AAOMP
American Academy of Oral
& Maxillofacial Pathology

Poster Program

Tuesday – June 18, 2013
8:30 am – 11:30 am
#1

**ORAL LESIONS AS THE PRESENTING MANIFESTATION OF CROHN'S DISEASE V**

**Woo, E Herschaft, J Wang**  
U of Nevada, Las Vegas  
Crohn's disease (CD) is an immune-mediated disorder of the gastrointestinal tract which together with ulcerative colitis, comprise the two major types of inflammatory bowel disease (IBD). The underlying etiology has been attributed to defects in mucosal immunity and the intestinal epithelial barrier in a genetically susceptible host, resulting in an inappropriate inflammatory response to intestinal microbes. The lesions of CD can affect any region of the alimentary tract as well as extraintestinal sites such as the skin, joints and eyes. The most common presenting symptoms are periumbilical pain and diarrhea associated with fevers, malaise and anemia. Oral involvement has been termed oral CD and may manifest as lip swelling, cobblestoned mucosa, mucogingivitis and linear ulcerations and fissures. Oral lesions may precede gastrointestinal involvement and can serve as early markers of CD. We describe a 6-year-old male who presented for evaluation of multifocal gingival erythema and swellings. His medical history was unremarkable for gastrointestinal disorders or distress. Histopathologic examination showed multiple well-formed granulomas that were negative for special stains and foreign body material. A diagnosis of granulomatous gingivitis was rendered. The patient was advised to seek consultation with a pediatric gastroenterologist and following colonoscopy, was diagnosed with early stage CD. Timely recognition of the oral manifestations of CD is critical because only a minority of patients will continue to exhibit CD-specific oral lesions at follow-up. This places the dental practitioner in a unique position to detect occult CD in an otherwise asymptomatic patient, which may ultimately lead to early diagnosis and treatment.

#2

**DRY MOUTH AND NUTRITION: QUALITY OF LIFE IN SJÖGREN’S SYNDROME PATIENTS H Lanfranchi, M Ansola**  
Department of Oral Medicine, School of Dentistry, U of Buenos Aires. Buenos Aires  
Sjögren’s Syndrome (SS) is an autoimmune disease that affects the exocrine glands, causing dry mouth. Local moisturizers, immunosuppressants, and antimuscarinic medications are commonly prescribed. However, there are few works addressing the influence of eating habits caused by dry mouth on the quality of life of patients with this disease. In the present study we evaluate the nutritional status and eating habits in patients with SS and make adjustments to their diet by adding moisture and soft textures that avoid damaging the oral mucosa. We studied 25 SS* patients through nutritional interviews to inquire about their eating habits. Ninety percent of the patients (n=22) reported having to limit their food selection because certain foods caused them discomforts. Nutritional status was assessed based on BMI, laboratory assays, and asking about changes in weight since disease onset. Based on the results, each patient’s diet was modified focusing on changes in the consistency and moisture of foods. Each patient was given a healthy eating plan and a guide to the selection and preparation of foods for dry mouth patients that ensure the inclusion of all necessary nutrients, designed by our Nutrition Service. 85% of the patients self-reported improvements in their diet and ability to swallow foods and therefore in quality of life. These results show the importance of the creation of a personalized eating plan that is consistent with the patient’s degree of oral dryness and that ensures the nutritional value is maintained. *Patients were diagnosed with Sjögren’s Syndrome according to the criteria established by Sjögren’s International Clinical Collaborative Alliance (SICCA). Supported by National Institutes of Health contract NOI-DE-32636.
ORAL LESION OF PSEUDOXANTHOMA ELASTICUM  B Trump, R Assi, A Bhattacharya  Texas A&M U Baylor College of Dentistry, Dallas  We report a case of pseudo xanthoma elasticum involving the oral cavity in a 65-year-old female. Pseudo xanthoma elasticum is a heritable multisystem disorder with cutaneous, ophthalmologic, and cardiovascular manifestations. PXE is caused by mutations in the ABCC6 gene located on chromosome 16p13.1. A recent study found an association of oral mucosal lesions and cardiovascular disease. Our patient presented with two areas of leukoplakia on the soft palate. Both lesions were excised and submitted for histopathologic diagnosis to rule out epithelial dysplasia. One lesion was diagnosed as hyperkeratosis and mild acanthosis while the other was consistent with pseudo xanthoma elasticum.

QUANTITATIVE SCORE OF LANGERHANS CELLS IN LICHEN PLANUS AND AMALGAM LICHENOID REACTION. SMC Grossmann, GR Souto, RA Mesquita, U Federal De Minas Gerais, Belo Horizonte  Amalgam lichenoid reaction (ALR) presents clinical and histological features of oral lichen planus (OLP); however both are differences on the evolution and on evidences of hypersensitivity to amalgam. Langerhans cells (LCs) are cells of the innate immune system and responsible for initiating adaptive immune response. The aim of this study was to measure the number of CLs in ALR, OLP and normal oral mucosa (NOM). CLs were identified by immunohistochemistry for CD1a protein in 21 samples of OLP, ALR and NOM in oral epithelium (OE) and lamina propria (LP). They also were evaluated separately in the region of subepithelial inflammatory infiltrate (RSII) and region down the subepithelial inflammatory infiltrate (RDSII) for samples of OLP and ALR. Density of LCs (cells/mm2) was determined using the AxioVision 2.4 software (Microsoft, Carl Zeiss Vision Gm bH, Gottingen, Germ). Samples were obtained from file of Service Maxillofacial Pathology, School of Dentistry, Universidade Federal de Minas Gerais (1965 to 2010). It was observed significantly higher density of CD1a + LCs in RSII (95.73 cells/mm2) and OLP (87.04 cells/mm2) with the OLP when compare with RSII (44.71 cells/mm2), OLP-ALR (42.12 cells/mm2), and OLP-NOM (29.82 cells/mm2). The densities of CLs were correlated with the density of inflammatory cells and were strong and positive correlations between OE cells, RSII, RDSII and OLP, when compare with inflammatory infiltrate in OLP (p> 0.05). Strong positive correlation was also observed in the RDSII in ALR (p <0.05). It was observed higher densities of inflammatory cells in the OLP and ALR when compare with NOM (p <0.05). Ours results suggest that different immunologic mechanisms can be associated with the pathogenesis of OLP and ALR. (Support: CNPq; Fapemig; CAPES)
#5

RECURRENT ALLERGIC CONTACT STOMATITIS PRESENTING AS ORAL BULLAE  
C Fisher, P Edwards  
U of Michigan, Ann Arbor  
Purpose: To report a case of allergic contact stomatitis, initially presenting clinically as oral bullae. Background: Allergic contact stomatitis is a delayed-type hypersensitivity reaction (Type IV). Its oral manifestations include inflammation followed by erosions and can present as bullae, occasionally resembling lesions of pemphigoid or pemphigus. These manifestations usually occur within 24-72 hours of contact with the antigen. Methods: A 25 y.o. Asian male presented with a chief complaint of recurring multiple, non-painful blisters occurring on the hard palate. Patient reported no lesions anywhere else on the body. A large (>1cm), fluid filled bulla on the left hard palate and multiple smaller bullae and erosions were noted on the right hard palate. These lesions resolved within one week without therapy. Results: Subsequently, a 4mm punch biopsy was performed. The H&E stained frozen sections demonstrated stratified squamous epithelium with underlying connective tissue containing a mild mixed inflammatory infiltrate. Direct immunofluorescence demonstrated strong fibrin positivity along the BMZ; weak positivity for C3 and IgM along the BMZ; and was negative for IgG and IgA. A CBC with differential, and serology for ANA and double-stranded DNA were unremarkable. Conclusion: The patient later revealed that the bullae only occurred after consuming large quantities of alcoholic beverages. Based on the clinical history and presentation, a diagnosis of allergic contact stomatitis was made.

#6

OROFACIAL LESIONS IN INFANTS MANAGED BY NASOALVEOLAR MOLDING APPLIANCES  
B Taylor, B Acharya, C Flaitz  
U of Texas School of Dentistry at Houston, Houston  
Purpose: This retrospective clinical study documented common orofacial lesions in infants with cleft lip and palate (CL/P), who were managed using a nasoalveolar molding appliance (NAMA). Methods: Following approval by the institutional board review, a convenience sample of infants referred for presurgical treatment of CL/P with NAMA were studied between 2006-2011 at pediatric dental residency clinic. Dental records and photographs were evaluated, including demographics, medical history, cleft type, orofacial lesions and treatment complications. Data were analyzed using descriptive statistics, t-test and chi-square with significance at p <.05. Results: Records of 141 infants were evaluated including 101 (71.6%) unilateral (U) CL/P and 40 (28.3%) bilateral (B) CL/P; 56% males and 44% females. Ethnicity included: 23% White, 10% Black, 58% Hispanic, 5% Asian, 4% Other. Mean age at initial examination was 48 days and mean length of treatment was 80 days. Documented oral lesions were sucking callus (70%), palatal keratosis (29%), candidiasis (22.5%), neonatal palatal and gingival cyst (28%), traumatic ulcer (21%), sucking ulcer (6%), neonatal tooth (6%), eruption cyst (1.4%), neonatal alveolar lymphangioma (.7%), pyogenic granuloma (.7%). In total, 77% of infants had > 1 oral lesion. Facial lesions included milia/ acne (64%), contact tape irritation (63%), nevus simplex (33%), seborrheic dermatitis (27%), impetigo (.7%), abrasion (.7%), eczema (.7%) with 77% of infants having facial lesions. Except for palatal keratosis (p = .03), there was no significant difference in orofacial lesions based on cleft type. Conclusion: Orofacial lesions, including NAMA complications, are common in infants CL/P undergoing NAMA therapy, but the majority are not associated with type of cleft.
#7

INTRAVASCULAR FASCIITIS INVOLVING THE UPPER LIP  J O'Donnell Jr., T Rosenberg, L Kahn, R Kelsch  Hofstra North Shore-LIJ School of Medicine, New Hyde Park  

Background: Intravascular fasciitis is a rare variant of nodular fasciitis involving arteries or veins typically in an extremity. However, approximately 10% of published cases involve the head and neck. Observation: We report a case of a 46-year-old female with an upper lip submucosal mass. The lesion was noted following minor trauma to the lip and was present for several months. At the time of evaluation there was neither ulceration nor paresthesia appreciated. An excisional biopsy was performed. Histopathological evaluation revealed a proliferative spindle cell lesion with readily identifiable mitoses. The lesion was multinodular with individual nodules occluding the lumina of medium-sized vessels. Immunohistochemical analysis demonstrated positive staining of lesional spindle cells for smooth muscle actin and positive staining of the accompanying prominent endothelial cell population by CD34. Intense positivity of smooth muscle markers highlighted the surrounding vessel wall. Based on these features a definitive diagnosis of intravascular fasciitis was rendered. Conclusion: Intravascular fasciitis has been infrequently reported in the oral cavity. The lesional cellularity and brisk mitotic activity, as seen in the family of pseudosarcomatous myofibroblastic proliferations, could lead to misdiagnosis of sarcoma. Recognition of the typical histological features of intravascular fasciitis is important for the practicing oral pathologist.

#8

DETECTION OF HERPESVIRUS TYPE 8 (HHV8) IN ORAL BIOPSIES OF KAPOSI SARCOMA BY IMMUNOHISTOCHEMISTRY AND PCR  J Seo, P Tobouti, N Sugaya, S Sousa  U of São Paulo, São Paulo  

Kaposi's sarcoma (KS) is a multifocal vascular tumor that occurs most commonly in patients who have immunosuppression caused by HIV. KS-associated herpes virus (human herpes virus 8, KSHV) has been identified as the causative agent. KS growth involves the up regulation of many key HHV8 gene products, such as the latency-associated nuclear antigen (LANA-1 or LNA-1). Frequently oral cavity may represent the initial site of KS, often being also the first clinical indication of HIV infection in previously undiagnosed individuals. Due to the great variability of morphological aspects, it may be necessary to assess the positivity to HHV8 in order to differentiate KS from other vascular lesions. In our biopsy service the main problem can be the small size of most biopsies that sometimes difficulties the detection of HHV8. Thus, in the present study we aimed to compare IHC and PCR techniques to detect HHV8 in oral biopsies of supposed KS. Sixteen cases diagnosed in the last five years were retrieved from the files of the Oral Pathology Department at the University of São Paulo. The biopsies measured an average of 2.5mm in diameter. For IHC the material was submitted to the streptavidin-biotin method and the antibody used was anti HHV8-LNA (Novocastra). For PCR, DNA, from the lesions was isolated using the phenol-chloroform extraction protocol. Primers were obtained from NCBI-GenBank databases. Real-time PCR was performed on DNA using Jumpstart SYBR green mastermix (Sigma-Aldrich) on a thermocycler (Applied Biosystems 7500 Real-Time PCR System). Fifteen samples, which were HHV8-IHC positive, were positive to HHV8- DNA detection. Conclusion: IHC showed to be as sensitive and reliable as RT-PCR even in small biopsies, being a cheaper and faster method.
ECHINOCOCCUS OF THE TONGUE: A CASE REPORT AND REVIEW OF THE LITERATURE
S Wetzel, K Murtagh, H Sacks, R Reich, P Freedman  New York Hospital, Queens  Echinococcosis is a zoonosis caused by a canine tapeworm. Although dogs are the definitive target for this parasite, humans can serve as an intermediate host when eggs from the organism are ingested. Infection results in formation of a hydatid cyst. Echinococcal infections are common in areas endemic to the entity including the Middle East, Europe and parts of Africa. However, there is an increase in cases arising outside of these parameters. The most common site for hydatid cyst formation is the liver and lungs. Only 1-2% of cases are seen in the head and neck region. The current case is that of an adult African male that presented with an enlarged tongue. Imaging studies revealed a cystic lesion involving the deep musculature. Histological examination of the excision specimen showed an echinococcal organism residing within the hydatid cyst. This case represents an unusual anatomic location for this occurrence, as well as, an uncommon geographical location. For these reasons, a parasitic infection did not enter into the differential diagnosis. Failure to recognize this condition can lead to complications during the excision process. These risks include anaphylaxis and secondary infection from removal of the cyst. The following case represents the significance of proper recognition and treatment for this condition.
#11

NK/T CELL LYMPHOMA, NASAL TYPE  J Doscher, M Johnson  Yale New Haven Hospital, New Haven

NK/T cell lymphoma, nasal type is an aggressive lymphoma commonly presenting as a destructive process of the facial midline, often demonstrating clinical and histologic features of necrosis and angioinvasion. It was initially categorized as an angiocentric lymphoma in the Revised European-American Lymphoma (REAL) classification. It has been recategorized as extranodal NK/T cell lymphoma in the WHO classification of lymphoid neoplasms. Historically these tumors were considered part of “midline lethal granuloma”. This entity frequently arises in extranodal regions: nasal or paranasal sinus. In addition, other extranodal sites include palate, trachea, skin and, the gastrointestinal tract. In most instances, EBV genomes are detectable in the tumor cells and immunohistochemistry detects CD56 positivity. The pathologic diagnosis of nasal-type NK/T cell lymphoma is based on the following criteria: expression of cytoplasmic CD3, CD56 and positivity for EBV in situ hybridization. If EBV in situ hybridization is negative, the immunophenotype studies should demonstrate cytoplasmic CD3 expression and positive cytotoxic molecules such as TIA-1. Nasal-type NK/T cell lymphoma is known to be one of the most aggressive lymphomas, so it is imperative to offer an appropriately aggressive treatment at an early stage of disease. We present the clinical and therapeutic treatment course and outcome of a 60 year-old caucasian male diagnosed with NK/T cell lymphoma. This includes a clinicopathologic review, approaches to diagnosis and surgical and prosthetic treatment strategies for patient optimization throughout oncologic therapies.

#12

CD30-POSITIVE IMMUNOBLASTIC PROLIFERATIONS OF THE ORAL CAVITY. IS ATYPICAL HISTIOCYTIC GRANULOMA RELATED?  R Eversole, A Dovigi  Oral Pathology Diagnostic Services, San Diego

Background/Introduction: CD30 (Ki-1) is a surface differentiation protein that is demonstrable in 1% of bone marrow cells and is a member of the tumor necrosis superfamily 8 (TNFRSF8). This glycoprotein is expressed on Reed-Sternberg immunoblasts in Hodgkin lymphoma and other lymphoblastic lesions of the skin and internal organ sites. CD30 positive lymphoproliferative diseases are rare in the oral cavity and may be seen in conjunction with systemic and cutaneous counterparts. Material and Methods: Archived formalin-fixed sections from 67 cases of oral CD30-positive lymphoproliferative lesions with large cell lymphoblasts were analyzed for lymphoid markers. Cytologically, the large cell nuclei varied from Sternberg to cerebriform to pronate. Stromal infiltrates varied within these lesions including small lymphocytes, commaform lymphocytes, plasma cells, neutrophils and eosinophils. Follow-up data were obtained. Results: CD30-positive cells were encountered in two lesions with benign outcomes: traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) and atypical lymphoblastic (histiocytic) granuloma (ALG). Cases with malignant behavior included solitary mucosal and systemic large anaplastic CD30-positive T cell lymphoma. Conclusion: Mucosal CD 30-positive lymphoproliferative lesions can be classified in similar fashion to cutaneous lesions with TUGSE and ALG behavior being similar to cutaneous lymphomatoid papulosis.
UNUSUAL ATYPICAL EPITHELIAL PROLIFERATION OF UNKNOWN BIOLOGICAL BEHAVIOR IN A 5-YEAR OLD
R. Gopalakrishnan, G Velasco-Peña, MD Rohrer, IG Koutlas
U of Minnesota, Minneapolis; Private Practice, St. Cloud
A 5-year-old male presented clinically with a subgingival supraperiosteal expansile mass associated with the mandibular anterior central incisors. There was no obvious clinical intraosseous involvement. The oral mucosal epithelium did not reveal any clinical changes suggestive of a process originating from the surface. Histopathologic evaluation revealed soft tissue fragments that were surfaced or lined by hyperkeratinizing and acanthotic stratified squamous epithelium exhibiting cytologic variations that include irregular rete pegs, loss of polarity, individual cell keratinization, increased nuclear/cytoplasmic ratio and prominent nucleoli. Areas of prominent endophytic epithelial proliferation comprised of cords and islands that showed cytological atypia were also present. The differential diagnosis was between a well-differentiated squamous cell carcinoma versus atypical epithelial proliferation of uncertain biologic behavior. Review of the literature revealed a similar lesion reported by Elzay and O’Keefe (Oral Surg Oral Med Oral Pathol 1979;47:436) who also discussed the unusual presentation of their case and the inability to completely exclude well differentiated squamous cell carcinoma. External consultation with three senior oral pathologists resulted in the diagnoses of atypical epithelial proliferation (2) and squamous cell carcinoma (1) further underscoring the difficulty in obtaining a definitive diagnosis. No additional treatment was performed and the patient remained lesion-free in a ten-month follow-up.

SUSTAINED OVEREXPRESSION OF INSULIN-LIKE GROWTH FACTOR II mRNA-BINDING PROTEIN 3 (IMP3) IN ORAL EPITHELIAL DYSPLASIA (OED): A POTENTIAL PROGNOSTIC BIOMARKER OF IMPENDING INVASION
G Mainville, C Allen, L Ayers, J Hagen, M Tong, D Kellough, S Mallery
The Ohio State U, Columbus
Identification of an invasion-predictive biomarker that identifies OED lesions with a high transformation potential could have clinical implications e.g. help target chemoprevention. IMP3, which is a recently identified oncofetal protein essential for mRNA binding, trafficking and stabilization, is overexpressed in many cancers, including OSCC. Clinical studies have shown that IMP3 expression in biopsies correlates with the presence of carcinoma in excised cervical epithelium. The objective of this case-control retrospective pilot study was to determine whether or not patterns of IMP3 expression are associated with OED progression to OSCC. IHC-stained archival tissues [2 groups, age and site matched, e 4 biopsies followed for e 4 years, transformed (n=5) and nontransformed (n=4) to OSCC)] were analyzed by light microscopy and with TissueStudio 3.5 image analysis software. Statistics confirmed comparable initial clinical and histologic features between the transformed and nontransformed groups. Microscopic evaluation revealed greater distribution of moderate to intense IMP3 staining in OED lesions that transformed. Image-analyzed samples revealed that intense IMP3 staining increased over time in those OED lesions that transformed to OSCC (Pearson’s r, p = 0.002, all patients combined). In contrast, no such association was noted in nontransformed OED lesions (p = 0.105). Pearson’s r values (intense IMP3 expression over time) for each individual patient were: transformed group - 0.82, 0.37, 0.99*, 0.92 and 0.72* (*p = 0.015); nontransformed group - 0.11, 0.53, -0.70, 0.57. These data, which show increasing levels of intense IMP3 expression in progressive OED lesions, suggest high sustained levels of intracellular IMP3 contribute to development of an invasive phenotype.
**#15**

**REPRODUCIBILITY OF THE BRANDWEIN VS. BRYNE PREDICTIVE HISTOLOGIC RISK SCORE MODELS IN HEAD AND NECK SQUAMOUS CELL CARCINOMA: A PILOT STUDY**

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Mount Sinai Medical Center, New York; U North Carolina, Chapel Hill; Howard U, Washington; Rambam Medical Center, Haifa; Tufts U, Boston  

**BACKGROUND** The Histologic Risk Model (HRM) is a validated outcome predictor for H&N Squamous Cell Carcinoma (HNSCC). We attempted to determine the reproducibility of the Brandwein and Bryne HRMs in HNSCC resection specimens.  

**STUDY METHODS** Two de-identified coded slides from 10 HNSCC were selected for review by six oral pathologists. A detailed scoring guide for both HRMs was provided. Brandwein criteria include perineural invasion, lymphocytic infiltrate, Worse Pattern of Invasion at interface vs. Bryne criteria of histologic variability, host response, degree keratinization, POI & pleomorphism. Total scores for each HRM were calculated for each OMP. The Cohen's kappa weighted coefficient was calculated for evaluating the agreement between the Bryne and Brandwein scoring methods. Consistency (reliability) between pathologists and intraclass coefficients of correlation were computed separately for each scoring method.  

**RESULTS** Six OMPs completed the study. The weighted Kappa coefficient of agreement was calculated using the online QuickCalcs kappa calculator. Agreement between the two scoring methods was $R=0.216$. The intraclass coefficients of correlation for assessing the consistency of tumor grading among the pathologists were computed using the Medcalc software. Both methods showed a very good inter rater consistency, with the Bryne score showing a slightly lower intraclass coefficient of correlation ($R=92$, 95% CI: 0.80-0.98) than the Brandwein score ($R=93$, 95% CI=0.83-0.98).  

**CONCLUSION** In this pilot study, we compared the reproducibility of the Brandwein and Bryne Histologic Risk score for HNSCC among oral pathologists. A larger study with more cases and additional scorer training is needed to determine if agreement can be sustained or improved.

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**#16**

**OLFACTORY CARCINOMA: A REPORT OF A RARE ENTITY**  
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ORL H&N surgery department and Department of Pathology, Zain & Sabah hospitals and Kuwait Cancer Control Center, Hawally  

Olfactory neuroblastoma is a rare neoplasm that arises from the olfactory membrane in the sinonasal tract. Recently, some cases have been shown to possess a divergent differentiation from a ganglioneuroblastoma to carcinoma, adenocarcinoma or even sarcoma. Herein, we report a rare case of olfactory neuroblastoma with a divergent carcinomatous differentiation in a 49 year old Kuwaiti male. The patient presented with a one year history of nasal obstruction, pressure on the left eye, epiphora, and anosmia. He had a history of head trauma and craniotomy for CSF leak in the 1990s. Endoscopic examination revealed a large bulky mass originating for the skull base and occupying the left nasal cavity, protruding posteriorly into the nasopharynx. CT and MRI studies showed a mass with a well-defined pedicle originating from the skull base with a bony defect. With high suspicion for a meningoencephalocele, though a neoplastic process could not be completely excluded, the mass was excised endoscopically. The histologic section of the mass revealed a small round blue cell tumor with lobular growth pattern in an edematous and somewhat vascular stroma. Pleomorphism, mitotic figures and pseudorosettes were also present in the tumor. Immunohistochemical stains were performed and the tumor was positive for synaptophysin and chromogranin. S100 highlighted the sustentacular cells. Of note, AE1/AE3 cytokeratin was diffusely positive. With a dual phenotype of olfactory neuroblastoma and a carcinoma, this tumor is best described as olfactory carcinoma.
#17

**BIOLOGICAL CONSEQUENCES OF INCISIONAL BIOPSY ON THE PRIMARY TUMOR**

M Donoghue, M Selvamani, KP Mohan, PS Basandi, A Ramakrishna, M Joshi, KSN Siva Bharani

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**Background:** Incisional biopsy (IB) is the most effective means of diagnosing and grading tumors. The proliferative phase lasting 6-7 days post biopsy has been established. In tumors the subsequent healing response can be lacking. Thus, IB can be expected to have some biological consequences on the primary tumor. Objectives: To evaluate the changes in tumor biology post-IB by comparing the IB and surgical resection (SR) tumor specimens.

**Methodology:** SR specimens of squamous cell carcinoma (SCC) and their IB specimens (N=60) with a gap of 6 days or more were retrieved from the archives. H& E stained sections were evaluated for Mean Vascular density (MVD), number of mitosis /10 HPF, atypical mitosis and the type, intensity and distribution of inflammatory cells. Sections were also prepared for estimation of mitotic index using KI 67 staining.

**Results:** Partial results from analysis of H & E stained sections showed statistically insignificant rise of MVD in SR in comparison to IB samples (0.106 ± 0.163 & 0.089 ± 0.145 respectively). The number of mitosis/10HPF was reduced in the SR in comparison to IB samples (4.20 ± 3.6 & 4.43 ± 3.7 respectively). The number of specimens showing normal mitosis was slightly reduced and those with abnormal mitosis slightly increased in the SR samples. Among the inflammatory cells eosinophils showed a significant (p=0.05, P < 0.05) rise in the SR samples. The distribution, nature and severity of the inflammatory response were not significantly altered. Mitotic index is pending completion.

**Conclusions:** This preliminary study suggests that there are some biological consequences of IB on the primary tumor. Larger studies are needed to quantify these consequences and determine their effects in terms of tumor behavior.

#18

**ORAL BIOPSY AS A BEHAVIOR MODIFICATION AGENT IN TOBACCO/ALCOHOL CESSATION**

T Peters, C Phillips, V Murrah

U of North Carolina, Chapel Hill

Tobacco is the predominant etiologic factor for oral cancer (OC). Prognosis for OC has essentially remained unchanged for many years. Moreover, field cancerization increases risk for second primaries. Strategies to decrease etiologic factors are critical. Biopsy results may act as a behavioral change agent. Our objective was to determine whether biopsy diagnoses are associated with risk factor cessation. Surveys were sent to subjects identified in the UNC Oral Pathology database with a diagnosis of hyperkeratosis, dysplasia or carcinoma. Data obtained included demographics, risk factor use and any change in use since biopsy. 1632 questionnaires yielded a 38% response rate. Results from Fisher’s exact test showed a higher quit rate for both cigarettes and alcohol in those with carcinoma versus those with hyperkeratosis (p< 0.01). The same was true for cigarette use in those with dysplasia versus hyperkeratosis (p< 0.03). Subjects with dysplasia or carcinoma were 2.78 times more likely to quit or reduce cigarette use versus those with hyperkeratosis (95% CI=1.38-5.59). Significantly higher cigarette quit rates were seen in males with carcinoma versus those with hyperkeratosis (p< 0.01); this was not true for females. Results convey important implications for patient education. Biopsied patients with premalignant or malignant diagnoses are likely to discontinue etiologic habits; however, there is a need to educate patients with “benign” diagnoses as our study indicates that they are more likely to continue their habits. These patients may have a false sense of reassurance which is not supported by current carcinogenesis statistics. Surgeons can play a role in preventing OC by emphasizing strategies for tobacco/alcohol cessation for all patients whom they biopsy.
#19

**ORAL SQUAMOUS PAPILLOMA, MULTIPLY RECURRENT, IN A PATIENT WITH PSORIASIS RECEIVING THE TNF-ALPHA ANTAGONIST ADALIMUMAB**
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**Background:** Tumor necrosis factor-alpha (TNF\(\alpha\)) is a cytokine secreted by macrophages that regulates various biological processes including cell proliferation, differentiation, apoptosis, lipid metabolism and coagulation. Increased TNF\(\alpha\) has been implicated in autoimmune diseases, insulin resistance and cancer. TNF\(\alpha\) antagonists are known to cause reactivation of many bacterial and viral infections. Adalimumab is a monoclonal antibody that neutralizes TNF\(\alpha\) activity and induces complement mediated cell lysis of mononuclear cells expressing TNF\(\alpha\).

**Objective:** To describe TNF\(\alpha\) antagonists and their role in reactivation of low risk human papilloma virus in the oral cavity.

**Observation:** A 50-year old male presented initially with a squamous papilloma of the midline mandibular gingiva that recurred three times after the initial excision. The medical history revealed treatment for psoriasis with adalimumab, subsequent to the first excision.

Potential causes for persistence or recurrence of the squamous papilloma include: incomplete excision, viral reinfection, viral reactivation in an immunocompetent or immunosuppressed patient, and TNF\(\alpha\) antagonist associated human papilloma virus infection.

**Conclusion:** TNF\(\alpha\) antagonist medications can reactivate latent viral infections or increase susceptibility. Reporting of similar cases is encouraged to further establish a link between TNF\(\alpha\) antagonist drug use and various oral pathologies including infection, inflammation, and neoplasia.

#20

**NEUROEPITHELIAL STRUCTURES ASSOCIATED WITH THE SUBEPITHELIAL NERVE PLEXUS OF TASTE BUDS: A FORTUITOUS FINDING RESEMBLING THE JUXTAORAL ORGAN OF CHIEVITZ. CASE REPORT AND REVIEW OF THE LITERATURE**
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**Background:** Numerous embryologic epithelial remnants have been described in the oral region and when they are intimately associated with peripheral nerves may pose a diagnostic pitfall for pathologists. The literature contains well documented cases in which the juxtaoral organ of Chievitz (JOC) was identified in a surgical specimen removed because of an oral malignancy and correct recognition of this anatomical structure potentially avoided unnecessary treatment to patients. To the best of our knowledge, this is the first report of such a case in which a neuroepithelial structure similar, if not morphologically identical to that of the JOC, was found in the posterior lateral border of the tongue in close association with the subepithelial nerve plexus of taste buds.

**Methods:** Review and interpretation of the English literature pertaining to the juxtaoral organ of Chievitz, subepithelial nerve plexus and embryologic epithelial remnants of the oral region.

**Results:** Embryologic epithelial remnants are normally found in the soft tissues and jawbones of the oral and para-oral regions. These remnants have been found in close association with peripheral nerves.

**Conclusions:** Proper recognition of these anatomical structures is crucial to prevent misdiagnosis of squamous cell carcinoma, or perineural invasion.
PLEXIFORM ENCAPSULATED NEUROMA: ANALYSIS OF CASES WITH SPECIAL EMPHASIS ON THE PLEXIFORM VARIANT  C Fowler, D Damm, D White  U of Kentucky College of Dentistry

Introduction: Palisaded encapsulated neuroma/solitary circumscribed neuroma (PEN) is a benign neural lesion that occurs on skin or mucosa. The plexiform variant and some examples of multilobular PEN may be confused with other plexiform neural lesions such as plexiform neurofibroma (PNF), plexiform neurilemoma (PNL), and mucosal neuromas of MEN 2B (MN).

Objectives: To perform a clinicopathologic analysis of cases of PEN, and to determine if morphology and immunohistochemistry (IHC) can be used to accurately differentiate plexiform PEN from other plexiform neural lesions. Materials/Methods: Cases of PEN, PNF, PNL, and MN from UKCD files were reviewed. IHC was performed on selected cases with S-100, NFP, EMA, GFAP, and CD34. Examples of neurofibroma, neurilemoma, and traumatic neuroma were used as IHC controls.

Results: Of 50 cases of PEN, 31 occurred in males while 19 were found in females. A wide age range was noted (12-86 years, mean=29.5 years) yet half of the lesions occurred in 5th-6th decades. The lip was the most common site (18), followed by hard palate/gingiva (17), head/neck skin (6), soft palate (4), tongue (3), and buccal mucosa/vestibule (2). PEN cases consistently exhibited fascicular arrangements of Schwann cells and axons. Artifactual clefting was identified in 41 cases. Myxoid change was never prominent and focally identified in only 4 cases. 36 cases appeared lobular, 12 cases were multilobular, and 2 were plexiform. All tested cases of PEN were diffusely positive for S-100 protein, NFP highlighted variable numbers of axons, and EMA marked a thin investing perineurium. GFAP expression was negative in both plexiform PEN cases and was variable among the other neural lesions. CD34 staining also varied and was unreliable in discriminating PEN from the other lesions. The pattern and intensity of S-100 protein reactivity was most helpful in separating plexiform PEN from PNF: strong, diffuse S-100 expression was found in PENs while more modest reactivity was seen in PNLs. NFP confirmed the presence of axons in plexiform PEN cases. MNs displayed a thick, EMA-positive perineurium, showed variable numbers of axons with NFP, and were positive for GFAP. Conclusions: IHC analysis with S-100, NFP, EMA, and GFAP along with careful attention to morphologic features should allow accurate diagnosis of plexiform PEN. Correlation with clinical appearance and family history also is recommended, especially in problematic cases.

ECTOMESENCHYMAL CHONDROMYXOID TUMOR: A SERIES OF 5 CASES  A Aldojain, J Jaradat, K Summersgill, E Bilodeau  U Pittsburgh, Pittsburgh

Ectomesenchymal chondromyxoid tumors (ECT) are rare, benign, intraoral mesenchymal soft tissue tumors, with only 44 cases reported in the English literature. We herein report our experience with 5 ECT and review the immunohistochemical and clinicopathologic features of this rare entity. The mean age of patients was 42 years (range 7-53 years), with no gender predilection. All tumors were located on the dorsal tongue. The pathologic differential diagnosis of the ECT was myoepithelioma, nerve sheath myxoma and cellular neurothekeoma. Histologically, all tumors exhibited a lobular proliferation of ovoid to round cells with cellular pleomorphism, a net-like growth pattern, and slit-like cystic spaces, with at least a focally myxoid background (5/5, 100%). Muscle entrapment was seen in 3 cases (3/5, 60%). Fine calcifications and multinucleated giant cells were noted in two cases (2/5, 40%). Immunohistochemically, the tumors were positive for S-100 (5/5, 100%), glial fibrillar acidic protein (GFAP) (4/5, 80%), focally to diffusely positive for SMA (4/5, 80%), with rare P63 expression (2/5, 40%). The extracellular myxoid material was positive for mucicarmine (2/2, 100%). Calponin and AE1/AE3 were consistently negative (4/4, 100% and 3/3, 100%, respectively). Some cases exhibited rare positivity for CD57 (2/3, 66%), CD68 (1/2, 50%), and focal positivity for CAM 5.2 (1/5, 20%).
#23
EVALUATION OF ANGIOGENESIS IN CENTRAL AND PERIPHERAL GIANT CELL GRANULOMAS OF THE JAWS AND ORAL CAVITY  
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Background and objective: Peripheral giant cell granuloma (PGCG) and central giant cell granuloma (CGCG) are two pathologic lesions with similar histopathologic features. PGCG is a reactive lesion, while controversies exist regarding the true nature of CGCG (reactive vs. neoplasm). The aim of this study was to evaluate the angiogenic potential of these lesions using CD105 immunostaining and MVD count. 
Materials and methods: In this descriptive study, 30 cases from each lesion were selected and 4µ sections were stained with CD105 antibody. Microvessel density (MVD) was evaluated by counting vessels in areas of highest vascularity (hot spots) using light microscopy at 400X magnification. 
Results: All cases in both groups showed immunoreactivity with anti-CD105 antibody. MVD was not significantly different between CGCG and PGCG groups (P=0.390). Similar findings were observed comparing peripheral supportive stroma and central mass in PGCG group (P=0.402). In CGCG group, no significant relation existed between MVD and clinical signs (or symptoms) (P=0.317) or cortical perforation (P=0.434). 
Conclusion: Based on our findings, there was no difference between PGCG and CGCG regarding microvessel density and angiogenesis which could suggest a similar pathogenesis for these lesions.

#24
CASE REPORT: METASTATIC LUNG CARCINOMA PRESENTING AS VASCULAR SWELLING OF THE MAXILLARY GINGIVA  
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We report the case of a 80 year old female who presented with a two week history of a bulky and vascular appearing swelling of the maxillary gingiva which involved the facial and palatal gingiva of teeth # 2 and 3. The swelling was in close approximation to implants that were supporting a fixed partial denture. There was no history of pain. Our differential diagnosis included reactive lesions (pyogenic granuloma, peripheral ossifying fibroma and inflammatory fibrous hyperplasia), hemangioma and malignant neoplasm (due to extension of the lesion to the facial and palatal gingiva). The lesion was biopsied and hematoxylin and eosin stained sections revealed a soft tissue specimen composed of a nodular mass of mucosa. The surface was lined by stratified squamous epithelium and appeared cytomorphologically bland. Within the underlying lamina propria a proliferating malignant epithelial neoplasm was seen which involved the margins. The proliferating epithelium exhibited a somewhat nest like growth pattern. The nuclei were very large with a vesicular pattern and large prominent pink nucleoli. Mitotic activity was extraordinarily brisk and atypical mitotic spindles were easily identified. The proliferating neoplasm was highly infiltrative and evidence of ductal differentiation was observed in the deepest portions. Our impression was metastatic neoplasm suggestive of lung carcinoma. Immunohistochemical staining with ttf-1 was performed to confirm the diagnosis, which was subsequently reconfirmed clinically.
#25

THE INCREASED P16 AND CYCLIN D1 EXPRESSION IN RECURRENCE OF SALIVARY GLAND PLEOMORPHIC ADENOMA

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Pleomorphic adenoma (PA) is the most common salivary gland tumor. Although classified as a benign tumor, the incidence of recurrence after initial surgical treatment is significant and varies largely due to differences in surgical technique. Furthermore, recurrent pleomorphic adenoma (RPA) has been associated with an increased risk of malignant transformation. However, few studies were found on RPA, its biological behavior and risk factors. Recently, cell cycle markes have been receiving increasing attention, with regard to their importance in biological behavior tumor. The aim of the present study was to investigate the participation of the cell cycle markers: p16, cyclin D1 and retinoblastoma (Rb) in PA, RPA and RPA with malignant transformation (RPAT). Twenty four cases of PA, 21 cases of RPA and 2 cases of RPAT were studied. The immunohistochemical reactions to p16, cyclin D1 and Rb were evaluated. Expression scores were assigned according to the percentage of positive nuclear tumor cells, from 0 to 3 (0, less than 10%; 1, 10-25%; 2, 25-50%; 3, staining of more than 50% of cells). The Mann-Whitney test was used to compare different tumor groups. The majority of the PA cases showed negative or weak p16 and cyclin D1 expression, whilst in the majority of the RPA cases, as well as in the two TRPA cases, strong expression of these proteins was demonstrated. Regarding Rb, all groups (PA, RPA and TRPA) were shown to be negative or to have weak expression. In conclusion, this study may suggest that the p16, cyclin D1 and Rb pathway were not affected in PA, whilst in RPA and TRPA, the p16 and cyclin D1 pathways were altered, indicating that those proteins are probably important in the development of recurrence of pleomorphic adenoma and malignant transformation.

#26

POLYMORPHOUS LOW GRADE ADENOCARCINOMA OF THE UPPER LIP WITH METACHRONOUS MYOEPITHELIOMA OF THE BUCHAL MUCOSA. CASE REPORT AND LITERATURE REVIEW OF SYNCHRONOUS AND METACHRONOUS MINOR SALIVARY GLAND TUMORS

I. Koutlas, P Argyris, S Pumbuccian, K Tosios, R Gopalakrishnan

Examples of synchronous and metachronous minor salivary gland tumors (MSGTs) are uncommon. We report of a patient who initially presented with polymorphous low-grade adenocarcinoma (PLGA) and subsequently with myoepithelioma (MYO). A 91-year old Caucasian female presented in 2009 with a 1 cm, firm, non-tender, well-circumscribed nodule of the left side of the upper lip extending to the anterior buccal mucosa. Excisional biopsy revealed PLGA. While the margins were positive, further treatment was not recommended due to the patient’s age. In 2011, the patient returned with a 1.5 cm asymptomatic mass of the left buccal vestibule. Excision of the lesion revealed a circumscribed proliferation of ductal and plasmacytoid cells arranged in spherical or whorl-like islands and immersed in a mucinous stroma, consistent with MYO. The PLGA recurred 3 years after initial diagnosis. Excision was again associated with positive margins and again no further treatment was recommended. A few months later, at a scheduled follow-up appointment, she presented with a painless nodule of the left upper lip, consistent with recurrent PLGA. One month later, the patient died of unrelated causes. We also present a review of the literature regarding MSGTs.
FAMILIAL CHERUBISM WITH ODONTOGENIC TUMOROUS PROLIFERATIONS
Koutlas, Y Hu, E Reichenberger, D Primley, R Gopalakrishnan U Minnesota, Minneapolis; U of Connecticut; Private Practice, Oral Surgery, St. Cloud Cherubism is a rare autosomal dominant condition affecting the jaws that maps to chromosome 4p16 with mutations identified in the gene encoding c-Abl-binding protein SH3BP2. The lesions of cherubism have been well characterized radiographically and histopathologically. Generally, lesions are multilocular and expansile and feature vascular fibrous tissue in association with multinucleated giant cells. Herein, we describe a family with cherubism, two members of which, in addition to giant cell lesions presented with odontogenic tumorous proliferations, one, the son and proband at age 25, with central odontogenic fibroma-like features, the other, his mother, at age 57, with primary intra-osseous odontogenic carcinoma and areas of benign fibro-osseous lesion. In both patients the lesions occurred in the mandible and presented with unusual for cherubism focal enlargement. The son underwent incisional biopsy and did not have additional treatment. His mother underwent extensive mandibulectomy due to widespread tumor. The son has two affected children while a third child is at age 5 and has not shown yet any features of the disease. Mutation analysis affected members of the family resulted in the identification of heterozygous mutation in SH3BP2 is c.1244G>C (p.Arg415Pro). To the best of our knowledge, association of cherubism with odontogenic lesions has not been reported in the literature. This association further supports the theory linking cherubism with disturbed odontogenesis.

COMBINED ODONTOGENIC TUMORS: A REPORT OF 3 CASES
A Neuman, I Bhattacharyya, D Cohen, C Dunlap U Florida Combined odontogenic tumors have rarely been reported in the literature. We present three new cases exhibiting distinctly separate histologic features including: ameloblastoma with ameloblastic fibro-odontoma (AFO), ameloblastoma with odontogenic keratocyst (OKC), and calcifying odontogenic cyst (COC) with ameloblastic fibroma (AF). We review the histopathology of these unusual lesions and discuss relevant literature. A search of archival material of University of Florida’s Biopsy Service revealed only 3 such lesions among the 72,171 specimens submitted from 2004-2012. The location, radiographic and clinical findings, and histology of these lesions are presented. Lin et al, Seim et al and others believe that hybrid odontogenic lesions are not a result of collision between two distinct entities but rather due to the pluripotentiality of the odontogenic epithelium with both lesions likely developing from a common epithelial source. In addition, though we have in our archives a combined adenomatoid odontogenic tumor (AOT) with calcifying epithelial odontogenic tumor (CEOT), we did not include this lesion as many authors believe that CEOT-like areas often occur in AOTs and therefore the combined AOT-CEOT should be considered a variant of AOT. A review of the English language literature revealed examples of “hybrid” odontogenic lesions including 6 cases of COC with AF and a few other rare combinations. Brannon noted two OKCs with ameloblastomatous change. To the best of our knowledge no case of concurrent AFO with ameloblastoma has been reported. Treatment of these lesions is typically based on appropriate treatment for the more aggressive component.
MULTIFOCAL CALCIFYING EPITHELIAL ODONTOGENIC TUMOR: A CASE REPORT AND REVIEW OF THE LITERATURE
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The development of multifocal calcifying epithelial odontogenic tumors (CEOTs) is most unusual, with only 5 cases reported thus far in the English literature. Here we present an additional case. A 24-year old female complained of wisdom tooth pain, and radiographic evaluation showed 2 lesions. The first was associated with the crown of an impacted mandibular third molar and appeared as a radiolucency with radiopaque flecks. The second appeared as a small radiolucency along the mesiolateral aspect of a mandibular second premolar. Among the present case and the 5 previously reported cases, there are 15 total lesions (4 extraosseous, 11 intraosseous). All except one patient had synchronous lesions. The average age was 42 years (range of 24 to 55 years). The lesions were located in the posterior mandible (n=6), posterior maxilla (n=5), anterior maxilla (n=2), and anterior mandible (n=2). Clinical findings included swelling (n=2), pain (n=1), and tooth displacement (n=3). Most of the intraosseous lesions presented as well-defined, mixed radiolucent/radiopaque lesions, although 4 were entirely radiolucent. Most cases exhibited typical histopathologic features of CEOT, except for one which exhibited foci of squamous odontogenic tumor. The preferred treatment was conservative enucleation, with recurrent lesions typically managed by curettage or removal of a rim of normal tissue. Follow-up information was provided for 5 patients. One had an intraosseous lesion recur twice, at 7 and 17 years after initial presentation; another patient had two peripheral lesions that recurred after 1 year. In all cases, these multifocal tumors appear to represent isolated findings, with no known syndromic association or familial tendency.

NON-CALCIFYING LANGERHANS CELL ASSOCIATED EPITHELIAL ODONTOGENIC TUMOR
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The calcifying epithelial odontogenic tumor (CEOT) is a benign, locally aggressive lesion, representing less than 1% of all odontogenic tumors. Most cases are intraosseous with a predilection for the posterior mandible, often associated with an impacted tooth. Characteristic microscopic features include sheets and cords of variably pleomorphic, polygonal epithelial cells with accumulations of eosinophilic amyloid-like material and spherical laminated calcifications. Alternate tumor presentations have been described, which include a clear cell variant, a cystic variant and a variant showing cementum or bone like components. There is a further rare variant that is characterized by lack of calcification and association of the epithelial component with Langerhans cells. To date, there have been 3 well documented cases, all involving the anterior maxilla in the canine/bicuspid area. We present a fourth case of the non-calcifying Langerhans cell associated (NLCA) variant, which involved the left anterior maxilla of a 53 year old Caucasian female. The lesion extended from the interproximal bone between the lateral incisor and canine into palatal bone. The initial conservative surgical management with curettage was unsuccessful and after one year, there was a recurrence which extended to the sinus. This is the first reported recurrence of the NLCA variant. In this regard, the biologic behavior of non-calcifying CEOTs is not well defined with suggestions that the lack of calcification is indicative of more aggressive behavior and alternately, that non-calcifying cases associated with Langerhans cells are less aggressive. The limited NLCA literature is reviewed with emphasis on comparison to immunologic findings in other CEOT types.
CELLULAR ATYPICAL CALCIFYING EPITHELIAL ODONTOGENIC GHOST CELL TUMOR: A REPORT OF AN UNUSUAL CASE  J Wollenberg, M Markoff, P Freedman  New York Hospital Queens, Flushing; Private Practice, Randolph  Calcifying epithelial odontogenic ghost cell tumor (dentinogenic ghost cell tumor) is a locally invasive, solid neoplasm once believed to be the solid variant of calcifying odontogenic cyst. In some cases, the tumor may cause bony expansion or displacement of teeth, but it is usually asymptomatic and discovered during routine radiographic examination. Radiographically, it presents as a well- or poorly-defined radiolucency with variable amounts of radiopaque calcification. Histologically, the calcifying epithelial odontogenic ghost cell tumor is composed of infiltrating sheets and islands of odontogenic epithelium in a mature connective tissue stroma. A characteristic feature is the transformation of epithelial cells to ghost cells, some of which undergo calcification. Calcifying epithelial odontogenic ghost cell tumor has a high recurrence rate following enucleation. Therefore, resection with an adequate disease-free margin is recommended. The current case is that of a 91-year-old female who was referred to an oral surgeon by her general dentist for evaluation of a radiolucency in the #22 area in 2010. At that time, a biopsy was performed and diagnosed as cellular atypical calcifying epithelial odontogenic ghost cell tumor. Due to multiple co-morbidities the patient decided against undergoing any further treatment. Over the next two years, imaging studies show progressive destruction of the mandible. For the first time, the natural course of a calcifying epithelial odontogenic ghost cell tumor has been documented through serial radiographic studies without any surgical treatment.

EWSR1 REARRANGEMENT IN CLEAR CELL ODONTOGENIC CARCINOMA: REPORT OF A CASE  A Yancoskie, C Sreekantaiah, A Rosenberg, J Fantasia  Hofstra North Shore-LIJ School of Medicine, Hempstead  Background: The Ewing sarcoma breakpoint region 1 (EWSR1) is the most frequently involved gene in translocations in sarcomas and several different gene partners have been documented. The translocations resulting in EWSR1-CREB1 and EWSR1-ATF1 gene fusions have been recurrently described in several neoplasms that are histopathologically and behaviorally different and include angiomatoid fibrous histiocytoma, clear cell sarcoma, hyalinizing clear cell carcinoma of salivary gland, and more recently soft tissue myoepithelioma and clear cell odontogenic carcinoma (CCOC). Objective: To evaluate a case of CCOC for EWSR1 rearrangement. Observation: A 59 year-old woman presented with paresthesia of the left lower lip and a radiolucency in the left body of the mandible. Upon biopsy a diagnosis of CCOC was rendered and treatment consisted of segmental mandibulectomy. Formalin fixed paraffin embedded tumor tissue was evaluated using fluorescence in situ hybridization (FISH) for the presence of EWSR1 rearrangement with the EWRS1 dual color break-apart probe. Results: FISH analysis of this CCOC demonstrated EWSR1 rearrangement in >20% of cells.
GLANDULAR ODONTOGENIC CYST: REPORT OF A SERIES OF 24 CASES  JC Whitt, BF Barker, TM Gibson, CL Dunlap  University of Missouri Kansas City, Kansas City  The glandular odontogenic cyst (GOC) is an uncommon odontogenic cyst that exhibits a high rate of recurrence when conservatively excised. A recent report of a large series of GOCs indicated a recurrence rate of 50%. Since its original description as sialo-odontogenic cyst in 1987, over 150 cases have been reported in the English language literature. We report a series of 24 cases of GOC arising in patients ranging in age from 29 to 79 years with an average age at surgery of 50 years. Seventy-five percent of the lesions arose in the mandible, with 42% (10/24) located in the anterior mandible. Overall, 63% (15/24) percent of the lesions were located in the anterior portion of the jaws. There was a strong male (2:1) gender predilection. Twenty-one (5/24) percent of the lesions were located in the dentigerous position. The remainder presented as radiolucencies associated with the roots of teeth, ranging in size from 2.0 to 6.0 cm. The recurrence rate of lesions in this series, based in information available in surgical pathology reports, was 21% with two lesions recurring three or more times over a 10 year follow-up period. A variety of microscopic features have been identified in GOC, including cobblestoned surface eosinophilic cells, microcysts, apocrine snouting, clear cells, variable thickness epithelial lining, papillary tufting, mucous goblet cells, plaque-like thickenings, epithelial spheres, cilia and multiple compartmentation. These cases exhibited many, but rarely all of these histologic features. The differential diagnosis of GOC includes lateral periodontal cyst, botryoid odontogenic cyst, odontogenic cyst with mucous metaplasia, surgical ciliated cyst and low-grade intra-osseous mucoepidermoid carcinoma.

DENTINOAMELOBLASTOMA, A RARE ODONTOGENIC TUMOR: REPORT OF TWO CASES WITH REVIEW OF LITERATURE M Alqahtani, DM Cohen, MN Islam, I Bhattacharyya  U Florida, Gainesville  Dentinoameloblastoma (DA) is a rare odontogenic tumor characterized by classic ameloblastoma-like areas with unusual induction of dentinoid by the neoplastic odontogenic epithelium without any evidence of enamel matrix or tooth formation. We present two cases of DA presenting as expansile radiolucent lesions of the maxilla. Microscopic examination of the biopsy samples revealed proliferative ameloblastoma-like areas along with numerous foci of eosinophilic partially calcified dentinoid material. The term DA has been in use since it was first defined by the WHO in 1970 and has been frequently confused with ameloblastic odontoma/odontomaameloblastoma. In DA, no enamel matrix is seen but presence of dentinoid is consistently noted, with occasional cases also exhibiting cementum-like material. Moreover, in our two cases the tumor presented with numerous gland-like structures with dentinoid material and in focal areas bore strong resemblance to an adenomatoid odontogenic tumor (AOT). We identified only 6 additional cases of unequivocal DA in the literature which have also been termed by some authors as adenoid ameloblastoma with dentinoid. Most cases previously reported as DA did not meet the histologic criteria laid down by the WHO. The dentinoid material seen in DA is believed to be an induction effect of the proliferative ameloblastic epithelium on the mesenchymal tissue. Notably, these lesions exhibit aggressive biologic behavior comparable to conventional ameloblastoma thereby warranting similar treatment. Importantly, the combination of a calcified product so unusual in ameloblastomas, glandular AOT-like (adenoid) areas and maxillary location may lead to a misdiagnosis of AOT if limited clinical information and only a small biopsy sample are available.
**#35**

**DENTINOMA: A REPORT OF TWO CASES**

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Dentinoma is a rare odontogenic tumor. Four histologic variants have been reported: immature dentinoma (containing tubular dentine, dysplastic dentine or dentinoid and odontogenic epithelium), mature dentinoma (immature dentinoma without epithelium), ameloblastic fibro-dentinoma (ameloblastic fibroma-like tumors with tubular dentine or dentinoid) and adenomatoid dentinoma. We present two cases of dentinoma. The first patient was an 8-year old male with an ill-defined multilocular radiolucent/radiopaque lesion overlying unerupted teeth #13 and #14. The lesion was curetted. Histopathologically, the biopsy contained abundant hyalinized, eosinophilic material in a vaguely lobular configuration, with entrapped islands of benign odontogenic epithelium as well as stellate and spindled cells. The odontogenic epithelium was present in small and large islands, and strands. Two years later, there was a persistent radiolucency at the site and this area was re-curetted. This showed typical deposits of dentinoid with entrapped odontogenic epithelium and stellate and spindled cells. Tubular dentine, enamel matrix and ghost cells were not identified in either specimen. The second patient was a 12-year old female with a well-defined radiolucent lesion in the mandible associated with the crown of an unerupted left second molar. The histopathology was identical to the re-curetted lesion from Case 1. The nature of dentinoid is still poorly understood; it is likely a product from an abnormal epithelial-mesenchymal interaction of tooth development. Ameloblastic fibrodentinoma and dentinogenic ghost cell tumor are found in the category of “odontogenic epithelium with odontogenic ectomesenchyme” under the WHO classification. The category under which dentinoma belongs is still controversial.

**#36**

**MULTIPLE ORTHOKERATINIZED ODONTOGENIC KERATOCYSTS IN A TWENTY-THREE YEAR OLD MAN**

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A 23-year-old Hispanic male presented with four radiolucent lesions around the impacted third molars in all four quadrants. A biopsy was performed for three of the lesions around teeth #1, 17 and 32, and the diagnoses were orthokeratinized odontogenic keratocysts (orthoOKC) for all three. The cystic lesion around tooth #16 was marsupialized. To the best of our knowledge, a case of multiple orthoOKCs has never been reported in the literature. As loss of heterozygosity of the PTCH gene has recently been reported in some sporadic orthoOKC cases, a patient with multiple orthoOKCs raises the possibility of being a nevoid basal cell carcinoma syndrome patient. Frontal bossing, multiple skin cysts and pigmented lesions were found clinically on this patient. Other family members also showed some of these clinical features. The investigation on the clinical and radiographic features of nevoid basal cell carcinoma syndrome is still ongoing. A genetic test for PTCH gene sequencing was suggested, but has not been performed to date.
PERIPHERAL CALCIFYING CYSTIC ODONTOGENIC TUMOR (SOLID TYPE): TWO CASES AND REVIEW OF LITERATURE

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WHO 2005 has classified calcifying cystic odontogenic tumor (CCOT) and dentinogenic ghost cell tumor (DGCT) as mixed odontogenic tumors with or without hard tissue formation. Both these lesions occur as an intraosseous or extraosseous process at any age. CCOT occurs in either sex, in the canine-incisor area while DGCT is more common in males in the canine-molar area. Most of the intraosseous lesions of CCOT and DGCT are described as well-defined unilocular radiolucencies while extraosseous lesions may show saucerization. The histopathology of these lesions show ghost cells, odontogenic epithelial islands similar to ameloblastoma, epithelial cells transforming into ghost cells with focal areas of calcification and dentinoid. Review of the archives of the oral biopsy service at UNMC, College of Dentistry NE, revealed 2 lesions in the gingiva presenting as gingival nodules. The first patient was a 32 year-old female with a firm asymptomatic nodule in the anterior maxilla while the second was a 67 year-old male with an ulcerated firm nodule in the mandibular premolar area. Histologically, features of odontogenic epithelium similar to ameloblastoma, ghost cells, areas of calcification and dentinoid were noted. These lesions were reported as CCOT (solid type). The literature reports similarities between CCOT and DGCT resulting in confusion regarding nomenclature, treatment and prognosis. It appears that these lesions could be combined as DGCT with or without cystic transformation. These cases are presented to clarify the nomenclature and to suggest a reclassification of these lesions.

LOBODONTIA: REPORT OF A FAMILY WITH A RARE INHERITED DENTAL ANOMALY

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Lobodontia is a very rare, autosomal dominant dental dysmorphology that may involve some or all of the teeth. Microdontia, hypodontia, shovel-shaped incisors, conical premolars and canines with accentuated cusps, multituberculate molars with single conical roots, and dens invaginatus have been described in association with this condition. These changes were suggested to have some similarity to the canid (lupine) dentition, resulting in designation of the condition as “lobodontia”. Three teenage sisters were referred to Dental Faculty Practice for evaluation of their teeth. Their father was edentulous due to the removal of his “abnormal” teeth in his teenage years. Intraoral examination of the 2 younger girls showed multiple retained deciduous canine and molar teeth, hypodontia and multituberculate molars with pronounced, elongated cusps, resulting in “rosette-like” occlusal surfaces. Radiographs revealed multiple impacted canine and premolar teeth with pointed cusps. While most of the anterior dentition of the oldest sibling was well-formed and complete, her multituberculate molars had marked occlusal attrition. The molar teeth each had a single root and a single large pulp chamber. Based on the clinical and radiographic findings, as well as the family history, the diagnosis of lobodontia was made. The hereditary nature of this condition was discussed with the family and continued management with the pediatric dentist was recommended. The findings in our patients were similar to those described in the handful of previous reports of lobodontia.