltrate, zones of epithelial hyperplasia and atrophy and a thickened PAS positive basement membrane below the epithelium and around blood vessels. MXA immunohistochemical stain, a marker highly associated with discoid lupus erythematosus, revealed positive staining reported as strong and focal respectively in the cases. Positive MXA immunostaining is supportive of the diagnosis of lupus. Early diagnosis of DLE, when oral mucous membrane involvement is the initial presenting feature, requires careful history and histopathologic correlation. Proper work up and consultation with a rheumatologist and/or dermatologist can help solidify the diagnosis and thus aid in prompt intervention and management.

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## #10 Marjolin's ulcer: A case report of a rare but preventable lesion

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 43

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*Dr. Mark Hochberg (Ohio State University College of Dentistry), Dr. Kristin McNamara (Ohio State University College of Dentistry),  
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Marjolin's ulcer (MU) is a rare cutaneous malignancy first described in 1828. MU arises in the setting of injured skin with subsequent chronic scarring or ulceration and eventual malignant transformation. While most cases of MU are associated with burn scars, less common settings have included traumatic wounds, venous stasis ulcers, osteomyelitis and pressure sores. MU may develop at any age but tends to affect older individuals. This is likely due to the typically long latency period from inciting event to malignant transformation, which averages 30 to 35 years. A male predilection exists. Lesions occur at any anatomic site, but the lower extremities and the head and neck region are most frequently affected. The predominant malignancy in MU is squamous cell carcinoma (80-90%) followed by cases of basal cell carcinoma and malignant melanoma. The treatment of choice is wide surgical resection. MU is typically an aggressive malignancy with a high recurrence rate and poor 5-year survival.

A 70 year-old woman from Togo had suffered a burn injury to the left side of her head many years previously. The wounded skin remained unchanged for decades, but recently developed an enlarging ulcer. Incisional biopsy of the ulcerated tissue resulted in a final diagnosis of keratinizing squamous cell carcinoma. Given the clinical setting, the lesion was deemed consistent with a MU.

MU is thought to be a largely preventable form of cutaneous malignancy. Proper wound management, including efforts to minimize infection and healing by secondary intention, substantially reduces the risk of malignant transformation. For this reason, MU has a propensity to affect disadvantaged individuals living in poverty who have limited access to proper or adequate healthcare.

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## #11 A SINGLE-CENTER CASE SERIES OF ORAL CAVITY SARCOMAS

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 51

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*Dr. Maram Bawazir (University of Florida), Dr. Mohammed N. Islam (University of Florida), Dr. Donald Cohen (University of Florida), Dr. Indraneel Bhattacharyya (University of Florida), Dr. Sarah Fitzpatrick (University of Florida)*

**INTRODUCTION:** Oral cavity sarcomas are rare and account for 1% of all malignancies in this site. This study aims to evaluate the prevalence, variety and distribution of soft tissue and gnathic sarcoma in the oral cavity.

**METHODS:** An IRB approved retrospective review encompassing years 2010-2020 of the University of Florida Oral Pathology Biopsy Service was performed. All histologically proven sarcomas occurring in the oral and maxillofacial area were retrieved. Demographics, clinical data, immunohistochemical analysis, and microscopic diagnoses were recorded and analyzed.

**RESULTS:** A total of 34 oral sarcomas were identified. The most prevalent were osteosarcoma (10 cases; 29.4%) followed by Kaposi sarcoma (9 cases; 26.4%) followed by undifferentiated sarcoma (3 cases; 8.8%). Three cases were unclassifiable due to lack of evidence of any specific lineage. Additional examples of sarcoma included; angiosarcoma, synovial sarcoma, low grade myxofibrosarcoma, rhabdomyosarcoma and chondrosarcoma. Molecular testing was performed on two cases. Age range was 11 to 82 (mean 42.35 years). Males were more affected than females (73.5% and 26.5% respectively). Central lesions accounted for 58.8% of the cases and 41.2 % were soft tissue sarcoma. All patients presented with primary tumors. The most commonly affected sites were mandible (13 cases; 65%), maxilla (11 cases; 32.4%), and tongue (4 cases; 11.8%). Malignant neoplasm was the most common reported clinical impression by submitting clinicians (18 cases; 52.9%). Lesion evolution time ranged from 3 weeks to one year. The most commonly reported symptoms with soft tissue tumors were localized pain, swelling, and bleeding. Most of the central lesions were radiolucent and caused cortical expansion.

**CONCLUSION:** Oral cavity sarcomas are rare neoplasms; however, they should be considered in the differential diagnosis whenever a large expansile, destructive, ill-defined or rapidly growing lesion is seen in a relatively young individual or paresthesia is reported.

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## #12 Three Cases of Central Xanthoma of the Jaws with Review of Literature.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 61

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*Dr. Shankar Venkat (University of Florida College of Dentistry), Dr. Donald Cohen (University of Florida), Dr. Indraneel Bhattacharyya (University of Florida), Dr. Mohammed N. Islam (University of Florida)*

**Background:** Extragnathic xanthomas are rare benign tumors associated with excess lipid production. Its gnathic counterpart was first reported in 1964. Only 45 cases of central xanthoma of jaw (CXJ) have been reported in the English literature. The etiopathogenesis of CXJ is highly suggestive of a reactive process or a metabolic condition. **Materials:** Three cases of CXJ were identified from the UF Oral Pathology Biopsy archives. Clinical, radiographic and histopathologic features of all the cases were retrospectively analyzed. Immunohistochemical (IHC) stains for S100 and CD68 were performed. **Case Series:** All cases of CXJ involved younger females and incidental lesions discovered during routine radiographic examination. The first patient, a 36-year-old female presented with an asymptomatic swelling and cortical expansion of the mandible adjacent to tooth #31. The second patient was a 13-year-old female with an asymptomatic expansion in the left mandibular ramus. Lastly, a 15-year-old female with dull pain exhibited mild buccal and lingual bony expansion of the right mandible apical to tooth #18. Radiographically, two of the cases presented as multilocular radiolucent lesions and one demonstrated as a well-circumscribed unilocular radiolucency intermixed with radiopacities. All three lesions were treated with surgical curettage and no recurrence at one-year follow up. All cases exhibited sheets of foamy macrophages similar to other lytic xanthomatous lesions such as non-ossifying fibroma, benign fibrous histiocytoma, Rosai-Dorfman disease, Erdheim disease, Gaucher disease, etc. A diagnosis of CXJ was established by exclusion and involved careful correlation of the clinical, radiographic, histopathological and IHC features. All three cases revealed diffuse positivity for CD68 and were negative for S100. **Conclusion:** CXJ are rare gnathic lesions of unknown etiology. We highlight three rare cases of CXJ, a lesion that may mimic other benign or reactive jaw lesions especially in young patients.

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## #13 Heterozygous Mutations in ANTXR2 Gene in a Child with Oral Manifestations of Hyaline Fibromatosis Syndrome: A Case Report and Review of Phenotype-Genotype Correlations

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 66

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*Dr. Sarah Franklin (School of Medicine at Hofstra-Northwell, Department of Dental Medicine, Division of Oral and Maxillofacial Pathology), Dr. Rachel Ruotolo (School of Medicine at Hofstra-Northwell, Department of Surgery), Dr. Kathleen Schultz (School of Medicine at Hofstra-Northwell, Department of Dental Medicine, Divisions of Oral and Maxillofacial Pathology and Pediatric Dentistry), Dr. John Fantasia (School of Medicine at Hofstra-Northwell, Department of Dental Medicine, Division of Oral and Maxillofacial Pathology)*

**Introduction:** Hyaline fibromatosis syndrome (HFS) is an autosomal recessive condition with hyaline deposits in the oral mucosa, skin, and other tissues, causing severe deformity and disability. HFS is caused by homozygous and compound heterozygous mutations in the *ANTXR2* gene. *ANTXR2* is ubiquitously expressed in all tissues except for the brain, and mutations in this gene are responsible for the complex clinical manifestations of this condition. Clinical hallmarks of the syndrome include progressive joint contractures, gingival hypertrophy, skin nodules, perianal masses, and hyperpigmentation over bony prominences.

**Case Findings:** A 19 month old female diagnosed with HFS, heterozygous for two novel pathogenic mutations in two separate alleles in *ANTXR2*, was evaluated for gingival hypertrophy. Additional clinical findings included subcutaneous nodularity of her facial skin, ears, lip commissures and with joint contractures. Rapid gingival overgrowth obscuring the primary dentition over five months, as well as involvement of her facial skin and perianal region necessitated multidisciplinary treatment to debulk the fibromatoses.

**Results:** A gingivectomy was performed to expose the patient's erupted primary dentition to facilitate oral mastication and potential for speech and language development. A preauricular nodule and perianal mass were excised. The histopathology of the gingival, preauricular, and perianal lesions revealed characteristic diffuse vascular hyaline collagen, typical for the syndrome.

**Conclusion:** Hyaline fibromatosis syndrome is rare and has significant oral manifestations. The histopathology of lesional tissues are quite characteristic. The novel mutations discovered in our patient and the clinical presentation support the phenotype-genotype correlations that have been previously suggested. The clinical phenotype coupled with the genotypic findings would classify this case as Grade 3 (severe) using a proposed four tier grading system for HFS. The differential diagnosis for progressive gingival overgrowth in children should include hyaline fibromatosis syndrome, as gingival hyperplasia and subcutaneous nodules may be the initial presentation in milder cases.

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## #14 Human T-Lymphotropic Virus 1 Related Adult T-Cell Lymphoma/Leukemia Presenting as a Lytic Lesion of the Mandible. The Importance of Serology in Accurately Defining the Entity.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 67

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**Introduction:** Adult T-cell lymphoma/leukemia (ATLL) is a rare T-cell neoplasm pathogenetically caused by human T-lymphotropic virus 1 (HTLV-1). HTLV-1 is the first human retrovirus shown to cause malignant transformation. ATLL is endemic in several regions of the world, most notably Japan, the Caribbean, parts of Africa and Iran. The disease has a long latency period, as individuals are infected by the virus early in life. Most cases of ATLL are diagnosed in adults with a male-to-female ratio of 1.5:1. ATLL most commonly affects the peripheral blood and skin. Oral involvement is extremely rare.

**Case Findings:** A 61-year-old female presented for evaluation of a large radiolucency involving the right posterior mandible with associated mobile teeth and pain. CT revealed an expansile lytic lesion extending from first premolar to the mandibular angle. Notably, there were two additional destructive lesions identified in the right sinonasal and tympanomastoid areas.

**Results:** The histopathology of the mandibular mass was that of a lymphoproliferative process. Lesional cells expressed CD25, CD2, CD3, CD4, CD5 and CD43, and did not express CD7, CD20, or PAX5. The Ki-67 proliferative index was 90%. A diagnosis of peripheral T-cell lymphoma was rendered. Serological testing with Western Blot was positive for HTLV-1, further supporting the diagnosis of ATLL.

**Conclusion:** Four clinical variants of ATLL are identified: acute, lymphomatous, chronic, and smoldering. Acute and lymphomatous are highly aggressive, whereas chronic and smoldering have a prolonged clinical course. Prognosis for ATLL is poor as it is resistant to conventional chemotherapy, with the acute and lymphomatous variants having a median survival of less than 1 year and the chronic and smoldering variants having a median survival of less than 5 years. The diagnosis of ATLL relies on immunohistochemistry supplemented with serology for HTLV-1. Lymphoproliferative processes with this immunoprofile require HTLV-1 serology for definitive classification.

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# #15 Monoclonal Anti PD-1 Antibody (Pembrolizumab) Induced Granulomatous/Sarcoid-like Lesions in a Patient with Nasal Mucosal Melanoma: an Evolving Phenomenon of Targeted Therapies and Expanding the Differential Diagnosis for Granulomatous Inflammation

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 68

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*Dr. Laurel Henderson (Zucker School of Medicine at Hofstra-Northwell, Department of Dental Medicine), Dr. Steven Savona (Zucker School of Medicine at Hofstra-Northwell, Department of Medicine), Dr. Peter Farmer (Zucker School of Medicine at Hofstra-Northwell, Department of Pathology and Laboratory Medicine), Dr. John Fantasia (School of Medicine at Hofstra-Northwell, Department of Dental Medicine, Division of Oral and Maxillofacial Pathology)*

**Introduction:** Immune checkpoint inhibitors and kinase inhibitors are now part of the treatment regimen of many cancers. Granulomatous/sarcoid-like lesions (GSL) involving the skin, lungs, lymph nodes, and other organs and tissues in patients treated with these medications, including those with mucosal melanoma, are an emerging phenomenon. GSL can be clinically and radiographically concerning for metastatic disease.

**Case Findings:** A 61-year-old female was diagnosed with primary melanoma involving the left nasal mucosa. Initial PET was negative for metastasis. The patient underwent medial maxillectomy with negative margins. Adjuvant therapy consisted of pembrolizumab 200 mg infusion every three weeks.

**Results:** Active surveillance included a subsequent PET scan with increased SUV localizing to the lungs, mediastinal and hilar lymph nodes, and a bony lesion of the right hip. FNA of the hip lesion demonstrated defined non-caseating granulomas. Histochemical and immunohistochemical stains were negative for microorganisms, and melanoma. Lung, mediastinal and hilar lesions were presumed similar. Steroid therapy resulted in regression of the GSL.

**Conclusions:** This case of nasal mucosal melanoma treated with pembrolizumab highlights the phenomenon of therapy related GSL. These are typically identified during routine surveillance and often mistaken for disease progression. Documentation of these lesions will avoid misdiagnosis of disease progression and influence treatment strategies for such lesions and continuance of therapy, if appropriate. This may represent a positive prognostic indicator. The mechanism of granuloma formation in this setting is speculative and includes, cytokine imbalance promoting granuloma formation characterized by either a hypoactive or hyperactive immune response or unmasking of subclinical sarcoidosis. Pathologists must be aware of this phenomenon occurring in patients receiving various targeted therapeutics for cancerous and some non-cancerous diseases. Thus, medication related granulomatous inflammation is an additional entity in the differential diagnosis of microscopically identified granulomatous inflammation.

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## #16 Hyperkeratosis with Epithelial Atrophy is Common in Proliferative Leukoplakia Biopsies: a Multicenter Study

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 73

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*Dr. Lama Alabdulaaly (Harvard School of Dental Medicine), Dr. Alessandro Villa (University of California San Francisco), Dr. Tiffany Chen (Brigham and Women's Hospital), Dr. Alexander Kerr (New York University College of Dentistry), Dr. Fabio Alves (AC Camargo Cancer Center), Dr. Sook-Bin Woo (Harvard School of Dental Medicine)*

**Introduction:** The aim of this multicenter retrospective study is to characterize the histopathologic features of initial and early biopsies of proliferative leukoplakia (PL), and to analyze the correlation between histopathologic features and malignant transformation (MT) and mortality.

**Materials and methods:** Patients with a clinical diagnosis of PL who had at least one biopsy and one follow up visit were included in this study. Initial (first ever biopsy for the condition) and early (first biopsy when patient was seen by one of the authors) biopsy specimens were retrieved and reviewed. Pearson's chi-square test was used.

**Results:** There were 86 early/initial biopsies from 59 patients; 74.6% were females and median age was 66 years. Most of the cases had a smooth/homogenous (39.0%) or fissured appearance (29.3%), and only 12.2% had a verrucous appearance. The most common biopsy site was the gingiva/alveolar mucosa (40.8%). The most common histologic diagnosis was oral epithelial dysplasia (OED; 53.5%) followed by hyperkeratosis/parakeratosis that was not reactive (HkNR; 31.4%), which did not show obvious OED. Of note, two-thirds of the HkNR cases showed hyperkeratosis and epithelial atrophy. A lymphocytic band was seen in 66.7% of OED cases and 33.3% of HkNR cases, mostly associated with epithelial atrophy. Thirty-four patients (57.6%) underwent MT for a total of 51 carcinomas; only 33.7% of initial/early biopsy sites showed MT at the same site. The mortality rate was 11.9%, and patients with OED and HkNR at initial presentation had mortality rates of 19.6% and 14.8%, respectively ( $p > 0.05$ ).

**Conclusions:** Our findings show that approximately one-third of PL cases exhibit only HkNR, often with epithelial atrophy but undergo MT, regardless suggesting that HkNR is the precursor lesion to OED and then to invasive cancer.

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## #17 Hypopigmented variant of intraoral common blue nevus. A rare entity.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 79

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*Dr. Efstathios Pettas (Department of Oral Medicine and Pathology, School of Dentistry, National and Kapodistrian University of Athens (NKUA), Athens, Greece), Dr. Maria Georgaki (Department of Oral Medicine and Pathology, School of Dentistry, National and Kapodistrian University of Athens (NKUA), Athens, Greece), Ms. Nikoleta Papageorgiou (School of Dentistry, National and Kapodistrian University of Athens (NKUA), Athens, Greece), Dr. Patroklos Katafygiotis (Department of Pathology, School of Medicine, National and Kapodistrian University of Athens (NKUA), Athens, Greece), Prof. Nikolaos Nikitakis (Department of Oral Medicine and Pathology, School of Dentistry, National and Kapodistrian University of Athens (NKUA), Athens, Greece)*

**Objectives:** Blue nevus (BN) represents a clinicopathologically distinctive subset of benign melanocytic proliferations that may affect the oral mucosa. While diagnosis of conventional BN is usually straightforward, some of its rare variants may cause significant overlap with other entities, causing diagnostic pitfalls. Herein we present a case of hypopigmented variant of common BN and discuss the diagnostic challenges.

**Findings:** A 52 year-old male of unremarkable medical history was referred for the evaluation of a painless palatal lesion of unknown duration. The clinical examination revealed a well-circumscribed, slightly elevated lesion of normal hue with bluish halo involving the hard palate. With a provisional diagnosis of BN an excisional biopsy was decided. Histopathologically, a proliferation composed of scattered elongated, finely branching melanocytes, located deeply within the lamina propria with non-infiltrating pattern and associated with a variably desmoplastic stroma, was observed. Cytoplasmic pigmentation was detected only in a small subset of dendritic processes after serial sectioning. Minimal cytologic atypia and mild nuclear pleomorphism were also present in the absence of mitotic activity. In order to rule out malignant melanocytic tumors, especially desmoplastic melanoma, extensive immunohistochemical examination was performed. Melanocytes were strongly and diffusely positive for Melan-A, HMB-45, SOX-10, S-100, p16, and vimentin. The proliferation index Ki-67 was <5%. A final diagnosis of hypopigmented common BN was rendered. No signs of recurrence were noted during follow-up.

**Conclusions:** To this date, few cases of BN with minimal or absent pigmentation have been reported in the literature. However, this pattern is extremely rare in lesions affecting the oral mucosa. Hypopigmented BN lesions require careful evaluation to be distinguished primarily from desmoplastic melanoma. Immunohistochemistry may be helpful, especially when tumors with atypia, pleomorphism, infiltrative pattern and/or mitoses are encountered.

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## #18 ECTOMESENCHYMAL CHONDROMYXOID TUMOR WITH HIGH-GRADE TRANSFORMATION

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Tuesday, 25th May - 08:00: Poster Presentations - Oral to Poster - Abstract ID: 64

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*Dr. Rana Alshagroud (King Saud University), Dr. Osama Alghamdi (King Saud University), Dr. Brendan Dickson (Mount Sinai Hospital, Ontario), Dr. Ioannis G. Koutlas (University of Minnesota)*

**Introduction:** Ectomesenchymal chondromyxoid tumor (ECT) is a rare neoplasm that has traditionally been considered limited to the tongue and thought to follow a benign clinical course. Recently these tumors have been found to be characterized by a recurrent fusion gene (*RREB1-MRTFB*[formerly*MKL2*]). This same product has been observed in a subset of mediastinal mesenchymal tumors and a single case of a biphenotypic oropharyngeal sarcoma, raising the possibility of a shared pathogenesis. Herein we report a case of ECT, that was confirmed to harbor the *RREB1-MRTFB* fusion product, which was found to show progression to a poorly differentiated malignancy.

**Materials and Methods:** An otherwise healthy 5-year-old girl presented with an asymptomatic mass of unknown duration, which was situated in the anterior maxillary vestibule extending to the lip. Axial MRI showed a hyperintense and heterogeneous mass with peripheral enhancement.

**Results:** An incisional biopsy was performed showing a tumor with morphologic and immunophenotypic features consistent with ECT. RNA sequencing was performed which confirmed the presence of a *RREB1-MRTFB* fusion product. The patient was subsequently referred for surgical resection. In addition to showing areas of conventional ECT this also revealed transition to highly cellular neoplasm characterized by sheets and nests of round-rhabdoid cells with brisk mitotic activity, bone involvement and lymphovascular invasion. Immunohistochemistry on this component was similar, suggesting high-grade progression rather than dedifferentiation.

**Conclusions:** This case adds to the recent literature expanding the anatomic distribution and morphology of ECT. More importantly, this is the first molecularly confirmed report showing progression from a conventional ECT to a high-grade neoplasm. It remains unclear whether overt dedifferentiation is likewise possible. This case suggests complete excision of ECT, to evaluate these tumors in their entity and exclude a high-grade component, is warranted. More detailed molecular studies, including management of tumors with malignant histologic attributes, are necessary.

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## #19 Assessment of Bone and Soft Tissue Margins in Resection of Mandibular Ameloblastoma: An Institutional Series

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Tuesday, 25th May - 08:00: Poster Presentations - Oral to Poster - Abstract ID: 84

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*Dr. Kelly Magliocca (Emory University), Dr. Anne McLean (Emory University), Dr. Faisal Saeed (Emory University), Dr. Zaid Mahdi (Emory University), Dr. Qiuying Shi (Emory University), Dr. Daniel Lubin (Emory University)*

**Introduction:** The treatment of gnathic ameloblastoma remains controversial. When resection is undertaken, a planned linear bone margin of at least 1.0 cm is often recommended for mandibular tumors. For cases in which tumor has extended beyond bone, inclusion of the adjacent anatomic barrier is recommended, however little attention has been devoted to the margin reporting of non-osseous margins.

**Materials and Methods:** Emory University records were searched for patients undergoing resection for ameloblastoma between 1/1/2020 and 3/01/2021. Twenty-one consecutive patients underwent surgical resection during the fourteen-month interval. In six cases, specimen fragmentation precluded margin evaluation. In three cases, only rare tumor islands remained microscopically in the resection specimen after initial biopsy inadvertently removed much of the lesion. The twelve remaining patients formed the study cohort.

**Results:** Patients ranged from 22 to 78 years of age, and 6 were male. Segmental mandibular resection was performed in 10 cases and marginal mandibular resection accomplished in 2. Average tumor size was 4.9 cm. In 10 of 12 cases, ameloblastoma extended beyond the mandibular bone. In all ten cases, the soft tissue margin was grossly identified as the margin nearest to tumor. Microscopically, soft tissue margin clearance measured 1.0 mm or less and in one, tumor involved the soft tissue margin. All bone margins measured 0.6 cm or greater from tumor, with 10 of 12 mandibular bone resection margins showing a linear distance of 1.0 cm or more from tumor.

**Conclusion:** In ameloblastoma perforating bone, soft tissue margins are most likely to represent the closest margin. Narrow clearance of soft tissue margins in ameloblastoma may be related to the lack of a specific guiding measurement in the literature. Close soft tissue margins can be identified grossly, and the clinical significance of ample bone margin clearance in the setting of narrow soft tissue clearance remains uncertain.

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## #20 Thirteen Synchronous Multifocal Calcifying Epithelial Odontogenic Tumors (CEOT): Case Report and Review of the Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 5

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*Dr. Hiba Qari (West Virginia University School of Dentistry), Dr. Ryan McCloy (West Virginia University School of Dentistry), Dr. Patrick Bacaj (West Virginia University School of Medicine), Dr. Jerry Bouquot (West Virginia University School of Dentistry)*

**Introduction:** Calcifying epithelial odontogenic tumor (CEOT, Pindborg tumor) is a rare benign, locally aggressive neoplasm of the jaws that accounts for approximately 1% of all odontogenic tumors. The CEOT was first defined by Pindborg in 1955 and has been reported approximately 350 times in the literature; 7 of these reported multiple, synchronous lesions (up to 4). We report the eighth case in an individual with the largest number (by far) of CEOTs reported to date and provide a literature review of multifocal CEOT cases. **Materials and methods:** A 30-year-old male presented to the Oral and Maxillofacial Surgery Department of the West Virginia University School of Dentistry (WVU SoD) to extract multiple impacted teeth previous to constructing a complete denture. **Results:** A pantograph showed 15 impacted teeth, almost all associated with well-demarcated cyst-like radiolucencies, some with small, ill-defined radiopaque flecks. Microscopically the lesions showed sheets of strands of polygonal epithelial cells with eosinophilic cytoplasm. Spread throughout the epithelium and connective tissue were small spherical amorphous pale purplish calcifications. Each lesion was similar and consistent with a diagnosis of CEOT. **Conclusion:** We report a patient with 13 independent CEOTs scattered throughout all quadrants, suggesting genetic mutation of a tumor suppressor gene, such as the PTCH1 gene, which has been previously identified in patients with solitary CEOTs. This is the largest number of Pindborg tumors or any other type of odontogenic tumors, yet reported in a single individual.

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## #21 Previously Unreported Salivary Heterotopia of the TMJ – 23 Serous Choristomas of One Joint.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 9

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*Dr. Jerry Bouquot (West Virginia University School of Dentistry), Dr. Steven Whitaker (West Virginia University School of Dentistry), Dr. Hiba Qari (West Virginia University School of Dentistry), Dr. Scott Bolding (Private Practice of Oral Surgery, Fayetteville, Arkansas)*

**Introduction:** Salivary gland choristomas of the soft tissues of the TMJs, independent of adjacent parotid glands, have not been previously reported. **Objective:** To report a massive example of serous gland heterotopia of the TMJ soft tissues. **Methods and Materials.** 116 consecutive TMJ surgical samples from 57 subjects were microscopically reviewed for abnormalities. Of these, 27 joints contained previously undescribed heterotopic glandular lobules of parotid-like tissue (plus 2 mucus lobules), with no connection to parotid glands. The case of a 41 year of female with many years of joint dysfunction and severe pain was selected for specific description because she presented the most extreme example of this phenomenon. **Results:** Two clusters comprising a total of 57 distinct lobules of mostly normal serous acinar tissue and ductal structures were embedded in or attached to the posterior-lateral ligament of the subject's left TMJ. The ligament itself was largely replaced by dense fibrous scar, with an embedded traumatic neuroma and an hypertrophied artery, while fossa curettings showed osteoarthritis. The salivary lobules appeared to represent 23 different "glands" measuring altogether 2.5 x 2.5 x 0.5 cm. Glandular parenchyma was histologically normal and mature serous tissue, except for 6 glands which showed almost complete oncocytic change, 7 showing focal regions of oncocytic change, and 2 showing acinar atrophy with stromal fibrosis and small numbers of lymphocytes (chronic sialadenitis). No large or secretory ducts were found. Serial sectioning confirmed that there was no connection to normal parotid gland. **Conclusion:** We report a massive cluster of 23 serous gland choristomas within and attached to the posterolateral ligament of the left TMJ in a single individual. The ligament was severely scarred and some choristomas showed chronic sialadenitis, presumably from being repeatedly pressed into the joint spaces. Heterotopic serous tissue has not been previously reported from the TMJs.

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## #22 Central ossifying fibroma of mandible arising in association with cemento-osseous dysplasia. An unusual case presentation

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 22

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*Dr. Rafik Abdelsayed (Augusta University, Dental College of Georgia), Dr. Allison Buchanan (Augusta University, Dental College of Georgia)*

Introduction: Cemento-osseous dysplasia (COD) is known to develop complications including infection, necrosis, and traumatic bone cysts [1]. Neoplastic occurrence with COD has been reported, but is rare [2,3]. We report an unusual case of central ossifying fibroma (COF) of the mandibular symphysis in association with COD in a 46-year-old black female. Case Description: The patient presented with a mandibular symphysis mass that was associated with difficulty of eating, speaking and paresthesia. The radiographic images showed a predominantly radiopaque mass in the symphysis region, measuring 7 x 6 x 5cm. There were discrete, minimally expansile mixed radiolucent- opaque lesions in the posterior mandible and maxilla. The patient underwent mandibular resection due to the symptoms of eating and speaking difficulties as well as paresthesia. Before resection, three incisional biopsies including bilateral posterior regions of mandible and the central mass. Histopathologic examination of posterior specimens revealed similar findings of fragments consisting of cellular fibroblastic tissue intermixed with short bone spicules and cementum-like and hemorrhagic deposits associated with traumatic bone cysts. However, the central specimen showed solid tumor consisting of fibroblastic proliferation supporting spindled fibroblast-like cells. It supported cellular bone trabeculae as well as spherical cementum-like calcified deposits. This framework is interspersed with spaces lined by fibrovascular tissue supporting eosinophilic osteoid, consistent with traumatic bone cysts. Discussion: COD lacks genetic abnormalities noted in fibrous dysplasia or neoplastic fibro-osseous lesions. We present an unusual association of COF with florid COD. This association has been previously reported in a patient who developed COF with COD and odontoma, and in another who developed peripheral osteoma, odontoma, COF and COD [2,3]. To the best of our knowledge, this presentation describes only the third case report of the association of COF and COD. The co-existence of COF and COD in these cases is unclear, but seems to be very rare.

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## #23 Solitary Gingival Lymphangioma: Case Series and Review of Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 23

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*Dr. Moni Ahmadian (University of Nevada, Las Vegas School of Dental Medicine), Dr. Rachele Wolk (New York Presbyterian Queens), Dr. Paul Freedman (New York Presbyterian Queens), Dr. Alexis Russell (University of Nevada, Las Vegas School of Dental Medicine)*

Lymphangiomas are benign malformations of lymphatic vessels. These anomalies likely arise from proliferation of lymphatic channels that do not communicate normally with the rest of the lymphatic system. Lymphangiomas show a marked predilection for the head and neck region with nearly 90% of cases clinically evident at the time of birth or during the first two years of life. Oral lymphangiomas are less common. The most common intraoral location is the anterior two thirds of the tongue followed by buccal mucosa. Extremely rare cases of solitary gingival lymphangiomas have been reported in the English language literature. Here we report an additional twenty cases of these rare solitary gingival lymphangiomas. Our cases demonstrate a 2:1 female-to-male ratio with a predilection for the first two decades of life. These cases show a nearly equal distribution between the maxillary and mandibular gingiva with a notable predisposition for the anterior gingival tissue presenting as hyperplastic, pebbly, and vesicular lesions. Microscopically, all the cases exhibit superficial thin, endothelial-lined lymphatic channels containing a luminal proteinaceous material. Immunohistochemical staining for D2-40 (podoplanin) was available for one of our cases and demonstrates cytoplasmic immunoreactivity of endothelial cells. These findings suggest gingival lymphangiomas, while uncommon, should be included in the differential diagnosis of the vesicular lesions of the gingiva.

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## #24 Papillon-Lefèvre Syndrome: Report of Two Cases in Siblings

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 34

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*Dr. Faraj Alotaiby (College of Dentistry, Qassim University), Dr. Thiyezen AlDhelai (College of Dentistry, Qassim University), Mr. Ibrahim Alblihed (College of Dentistry, Qassim University), Dr. Indraneel Bhattacharyya (University of Florida)*

**Introduction:** Papillon-Lefèvre syndrome (PLS) is an unusual health condition manifested by pre-pubertal extensive periodontal disease that results in early tooth loss and palmoplantar keratosis. The etiology of PLS is loss-of-function mutations in the cathepsin C gene. This gene is a lysosomal protease that plays an essential role in immune and inflammatory responses. **Case Presentation:** We present two cases in siblings with PLS. The parents of the patients are relative. The first patient is 20 years old who presented to the clinic with highly resorbed maxillary and mandibular alveolar ridges with only wisdom teeth remaining in the oral cavity at the time of examination. According to the dental history, there was aggressive periodontitis followed by complete destruction of the periodontium and severe bone resorption. There was skin manifestations of PLS including palmoplantar and dorsal keratosis in their extremities as well as the knee and wrist joint regions. The second sibling was 9 years old with only skin manifestations similar to the older sibling. The periodontal status of the younger patient was stable at the time of clinical examination. Interestingly, they reported that their 16-year-old female cousin also suffers from the same condition, with almost the same clinical manifestations as the older sibling. **Conclusion:** We report two cases of PLS in patients of same family, which supports the hereditary nature of cathepsin C mutation in PLS patients. In addition, variability of clinical expression of PLS is very evident in the current cases, which reflects that the PLS can show a broad spectrum of clinical features.

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## #25 Metastatic Sarcomatous Tumor to Mandible Identifies Pleuropulmonary Blastoma Tumor Predisposition Syndrome

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 47

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*Dr. John Hicks (Texas Children's Hospital, Baylor College of Medicine), Dr. Catherine Flaitz (University of Colorado School of Dental Medicine)*

**Introduction:** Pleuropulmonary blastoma (PPB) affects infants and young children and may occur as a cystic (Type I), partially cystic-partially solid (Type 2), or solid (Type 3) tumor. PPB is an embryonal tumor derived from lung and pleural mesenchyme. This malignant tumor is composed of sarcomatous elements, most commonly a mixture of spindle cell sarcoma with focal rhabdomyosarcomatous elements. However, it may have a wide variety of sarcomatous components. The tumor is characterized by DICER1 mutation, which may be somatic (tumoral) or constitutional. Constitutional DICER1 mutation indicates a predisposition for the development of other tumors and may have implications for the child and other family members. **Clinical Presentation and Pathology Findings:** This 18-month-old male presented with a rapidly enlarging, expansile left mandibular mass. A biopsy was performed which showed a predominantly spindle cell sarcoma (vimentin-positive) with focal areas of readily identified rhabdomyoblasts (positive for desmin; negative for myogenin and MyoD1). Molecular genetic testing of the tumor identified a DICER1 mutation and was negative for rearrangements associated with rhabdomyosarcoma (FOXO1, FUS-TFCP2, EWSR1-TFCP2, NCOA2, VGLL2). Radiologic workup identified a predominantly solid pulmonary mass. Lobectomy was performed and diagnosis of PPB was confirmed. Sarcoma-based oncologic management was carried out. Subsequent genetic testing identified DICER1 constitutional mutation. **Conclusion:** PPB presenting as a metastatic disease is not unusual, with the involvement of various sites and organs (most common sites: brain, bone). Although exceedingly rare, PPB should be considered in an infant or young child when encountering a possible metastatic sarcomatous tumor and prompts radiologic evaluation for a possible primary lung tumor. Molecular genetics plays an essential role in identifying risk for PPB Tumor Predisposition Syndrome. Oncologic management is sarcoma-based and children with isolated lung tumors have variable outcomes (Type I - 85%, Type 2 - 60%, Type 3 - 40%).

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## #26 Genotyping An Autoimmune-Associated SIRPγ SNP From Archived Oral Biopsy FFPE Tissues

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 72

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*Dr. Scott Steward-Tharp (The University of Iowa), Dr. John Hellstein (The University of Iowa), Dr. Nitin Karandikar (The University of Iowa)*

Introduction: T-cells play critical roles in many autoimmune conditions, such as type I diabetes mellitus (T1D). They are also thought to play a significant role in many immune-mediated disorders affecting the oral soft tissues: lichen planus, graft vs. host disease, Sjogren syndrome. T-cells exert their functions via direct recognition through molecules expressed on the cell surface and secretion of factors that drive or suppress inflammatory responses locally. Signal regulatory protein gamma (SIRPγ) is an immunomodulatory protein that is uniquely expressed on the cell surface of human T-cells. Our laboratory has shown that a T1D-linked genetic variant of the gene for SIRPγ (rs2281808) correlates with reduced expression of SIRPγ on the T-cell surface and that T-cells with less SIRPγ surface expression produce more inflammatory molecules. We hypothesize that single nucleotide changes within the gene for SIRPγ can lead to a predisposition for the development of immune-mediated pathosis. Our objective for this study was to assess the feasibility of genotyping individuals at single nucleotide polymorphism (SNP) rs2281808 from genomic DNA extracted from formalin-fixed, paraffin-embedded (FFPE) tissue blocks.

Materials and Methods: Archived, de-identified formalin-fixed paraffin-embedded tissue blocks from oral soft tissue biopsies were used in this study. The tissue blocks ranged from 1-18 years of age. Three 5-mm thick sections per sample were deparaffinized with subsequent genomic DNA extraction using the QIAamp® DNA FFPE Tissue Kit and Deparaffinization Solution. DNA was quantitated using UV spectroscopy at 260 nm. Allelic discrimination PCR for rs2281808 genotyping was done using a TaqMan® assay and probes.

Results: Allelic discrimination PCR for rs2281808 was successful in ~77% (46/60) of specimens tested.

Conclusions: This pilot study supports the feasibility of our methodology, however, further studies are necessary to assess the accuracy of the genotyping results obtained via this method.

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## #27 Metastatic urothelial carcinoma to the mandible

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 75

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*Dr. Victoria Woo (TAMU College of Dentistry), Dr. Paras Patel (TAMU College of Dentistry), Dr. Gregory Taylor (Southlake Oral Surgery), Dr. Scott McCord (Texas Oncology)*

Urothelial carcinoma (UC) is the second most common malignancy of the genitourinary tract and sixth most common malignancy in the US. It predominately affects patients in the seventh decade with a male predilection. Presenting symptoms may include hematuria, infection, and urinary obstruction. First-line treatment of invasive UC consists of surgical resection with platinum-based chemotherapy. Promising roles for immunotherapy and targeted therapy have also emerged. We herein describe a 68-year-old female who presented for evaluation of long-standing pain involving her anterior mandible. Radiographs revealed an ill-defined radiolucency in the region of #22 to #27. Histologic examination of a biopsy taken from the #23 periapex showed sheets and nests of atypical epithelial cells with abundant eosinophilic to vacuolated cytoplasm and central nuclei. In several areas, the cells were markedly discohesive and exhibited prominent hobnailed configurations. On immunohistochemical analysis, the neoplastic population was reactive for AE1/3, CK7, and CK20. Follow-up discussion with the patient's physician revealed a history of invasive high-grade urothelial carcinoma diagnosed one year prior and treated with neoadjuvant chemotherapy followed by cystectomy. The prior specimens were reviewed and showed morphologic similarities to the mandibular biopsy, supporting a diagnosis of metastatic urothelial carcinoma in the latter. The patient subsequently underwent second- and third-line treatment but has continued to show progression of her disease. Metastasis to the oral cavity is an uncommon occurrence that accounts for only 1% of all oral malignancies. The majority of cases occur in the setting of disseminated disease with a known primary, although a small percentage of oral metastases represent the initial manifestation of occult disease. Metastasis of UC to the oral cavity is exceedingly rare with less than 20 cases reported in the literature. However, knowledge of the morphological and immunohistochemical hallmarks of this malignancy can aid in directing the diagnostic workup, thereby facilitating timely management.

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## #28 Hematolymphoid Tumors of the Oral and Maxillofacial Region: A clinicopathologic study of 41 cases.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 81

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*Dr. Darren P. Cox (University of Pacific Arthur A. Dugoni School of Dentistry), Dr. Leticia Ferreira (University of Pacific Arthur A. Dugoni School of Dentistry)*

**Introduction:** Oral hematolymphoid tumors represent a heterogeneous and complex group of neoplasms of lymphoid, plasma cell, histiocytic/dendritic, and myeloid origin. Our aim was to determine the frequency, classification, and clinicopathologic characteristics of all hematolymphoid tumors diagnosed at the Pacific Oral and Maxillofacial Pathology Laboratory (POMPL) in the past seven years.

**Materials and Methods:** Forty-one cases of hematolymphoid tumors were retrieved from the files of the POMPL and the data were analyzed with regard to clinical, histopathologic, and immunophenotypic features.

**Results:** Hematolymphoid tumors represented ~0.1% of the total number of accessioned cases since Jan 2014. Male:Female ratio was 1.1:1. Anatomic distribution: 23 were intraosseous (15 in tooth-bearing areas and two in maxillary sinus) and 18 originated in soft tissue (10 palatal mucosa; 6 gingival; 1 buccal mucosa; 1 upper lip). The age range was 35 to 83 years (mean: 59.2 years; median: 61 years). Diagnoses: 26 were B-cell lymphomas (15 diffuse large B-cell lymphoma, 5 low-grade B-cell lymphoma, 5 follicular lymphoma, and 1 marginal zone lymphoma (MALT lymphoma)); 8 plasmacytoma/myelomas; 2 T-cell lymphomas; 2 Langerhans cell histiocytosis, 2 myeloid sarcomas, and 1 atypical hematolymphoid proliferation.

**Conclusions:** Male and female patients are equally affected by hematolymphoid tumors in general and the most common type to involve the oral cavity is the diffuse large B cell lymphoma. Hematolymphoid tumors show a predilection for the tooth-bearing areas of the jaws and the palatal mucosa and typically appear as destructive bony lesions or red soft tissue swellings.

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## #29 Tubuloductal/syringoid variant of central odontogenic fibroma with amyloid?

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 85

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*Dr. Ioannis G. Koutlas (University of Minnesota), Dr. Katia Julissa Ponce (University of Montreal), Dr. Rima-Marie Wazen (University of Montreal), Dr. Antonio Nanci (University of Montreal)*

### **Introduction:**

Glandular tumors of jaw bones present, most often, histopathologic features of salivary gland and, rarely, of cutaneous glandular neoplasms and are thought to predominantly originate from odontogenic epithelium.

### **Materials and Methods:**

An unusual maxillary tumor in a 74-year-old male is presented exhibiting radiolucency and root resorption in the apical area of teeth #6-7.

### **Results:**

Preparations revealed occasionally branching tubular cords and ductal structures characterized, mostly, by a bi-layer composed of luminal Ber-EP4, cytokeratin (CK) 7 and occasionally CK8/18 positive cells and varying from cuboidal to low columnar, and abluminal, CK5/6 positive, basal/basaloid cells revealing nuclear reactivity for p63/p40. Smooth muscle actin and calponin were negative, save for a single focus for calponin, confirming absence of myoepithelial support or epithelial mesenchymal transition. CK19 exhibited staining of both layers, the luminal being more intense. Eosinophilic secretory material and, occasionally, a luminal pellicle were decorated with CK8/18 and polyclonal carcinoembryonic antigen (CEA). CD1a identified only rare Langerhans' cells and Ki67 decorated 1-2% of abluminal cell nuclei. Small solid nests of epithelial cells were also present. Infrequently, an apparent transition of a nest into a tubular structure was appreciated. The partially inflamed stroma featured multiple hyalinized acellular deposits consistent with amyloid, as confirmed by bright orange Congo red reactivity with apple-green birefringence, which reacted with antibodies against odontogenic ameloblast-associated (ODAM) protein but not amelotin and secretory calcium-binding phosphoprotein proline-glutamine rich 1.

**Conclusions:** Based on the above the diagnosis of tubuloductal/syringoid variant of central odontogenic fibroma with ODAM amyloid is favored.

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## #30 CEMENTO-OSSEOUS DYSPLASIA ASSOCIATED WITH AN IMPACTED CANINE INFERIOR TO A COMPOUND ODONTOMA

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 17

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*Dr. Mark Mintline (Western University College of Dental Medicine), Dr. Hanaa Selim Gobriel (Western University College of Dental Medicine), Dr. Setareh Lavasani (Western University College of Dental Medicine), Dr. Jeffrey Elo (Western University College of Dental Medicine), Dr. Shahrzad Aarup (Western University College of Dental Medicine), Dr. Gretchen Folk (Scripps Oral Pathology Service)*

We present a case of a 48-year-old Hispanic female with a fibro-osseous lesion consistent with cemento-osseous dysplasia (COD) that was located on the periapical aspect of an impacted mandibular canine inferior to a compound odontoma. COD is a non-neoplastic fibro-osseous lesion of the tooth bearing regions of the jaws. COD is traditionally divided into three clinical variants: periapical, focal, and florid. COD is associated with vital teeth and may be seen in edentulous areas. However, cemento-osseous dysplasia associated with the periapical region of an impacted tooth and adjacent to an odontoma is rare. We believe this is the first reported case of COD around the periapical aspect of an impacted tooth. The radiographic, surgical, and microscopic findings suggest that the compound odontoma and COD developed coincidentally as distinct lesions. But a common developmental origin for the odontoma and COD cannot be ruled out due to the proximity of the lesions. No recurrence has been recorded after one year. Additional case reports of COD arising adjacent to or in association with odontogenic tumors are needed.

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## #31 MULTIDISCIPLINARY MANAGEMENT OF DIFFUSE LARGE B-CELL LYMPHOMA OF THE MANDIBLE

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 21

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We present a case of a 25-year-old Hispanic female with a diffuse large B-cell lymphoma of the mandible. The case documents the patient's complete cancer treatment and the role of the dental team as part of a larger oncology team to help prevent and manage oral complications. The dental team collaborated with the medical and radiation oncology teams to screen, diagnose, stage, treat, and manage the patient. Prior to cancer treatment, the dental team completed an oral health examination and established a schedule of dental treatment. Treatment consisted of patient education, oral hygiene instruction, dental prophylaxis, elimination of acute oral infections, fabrication of custom gel-applicator trays, fluoride gel prescription, and fabrication of a customized tongue-displacing (CTD) stent with a novel design. We believe it is the first reported use of a CTD for a gnathic non-Hodgkin lymphoma. During and after cancer treatment, the dental team managed complications of chemotherapy and radiation therapy including dysgeusia, xerostomia, oral mucositis, and trismus. No recurrence has been recorded after eighteen months. Dentists play an important role in managing and preventing oral complications of cancer treatment.

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## #32 Regional Odontodysplasia: Report of Two Cases and Review of Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 44

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University)*

Regional odontodysplasia (RO) is a rare nonhereditary developmental anomaly affecting the formation of dental tissue. Although various theories have been proposed, the etiology is unknown. RO affects both deciduous and permanent dentition, more commonly the anterior maxilla. It is prevalent in females with no racial predilection. It shows unerupted teeth or erupted teeth with small crowns accompanied by abscess and gingival swelling. On radiographs, the affected teeth show thin enamel and dentin with wide pulp chambers commonly described as ghost teeth. The treatment depends on the degree of anomalies, the function, and the esthetics. Here, we report two cases of RO.

Case 1: An 8-year-old boy presented with a history of over-retained #F and G, delayed eruption of #9 and 10, and partially erupted hypoplastic #14. On radiographs, teeth #9, 10, and 14 showed large pulp chambers, minimal root development, and a well-defined corticated radiolucency. The patient had multiple follow-up visits over two years. Teeth #F, G, 9, and 10 were extracted along with an excisional biopsy. Histopathologic and radiographic findings were consistent with regional odontodysplasia. Tooth #14 eventually developed dental caries and was extracted. A removable partial denture was fabricated for missing teeth.

Case 2: An 11-year-old boy was referred for evaluation of delayed eruption of #25, 26, and 27. Teeth #P, Q, and R were extracted at the age of 6 due to their abnormal appearance and associated pain, however, the alveolar ridge had a good size and contour. On the panoramic radiograph, teeth #25, 26, and 27 were malformed with open apices, large pulp chambers, and a shell-like appearance. The findings were consistent with regional odontodysplasia. The plan was to maintain the unerupted teeth until the patient is old enough to have implants in place, and a removable partial denture was fabricated to replace the unerupted teeth.

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## #33 Malignant Transformation of Oral Lichen Planus: A Case Report and Review of Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 45

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*Ms. Gabrielle Dennis (Loma Linda University), Ms. Somyung Ji (Loma Linda University), Dr. Craig Zuppan (Loma Linda University),  
Dr. Chi Viet (Loma Linda University), Dr. Anupama Grandhi (Loma Linda University)*

Oral lichen planus (OLP) is a relatively common inflammatory mucocutaneous disease with unknown etiology. The disease affects 0.5 to 2% of the general population with a predilection for females and middle-aged adults. It commonly presents as bilateral symmetrical lesions on the buccal mucosa, dorsal tongue, and gingiva. The two most common subtypes are reticular and erosive forms. According to Gonzales-Moles, the overall malignant transformation rate is 1.1%, with the reported range of 0% to 12.5%. Here, we present a case of OLP transformed into oral squamous cell carcinoma (OSCC).

Case: A 58-year-old woman presented with a 19-year history of leukoplakia on the dorsal and ventral tongue and a 9-year history of painful and eroded gums. In 2002, she had a diagnosis of lichen planus with low-grade dysplasia on biopsy. An incisional biopsy of the lesions from the right mandibular buccal gingiva and right dorsal tongue was performed in 2010. The diagnosis was OLP of gingiva and benign verrucous keratosis of the dorsal tongue. The oral lesions were treated and regularly followed up for 2.5 years, and the patient was lost to follow-up for seven years. In 2019, she presented with biopsy-proven invasive poorly differentiated OSCC of the right mandibular gingiva. She underwent a right marginal mandibulectomy, right selective neck dissection, local flap, and teeth extraction. On an 8-month follow-up, the patient developed a palpable nodule at the base of the right neck, and a level 1 neck node biopsy was positive for metastatic SCC. She had modified radical neck dissection and right mandibular vestibuloplasty. The patient is currently undergoing radiation therapy. The severity and progression of this case highlight the malignant potential of lichen planus and the importance of careful follow-up.

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## #34 PERIPHERAL KERATOAMELOBLASTOMA/SOLID KERATOCYSTIC ODONTOGENIC TUMOR

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Regular) - Abstract ID: 83

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**Introduction:** Keratoameloblastoma (KAB) is considered a rare variant of ameloblastoma (AMEL). Three histomorphologic types exist: a) simple (acanthomatous ameloblastoma), b) complex with features of both AMEL and keratocyst odontogenic tumor (KOT/OKC), and c) papilliferous revealing papillary keratin-containing extensions in cystic ameloblastomatous lumina. There are two opposing theories on the development of KAB; 1) the tumor arises de novo as an ameloblastoma with exuberant keratin production, and 2) ameloblastomatous transformation occurs in preexisting KOT/OKC. Rare reports exist of solid KOT/OKC (SKOT) sharing features with KAB suggesting a hybrid lesion. Recently, it has been suggested that KAB should be reclassified as SKOT based on the presence of *ptch1* variants. Herein, we present a peripheral odontogenic tumor with features of KAB and SKOT.

**Materials and Methods:** 54-year-old male presented with a firm tender swelling of the right side of maxilla in the area of the premolars causing saucerization of the alveolar bone.

**Results:** Originating from surface epithelium, the tumor exhibited plexiform, follicular and cystic growth patterns with features encountered in KOT/OKC and stellate reticulum areas and reverse polarization in the periphery. Foci of mild pleomorphism and a few scattered mitoses were noted. Plexiform and cystic areas featured abundant keratin. Elongated lingulae infiltrating the connective tissue as well as plexiform extensions with predominantly ameloblastomatous features and few areas of keratinization were appreciated on the surface epithelium. The stroma revealed plasmacytic inflammation and multiple calcifications representing calcifying keratin deriving from burst cysts. Cytokeratin 19 plasmalemmal staining was observed. Keratin in calcifications was also stained. Ber-EP4 revealed focal plasmalemmal reaction in the surface epithelium with ameloblastomatous changes. There was no staining for calretinin and BRAF V600E. Variable Ki67 nuclear staining was noted.

**Conclusions:** Features of KAB and SKOT were observed. Next generation sequencing for the presence of *brafor ptch1* variants is underway.

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## #35 Metastatic Lesions to the Oral Soft Tissues and Bones: A Seventeen-Year Retrospective Study

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 49

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**Introduction:** Metastatic neoplasms to the oral soft tissues and bones are exceedingly rare and are associated with poor prognosis. Clinically, metastatic lesions exhibit a strong predilection to the jaw bones, specifically the posterior mandible, while soft tissue involvement includes the gingiva and tongue. Metastatic tumors may pose diagnostic challenges since their histopathologic appearance varies. Herein, we report the clinico-epidemiologic and histopathologic characteristics of forty-one metastatic lesions to the oral mucosa and jaws.

**Material and methods:** Cases diagnosed as metastatic lesions during the period 2003-2020 were retrieved. The demographic features (age and gender of the patients), clinical impression and location, as well as primary site and histologic type of the neoplasms were recorded.

**Results:** Among 126,229 biopsy specimens, only 41 (0.03%) comprised metastases. The patients' mean age was 67.6 years (age range= 18-94 years) without any apparent gender bias (male: female ratio=21:20). The possibility of metastasis was considered in only half (21, 51%) of the cases. Twenty-two cases affected the gingiva (54%); six (27.3%) presented with tautochronous bone involvement. Ten cases (24%) were exclusively intraosseous and manifested as ill-defined, unilocular, mandibular radiolucencies. Other anatomic sites included the alveolar mucosa and hard palate (3 each, 7.3%), tongue (2, 5%) and soft palate (1, 2.4%). Collectively, the most common primary site was the lung (12, 29%) followed by the colon (6, 15%), breast and kidney (4 each, 10%), pancreas (2, 5%), and prostate, heart, esophagus, sublingual gland, adrenal, bone and CNS (1 each, 2.4%). In six cases (15%) the primary was unknown. In both men and women, lung was the most common primary site (33.3% and 25%, respectively).

**Conclusions:** Metastases to the oral cavity and jaws are rare. When present, they show a predilection for the gingiva and mandible. Clinicians should remain aware about metastatic neoplasms since they can frequently mimic reactive pathoses.

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## #36 Lymphoepithelial carcinoma of the oral mucosa. A rare histopathologic subtype of squamous cell carcinoma.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 77

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**Objectives:** Lymphoepithelial carcinoma (LEC) represents a rare histopathologic subtype of squamous cell carcinoma (SCC), which shares morphologic similarities with non-keratinizing undifferentiated nasopharyngeal carcinoma (NPC). LEC may be etiologically associated with EBV, especially in endemic regions. Although being described in several locations of the head and neck, the oral mucosa seems to be an infrequent site of involvement. Herein, we present a case of an EBV-negative oral LEC and discuss the diagnostic challenges.

**Findings:** A 51 year-old male Filipino patient suffering from chronic kidney disease presented for the evaluation of a swelling in the right mandible, first noticed by the patient a few days ago. Clinically, the right posterior mandibular gingiva appeared enlarged and hemorrhagic, covered by ulcerated mucosa, while the adjacent teeth displayed high mobility. Extraorally, a painful and slightly movable lymph node in the right submandibular area was palpated. Radiographic evaluation revealed extensive bone loss in the right posterior mandible. With a provisional diagnosis of lymphoma or SCC, an incisional biopsy was performed. Histopathologic examination revealed invasion of the underlying connective tissue by pleomorphic cells of epithelioid morphology, occasionally arranged in syncytial islands, arising from a partially ulcerated, non-keratinized squamous epithelium. A prominent diffuse lymphocytic cell infiltrate surrounding the tumor cells was also detected. The neoplastic cells expressed pancytokeratin, CK5/6, p63, and p40, while p16, LMP-1, and in situ hybridization for EBV RNA were negative. A final diagnosis of moderately to poorly differentiated SCC with features indicative of LEC was rendered and the patient was referred for further management.

**Conclusions:** LEC should be considered in the differential diagnosis of undifferentiated malignant neoplasms displaying close association with diffuse lymphoid aggregates. The evaluation of nasopharynx is justified to rule out metastatic NPC, as the separation of these two entities microscopically is not feasible.

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## #37 Base of tongue metastasis of cutaneous malignant melanoma masquerading as a pedunculated reactive lesion. Report of a case with rhabdoid and neuroendocrine features.

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 78

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**Objectives:** Metastatic melanoma (MM) represents a highly aggressive malignancy associated with poor prognosis. MM has a propensity to microscopically resemble other neoplasms by displaying diverse morphologic variations. Herein, we present a case of cutaneous melanoma metastatic to the base of tongue clinically mimicking a reactive pedunculated lesion and histopathologically exhibiting unusual rhabdoid and neuroendocrine features.

**Findings:** A 63-year old male presented for evaluation of a painless tongue mass of 5 months duration. The patient had been surgically treated for cutaneous melanoma of the right chest 5 years ago. Clinically, a pedunculated, soft, partially ulcerated, hemorrhagic mass involving the left base of tongue was noted. With a provisional diagnosis of pyogenic granuloma or other reactive lesion, an excisional biopsy was performed. Histopathologic examination revealed invasion of the connective tissue by clusters of epithelioid and/or plasmacytoid neoplastic cells with prominent eosinophilic nucleoli and various degrees of cytoplasmic melanin pigmentation. Interestingly, a small subset of cells showed rhabdoid morphology. Most tumor cells expressed Melan-A, HMB-45, SOX-10, and S-100, while cells with rhabdoid morphology were positive for Myo-D1. Chromogranin and synaptophysin were also positive in a few scattered cells. A final diagnosis of MM with rhabdoid and neuroendocrine features was rendered. Molecular investigation revealed mutations for the BRAF V600E gene, while multiple brain metastatic foci were detected on further diagnostic work-up.

**Conclusions:** Divergent differentiation of melanoma cells may cause diagnostic pitfalls necessitating thorough immunohistochemical analysis. The presence of rhabdoid cells, although infrequent in primary melanomas, is relatively common in metastatic tumors. Furthermore, neuroendocrine differentiation in melanomas is very uncommon and its co-existence with rhabdoid features is exceedingly rare. Better characterization of such histopathologic variations in melanomas with evaluation of their potential biologic significance is warranted.

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## #38 Oral Focal Mucinosis, an uncommon entity: Report of three cases

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 86

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*Dr. Yamely Ruiz (UNAM), Dr. Beatriz Aldape (UNAM), Dr. Ana María Cano (INCan), Dr. Bernardo Cruz (UNAM)*

### **Introduction**

Oral focal mucinosis (OFM) is a rare soft tissue lesion of unknown etiology and it is considered the oral counterpart of cutaneous focal mucinosis or cutaneous myxoid cyst. The pathogenesis of OFM may be explained by fibroblast overproduction of hyaluronic acid. Clinically, it appears as painless swelling of the same color as the surrounding mucosa. The gingiva is the most common site and the diagnosis is established only by histopathological and immunohistological examination.

### **Materials and Methods**

We present three cases of gingival oral focal mucinosis.

Case 1: 40 year-old woman with painless and slow-growing swelling of two years of evolution at buccal mandibular gingiva, between lateral incisor and canine. The lesion measured 2 cm. and exhibited the same color as the overlying mucosa; there was not radiographic involvement.

Case 2: 48 year-old female with a hard palatal swelling. The lesion was sessile, firm, and not color change

Case 3: 36 year-old male with a painless and same color as the normal mucosa gingival swelling. The lesion was between central and lateral right incisors, it was sessile and measured 1.5 cm

### **Results**

In the three cases excisional biopsy was performed and histopathologically they shown the proliferation of elongated and mucinous material in a myxoid matrix. This myxoid stroma was surrounded by collagenous fibrous connective tissue and covered with squamous epithelium. Histochemical staining with Alcian blue was positive in the mucinous areas

### **Conclusions**

These three cases are significant clinically as well as histopathologically. The lesions clinically mimic an ossifying peripheral fibroma or a fibroma. Thus, the histopathological and histochemical findings with special stains, such as Alcian blue, are determinant to establish the diagnosis of these conditions. Treatment is the complete surgical excision and its recurrence is not reported.

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## #39 Intraosseous Benign Fibrous Histiocytoma of the Mandible

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 88

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*Dr. Sonia Sanadhya (University of Maryland, School of Dentistry), Dr. James Kassolis (Private Practice of Periodontics), Dr. John R Basile (University of Maryland, School of Dentistry)*

### **Introduction**

Fibrous histiocytomas are a diverse group of tumors that exhibit fibroblastic and histiocytic differentiation and are considered to be a true neoplasm. Intrabony fibrous histiocytomas of the jaws are rare, with most cases being reported in the femur, tibia, pelvis and occasionally the skull bones.

### **Material and Methods**

This report describes a case of 69 year old male patient who presented with pain in the area of tooth #18 (mandibular left second molar) and an endo-perio lesion that perforated through the cortical bone which appeared to be multilocular on radiographs. Clinically, there was a buccal swelling and when the buccal bifurcation was probed, a 10mm depth was revealed with a purulent exudate emanating from the pocket. The bridge was sectioned at the junction of #19 and 20 pontic followed by extraction of tooth #18. Reflection of the mucosa revealed a large bony lesion destroying the buccal plate of bone in the area of teeth #18 and 19. The purulent material was primarily associated with the tooth #18 and not the overall lesion. The majority of the lesion was enucleated and removed.

### **Results**

Histopathological examination revealed fragments of mildly inflamed connective tissue that demonstrated a focus of spindle or mesenchymal cells proliferating in a whorling or storiform pattern. The tissue contained large scattered deposits of mineralized material and fragments of vital bone. Immunohistochemical stains for Desmin, SMA, CD34 and factor XIIIa were negative while diffusely positive for CD68. The positivity for CD68 demonstrated that the lesion was composed of histiocytic cells.

### **Conclusions**

Benign fibrous histiocytomas of the soft tissues are well recognized entities, the rare intra-bony counterpart shares clinical, radiographic and histological features with odontogenic tumors, non-ossifying fibromas, osteoblastomas and fibrous dysplasia, and therefore should be considered in the differential diagnosis of these lesions.

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## #40 A case of B Cell Lymphoma, NOS – A Manifestation in the Labial Mucosa

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Tuesday, 25th May - 08:00: Poster Presentations - Oral to Poster - Abstract ID: 80

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*Dr. Sonia Sanadhya (University of Maryland, School of Dentistry), Dr. Elaine S Jaffe (Center for Cancer Research, National Cancer Institute, Bethesda, Md), Dr. John R Basile (University of Maryland, School of Dentistry)*

### **Introduction**

Non-Hodgkin's lymphoma of B cell lineage is the third most common malignancy of the maxillofacial region. DLBCL, NOS constitutes 25% - 35% of the adult non-Hodgkin's lymphomas and is more common in elderly individuals, as well as more prevalent in males than in females. The etiology of DLBCL, NOS remains unknown. Underlying immunodeficiency is a significant risk factor.

### **Materials and Methods**

We report a case of a 78-year old female patient who presented with a lump in the upper labial mucosa since two weeks, there were no symptoms prior to the stated time duration. The soft tissue mass was located approximately 4mm from the vermillion border, adjacent to the incisal edges of teeth #10 and 11 (left maxillary lateral incisor and canine), measured around 1cm in diameter and was firm on palpation. The clinical differential included benign salivary gland neoplasms, lipoma and a mucocele. The patient had a past medical history of diffuse large B cell lymphoma three years ago.

### **Results**

Microscopic examination showed an atypical lymphoid proliferation involving the soft tissue with infiltration into the adjacent skeletal muscles. A few residual ductular/glandular elements were also noted. The biopsy was composed of a monotonous infiltrate of medium sized cells with round to irregular nuclei, one or more prominent nucleoli and scant cytoplasm. A polymorphous inflammatory background was not observed, and the cells diffusely infiltrated in to the soft tissue. Immunohistochemistry revealed that the atypical cells were positive for CD20 and BCL2 while negative for CD10, BCL6 and c-Myc. Ki-67 proliferation index was moderate to high.

### **Conclusions**

In summary, the overall features were consistent with a B-cell lymphoma that was difficult to further classify due to limited sampling. Based on the relatively high proliferative rate, the lesion was likely consistent with recurrence of the previously diagnosed diffuse large B-cell lymphoma.

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## #41 Melanin Pigmented Odontogenic Lesions- Report of a Dentigerous Cyst and Review of the Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 11

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*Dr. Hajir Aldaod (Mount Sinai Hospital , New york), Dr. Sagar Khanna (Mount Sinai Hospital , New york), Dr. Suchie Chawla (Mount Sinai Hospital , New york), Dr. NAOMI RAMER (Mount Sinai Hospital , New york)*

### INTRODUCTION

The presence of melanocytes in the oral cavity, especially in the gingiva and buccal mucosa is a well documented finding, especially in dark complexioned individuals. Melanocytes are quite rare in odontogenic cysts and tumors. The melanotic neuroectodermal tumor of infancy was initially considered to be the only pigmented jaw tumor in existence. This was until 1961 when Lurie et al reported the first melanin pigmented calcifying odontogenic cyst. In 1964, Grand and Marwah reported the first case of a pigmented odontogenic cyst, that of a gingival cyst. Although most pigmented odontogenic cyst are odontogenic keratocysts , pigmented dentigerous cyst are second in frequency with three cases reported to date.

### MATERIALS AND METHODS

A case of a macroscopic and microscopic melanin pigmented dentigerous cyst is presented with histological and immunohistochemical findings. A pubmedline search of the literature was conducted to review all cases of melanin pigmented odontogenic cysts and neoplasms reported to date.

### RESULTS

The pigment in the lesion was confirmed to be melanin by Masson- Fontana staining. The presence of dendritic melanocytes within the lesion was demonstrated by S-100 immunostaining. Since 1961, sixty-three melanin pigmented odontogenic cysts and tumors have been reported. The odontogenic pigmented lesions found in the literature differ in type, histochemical, Immunohistochemical and ultrastructural methods used and have demonstrated that the pigment is always melanin. Most of the patients are Asians and Blacks, thereby implicating racial pigmentation as an important factor.

### CONCLUSION

We report a melanin pigmented dentigerous cyst associated with impacted teeth 16 and 16A in a 27 year old black male. Our dentigerous cyst demonstrated macroscopic and microscopic visible melanin pigmentation. Melanin pigmented dentigerous cysts are quite rare with only three other cases reported to date in the English language literature to which we add this first case involving the maxilla.

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## #42 Central Giant Cell Granuloma in a patient with Neurofibromatosis Type 1

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 12

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*Dr. Daria Vasilyeva (Columbia University), Dr. Yakov Yakubov (Columbia University), Dr. Elizabeth Philipone (Columbia University), Dr. Alia Koch (Columbia University)*

Neurofibromatosis type 1 (NF1) is an autosomal dominant genetic disorder with variable expressivity. Oral manifestations are found in a large percentage of NF1 patients. Central Giant Cell Granulomas (CGCG) have been reported in patients with NF1, but the association is not well understood. We present a longitudinal case study of a CGCG in a patient with NF1 treated with intralesional steroid injections.

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## #43 Lichen Planus and the Vulvovaginal Gingival Syndrome- Report of a Case and Review of the Literature

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 16

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*Dr. Margarita Katser (Mount Sinai Hospital , New york), Dr. Suchie Chawla (Mount Sinai Hospital , New york), Dr. Robert Phelps (Mount Sinai Hospital , New york), Dr. NAOMI RAMER (Mount Sinai Hospital , New york)*

### INTRODUCTION

Lichen planus is a dermatologic disease that affects both skin and mucosa. Cutaneous lesions of lichen planus present as small polygonal violaceous papules that present most often on the flexor surfaces of the extremities. These lesions may also be present on the vulvar skin and the hair-bearing aspects of the vulva.

Lichen planus of the mucous membranes may affect buccal or gingival mucosa and the vulvar vestibule and vagina. Examination of the mouth reveals the white, lacy pattern of hyperkeratotic striae present on buccal mucosa, often bilaterally. The gingiva may be diffusely swollen, erythematous, and exhibit desquamation.

In 1982 Pelisse first reported the vulvovaginal gingival syndrome and lichen planus. The main clinical manifestations of the syndrome are a triad of vulvar, vaginal and gingival lichen planus.

### CASE REPORT

A sixty-five year old female was referred to the oral surgeon for evaluation of red patches in her mouth. The patient reported that she had a biopsy of her vulvar for which a diagnosis of lichen sclerosis et atrophicus was made. A biopsy of her maxillary gingiva was submitted to the oral pathology laboratory and diagnosed as lichen planus. The vulvar biopsy was sent for comparison. After consultation with Dermatopathology the diagnosis of lichen planus and the vulvovaginal gingival syndrome was made.

### CONCLUSION

We report a case of severe erosive vulvovaginal lichen planus that preceded the onset of gingival lichen planus by nine months. Not uncommonly, patients with severe oral lesions displayed mild genital disease and patients with asymptomatic gingival disease suffered with severe erosive vulvovaginal disease. The diagnosis of vulvovaginal gingival syndrome requires multidisciplinary evaluations by various medical specialists. We have reviewed the literature of this rather rare condition and report an additional case, with the intention of making the practicing clinician aware of this unusual clinical entity.

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## #44 Sclerosing odontogenic carcinoma: a rare and challenging biopsy diagnosis

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Tuesday, 25th May - 08:00: Poster Presentations - Poster (Student/Resident) - Abstract ID: 53

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*Dr. Carleigh Canterbury (Columbia University Medical Center), Dr. Elizabeth Philipone (Columbia University Medical Center), Dr.*

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### Introduction

Sclerosing odontogenic carcinoma (SOC) is a primary intraosseous malignancy first described by Koutlas et al. in 2008 and recently listed as a separate entity in the 2017 World Health Organization Classification of Head and Neck Tumors. Fewer than fifteen cases of SOC have been reported in the literature to date, with many mischaracterized on initial biopsy diagnosis. This mischaracterization, particularly on biopsy, is likely attributable to the subtle and variable histologic features of SOC. We present a case of SOC and review the literature with emphasis on clinical and histologic features that should raise suspicion for this lesion on biopsy.

### Material and Methods

The biopsy specimen was submitted in a 10% formalin fixative and subsequently embedded in paraffin. The specimen was processed into four-micrometer-thick sections and stained for routine microscopic examination. Immunohistochemical studies were performed and included antibodies against CK5/6, CK19, e-cadherin, CK7, CK20, p63, and Ki67.

### Results

A review of the literature demonstrated only one set of authors reporting an initial biopsy diagnosis of SOC. The remainder of reported cases required a minimum of one repeat biopsy, and in several cases, diagnosis was only established following complete excision. Therapeutic approaches ranged from conservative marginal excision to radical excision with neck dissection and adjuvant chemotherapy and radiation. Regardless of therapy modality all cases demonstrated no recurrence, with one exception, likely attributable to positive margin status at resection.

### Conclusions

Given the subtle features of SOC that may often mimic other benign and malignant processes, sufficient awareness and suspicion for this entity is necessary for diagnosis, and ultimately to guide clinical management. In the absence of an established diagnosis of SOC, multiple reported patients have undergone more radical therapy than what appears to be necessary for the management of this low-grade entity with no apparent metastatic potential.