a significant mediator of cytokine (ie, interferon, tumor necrosis factor-α) production. Additionally, in previous studies unrelated to pSS, ETS1 was shown to regulate STAT4 expression at the transcription level. Thus, we hypothesized that ETS1 plays a significant role in regulating STAT4 transcription within SGECs of patients with pSS. Our objectives were as follows: (1) to determine whether downregulation of ETS1 mRNA by siRNA knockdown results in decreased mRNA and protein expression of STAT4 within the salivary gland cell line A253 and (2) to demonstrate the regulation of STAT4 mRNA expression by ETS1 siRNA knockdown in cultured SGECs from control patients with pSS and non-pSS sicca.

Methods: The salivary gland cell line, A253, was transfected with siRNA targeting ETS1 or a scrambled siRNA control. After 48 hours, mRNA and protein were isolated. cDNA synthesis was performed using 1 μg of mRNA. Expression of ETS1 and STAT4 mRNA was quantified by quantitative reverse transcription−polymerase chain reaction using the delta cycle threshold method to determine the relative fold changes normalized to glyceraldehyde 3-phosphate dehydrogenase in triplicate experiments. In addition, downregulation of STAT4 protein by ETS1 siRNA was determined by Western blot analysis and semiquantified by densitometry using cofilin as the loading control. SGECs were cultured from labial salivary gland biopsies of 2 patients with pSS (focus score [FS] $\geq$1) and 4 patients with non-pSS sicca (FS = 0, n = 2; 0 < FS < 1; n = 2) and transfected with siRNA targeting ETS1 or a scrambled siRNA control. After 48 hours, mRNA was isolated for cDNA synthesis and used to determine the mRNA expression of ETS1 and STAT4 as described above.

Results: In A253 cells, STAT4 mRNA expression was changed by −2.15-fold, on average, when treated with siRNA targeting ETS1. STAT4 protein expression was measured by Western blot analysis 48 hours post-ETS1 siRNA knockdown showing an average −1.25-fold change in A253 total cell lysate. SGECs cultured from patients with lymphocytic infiltration (ie, FS >0) showed a significantly higher expression of STAT4 (P = .0485) than patients with non-pSS sicca with FS = 0. However, we did not find a significant change in STAT4 mRNA expression (average, +1.14-fold) in cultured SGECs treated with siRNA targeting ETS1.

Conclusions: STAT4 is overexpressed by cultured SGECs in patients with pSS and those without pSS with focal lymphocytic infiltrates. STAT4 overexpression in SGECs of patients with pSS and those without pSS could be a result of inflammatory cytokine exposure by neighboring cells or effects of infiltrating lymphocyte exposure. Thus, our results suggest a separate regulatory mechanism of STAT4 in pSS bypassing ETS1 regulation, which might be due to pathogenic pathways involving other transcriptional regulation.

Case Report Award Presentations

Each year, quality case report abstracts submitted by students/residents/fellows are selected to receive case report awards. There are 5 award recipients this year:

OSTEONECROSIS OF JAW SECONDARY TO STEM CELL TRANSPLANT

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Background: Osteonecrosis of the jaw (ONJ) is necrosis of mandibular or maxillary bone, which may lead to bone exposure, regardless of the etiology. It is thought to be attributed to insufficient blood supply and altered bone turnover, resulting in a disrupted repair process and eventually leading to the collapse of bone in the setting of microfractures. We report what is, to the best of our knowledge, the first case of ONJ secondary to stem cell transplant (SCT).

Case Summary: A 69-year-old woman with a medical history significant for hypertension, osteopenia, and myelodysplastic syndrome, status post-haploidentical SCT in March 2018, presented for evaluation of asymptomatic exposed bone of the maxilla and mandible. She had no history of radiotherapy to her head and neck; however, she had undergone total body irradiation of 2 Gy in February 2018 as part of her conditioning regimen. She had no history of exposure to a bisphosphonate, receptor activator of nuclear factor-κB ligand inhibitor, antiangiogenic medicine, or corticosteroids. Her current medications included amlodipine, metoprolol, omeprazole, tacrolimus, sulfamethoxazole-trimethoprim, acyclovir, cholecalciferol, and folic acid. She did not have gingivitis or active periodontal disease. The patient had developed exposed necrotic bone sequestra in several areas of the buccal aspect of her maxillary and mandibular gingiva 2 months after her transplant in May 2018. The pathology report indicated fragments of necrotic bone with acute inflammation and bacterial overgrowth consistent with osteonecrosis.

Conclusions: There is evidence that the microenvironment of the marrow stromal system is severely and irreversibly damaged after SCT. The deficit in the quantity and quality of osteoblastic progenitors may compromise the ability to regenerate a normal osteogenic cell population, leading to an abnormality in bone remodeling/turnover. Although osteonecrosis of the appendicular skeleton is a common complication after SCT, there have been no reports of association with ONJ. Particularly interesting is the pattern of ONJ presentation in all the 4 quadrants in this patient.

OSTEONECROSIS OF MANDIBLE SECONDARY TO SICKLE CELL CRISIS

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Background: Sickle cell disease (SCD) is an autosomal recessive hemoglobinopathy causing amino acid substitution in the B-hemoglobin chain of erythrocytes. SCD affects 1 in 500 African American children. SCD is associated with multiorgan morbidity and significant reduction in life expectancy. Vaso-occlusive crisis associated with SCD is characterized by acute pain and local tissue hypoxia and necrosis. The extremities, back, and chest are frequently affected, and jaw involvement is very rare. To the best of our knowledge, only 3 cases of jaw osteonecrosis secondary to SCD have been reported in the English-language literature.

Case Summary: A 38-year-old African American man with a history of SCD and avascular necrosis of the hips presented to an emergency department with severe pain in his back, legs, and chest due to a vaso-occlusive episode precipitated by physical overexertion. A few days later, the patient complained of left jaw pain and paresthesia. Computed tomography (CT) of the head with intravenous (IV) contrast, maxillofacial CT with IV contrast, and the remainder of the neurologic examinations were unremarkable. The oral medicine service

was consulted, and physical examination of the patient demonstrated lingual gingival tissue swelling of the lower left molars. The mass was nonfluctuant with ulceration causing malalignment of tooth number 18. Bone biopsy revealed squamous mucosa with hemorrhagic foci, ulceration, and ectatic blood vessels with fragments of necrotic bone consistent with osteonecrosis. At the patient's 2-week follow-up visit, a small area of exposed necrotic bone was noted in the affected area. Maxillofacial CT without IV contrast showed mixed radiolucency and sclerosis with dehiscence along the lingual cortex of the posterior mandibular body and along the cortex of the retromolar trigone. Chlorhexidine gluconate 0.12% oral solution was prescribed, and debridement of the left posterior mandible and extraction of the lower left molars with local flap reconstruction were recommended by an oral and maxillofacial surgeon. The patient did not return for follow-up management.

Conclusions: Acute pain and chronic vasculopathy are significant complications of SCD. Jaw involvement in SCD is very rare, as illustrated in this case. It is important for oral health care professionals to understand the pathophysiology and clinical manifestations of SCD.

THE FIRST REPORTED CASE OF PROLIFERATIVE FASCIITIS IN THE ORAL CAVITY

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Background: Proliferative fasciitis is a benign and reactive lesion involving fibroblasts in the subcutaneous tissues and deep fascia, with a rare occurrence in the head and neck region. It is considered a variant of nodular fasciitis, which could involve trauma as an etiology. Proliferative fasciitis mostly occurs in adults, but cases in children have been reported. Clinically, it can present as an aggressive lesion with pain or no symptoms, mimicking sarcomas.

Case Summary: We report a case of a 53-year-old woman who presented to our clinic for an evaluation of a reddish lesion of the right mandibular gingiva around the molar areas with a relatively rapid onset. The patient had prediabetes with moderate oral hygiene and no other significant medical history. Intraoral examination revealed a poorly circumscribed gingival lesion on the posterior, <1 cm in diameter with no pain on palpation, and soft to firm in texture with no bleeding. A biopsy of the lesion was performed for histologic examination, and the microscopic differential diagnosis included benign and malignant spindle cell tumors; thus, immunohistochemistry was performed for more accurate diagnosis, and a specimen was sent to the pathology lab at Ohio (Central Ohio Skin and Cancer). The immunohistochemical findings were positive for vimentin and smooth muscle actin and negative for CD34, S100, and pancytokeratin. The lesion was diagnosed as proliferative fasciitis on the basis of histologic and immunohistochemical features. The feature that differentiates proliferative from nodular fasciitis is the basophilic component that closely resembles ganglion cells without Nissl substance. The treatment rendered was conservative surgical excision with 1-year follow-up, and no recurrence was observed.

Conclusions: Because proliferative fasciitis has not been reported in the oral cavity, to our knowledge, and because it poses a diagnostic challenge and can mimic malignancies, it is essential to know the salient diagnostic features to avoid aggressive treatment in patients presenting with such lesions in the oral cavity.

MASSON TUMOR OF THE LINGUAL TONGUE: A CASE REPORT

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Background: Intravascular papillary endothelial hyperplasia, or Masson tumor, is a benign lesion of the head and neck region. The etiology of the lesion arises within a blood vessel and is thought to be reactive and associated with vascular injury. Masson tumors comprise approximately 2% of all vascular tumors of skin and subcutaneous tissues; however, this is rarely seen intraorally. It is important to consider at the time of differential diagnosis to distinguish from malignancy and avoid aggressive surgery or unnecessary treatment.

Case Summary: We describe a case of a patient who presented to the Erie County Medical Center Department of Oral Oncology for evaluation of a soft, nontender, mobile mass in the right side of the ventral tongue. The patient first presented in June 2019 with an approximately 5-mm round mobile mass on the right side of the ventral tongue of 6 days' duration. The patient opted for no treatment in June 2019 and returned in December 2019 after the mass had grown in size and had begun to affect his everyday activities. Treatment options included excisional biopsy under general anesthesia or under local anesthesia. The patient opted for excision under local anesthesia. The vascular component was identified and tied off, and the tumor was removed in total. The tumor was a bluish lesion with a thick intact capsule. The final pathology revealed a thrombosed blood vessel with papillary endothelial hyperplasia consistent with Masson tumor. Immunostains for CD31 and D2-40 supported this diagnosis. The patient has some residual tethering of the right side of the tongue resulting from establishing primary closure.

Conclusions: The majority of tumors with this diagnosis have an excellent prognosis with complete excision. Malignant transformation and metastasis have not been reported.

"RINGLIKE HARD MASS" SURROUNDING THE ROOT OF A PRIMARY TOOTH IN A YOUNG CHILD: REPORT OF AN UNUSUAL CASE

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Background: Several lesions of diverse origin may be detected in the oral cavity of young children, affecting the oral mucosa, jaws, or teeth. Their clinicopathologic features may show considerable overlap. We present an interesting case of a "ringlike hard mass" of initially unknown nature around the cervical area of a primary tooth in a young child, and we discuss the diagnostic challenges.

Case Summary: A 2-year-old girl presented for evaluation of a painless lesion surrounding a primary tooth, first noticed before she was 5 months of age. Her medical history was unremarkable without any history of trauma. The clinical examination revealed a yellowish cylindrical mass, hard in consistency, completely surrounding the cervical area of the left first primary lower incisor. It was nonremovable, strongly adhered to the root surface. With a provisional clinical diagnosis of a tooth abnormality (eg, hypercementosis), a periapical radiograph revealed a