Dentistry and Practices

ISSN: 2581-4990

CASE REPORT

Orofacial Granulomatosa: A Case for Interprofessional Collaboration

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Introduction

Within the goals of interprofessional collaboration, the value of determining a correct diagnosis among participating healthcare providers is consistent with improving patient experiences and reducing healthcare costs. An atypical presentation of signs and symptoms of a disease entity can create a diagnostic challenge for healthcare providers that often require patients to navigate the healthcare system resulting in treatment delays. [1-4] the patient problem presented in this current case demonstrates the need for interprofessional collaboration among providers when facing a diagnostic challenge.

Case Description

A 59 year old female patient [Figure 1] presented to a primary care clinic with the chief complaint of upper lip swelling, which had been present for three days. A diagnosis of Herpes Labalis was made and Valtrex 500 mg prescribed. Four days later, the patient returned to the primary care clinic when the swelling spread to the left side of her face. No additional testing, diagnoses, or treatment was rendered at that time. Following an additional four days, the patient returned to the primary care clinic when the swelling had spread to the right side of her face. A bacterial infection was suspected and Clindamycin 300 mg prescribed. A blood specimen was drawn with lab order for a CBC with Differential/Platelet; Complete metabolic panel; TSH; Antinuclear Antibodies Direct; C-Reative Protein, Quant. [Table 1].

PATIENT
Female Caucasian DOB: 07/30/1957 (59)
CHIEF COMPLAINT/CONCERN
"Upper lip and Facial Swelling"
BACKROUND AND/OR PATIENT HISTORY
Penicillin allergy Current medications: Estradiol 2 mg Lorazepam 1 mg Paroxetine 37.5 mg Special Education High School Teacher
CURRENT FINDINGS
Diagnosis: Swelling, mass, or lump on face Temp: 37^{0}C BP: 135/80

Figure 1: Abbreviated patient history and diagnostic information.

On the following day, she then presented to the Emergency Department of a local hospital with the complaint of facial swelling. The swelling, which began in her upper lip two weeks earlier, had now spread to the left and right cheeks, just below her eyes, encompassing most of her mid-face [Figure 2]. Vital signs were within normal limits, with a slight increase in systolic and diastolic blood pressure readings. The patient had not experienced pain. The patient had no remarkable history of trauma or other environmental occurrence. The emergency room (ER) physician did not make a differential diagnosis of the facial swelling. The ER medical staff prescribed the following diagnostic tests, medication, and follow-up care:

Additional Procedure and tests performed during the Emergency Department visit:

C3 and C4

Positive Lab Results	Result	Reference Interval
WBC	11.8 x 10^3/µl	3.4-10.8 x 10^3/µl
Platelets	389 x 10^3/µl	150-379 x 10^3/µl
Neutrophils	9.2 x 10^3/µl	1.4-7.0 x 10^3/µl
Glucose, Serum	102 mg/dl	65-99 mg/dl

Table 1: Laboratory results from Day 4/Primary Care Provider visit. Positive results above the Reference Intervals, particularly WBC and Neutrophil, did not inform provider diagnosis.

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Received: June 25, 2018; **Accepted:** June 27, 2018; **Published:** June 29, 2018

Procalcitonin

Erythrocyte Sedimentation Rate

Medication prescribed during Emergency Department Visit:

Prednisone 10 mg for 3 weeks

Follow-up from emergency Department:

Upper lip biopsy

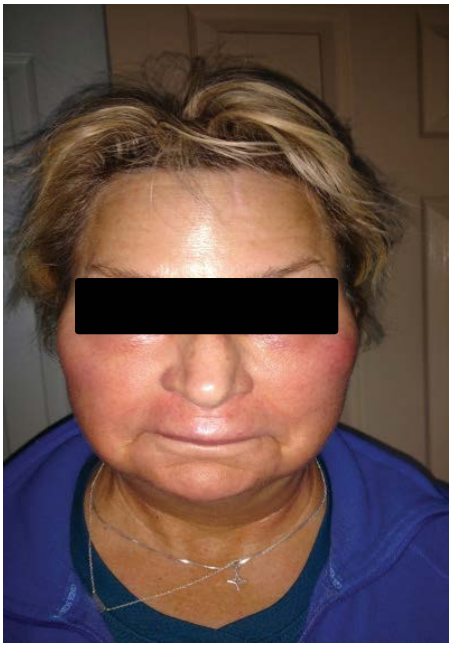

Figure 2: The patient presenting soft tissues swelling in the upper lip and bilaterally in the malar and infraorbital regions.

Allergist referral

Rheumatology referral

ER Diagnosis: Facial Swelling

Two days following the ER visit, the patient presented to the office of a local Allergy Group and was examined by a Physician's Assistant (PA) and allergist (MD). Her history, signs & symptoms were used to rule out any kind of typical allergic reaction. There was no differential diagnosis made at this appointment. Two days following, on the advice of her Esthetician, the patient consulted with her general dentist. The dentist ruled out any odontogenic issues (tooth/supporting structures). Following the dental visit [Table 2], the patient presented to a dermatologist (MD & PA) for assessment of her problem. After the history and examination, the dermatologist determined a working diagnosis of Chelitis Granulomatosa with idiopathic etiology. An incisional biopsy of the upper lip [Figure 3] was performed. The Dermatologist made a referral to an ENT for further evaluation.

At this point in the scenario [Table 2], two weeks following her first urgent care visit, the patient contacted her former dentist in a Midwestern city with photos of her facial swelling and a limited history of her medical journey. The dentist took a medical history over the phone and questioned the patient about any previous facial cosmetic procedures. The patient revealed having had Botox injections and dermal filler placement approximately 12 months prior from a plastic surgeon, which included injections of Juvéderm Voluma XC in the R and L nasolabial folds. The patient had not revealed the cosmetic procedures during history taking with each of the healthcare providers seen in the two-week period.

The patient then presented to a local Otolaryngology office and

Day 1,5 Primary Care	Day 9 Primary Care	Day 10 ER	Day 12 Allergist	Day 14 General Dentist	Day 16 Dermatologist	Day 19 General Dentist	Day 21 ENT	Day 22 Imaging	Day 26 Pathology Report	Day 31 Pathology Addendum	Day 68 Treatment completion
Viral/Bacterial infections Valtrex / clindamycin		Prednisone	Per exclusionem	Per exclusionem	Chelitis Granulomatosa	Dermal Fillers	CT Scan	Consistent with soft tissue swelling	Granulomatous inflammation	Diagnosis	Hyaluronidase
									Granulomatous inflammation		

Table 2: Diagnosis and treatment timeline.

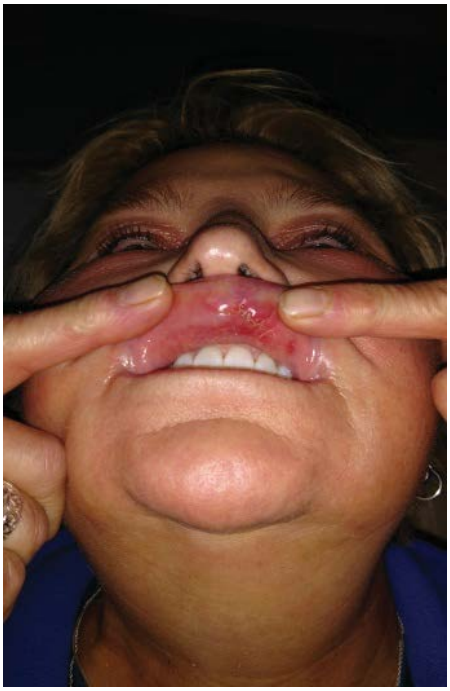

Figure 3: Upper lip incisional biopsy.

consulted with an ENT (MD). A head and neck **Maxillofacial CT** scan with intravenous contrast was ordered and completed with the following findings:

Facial bones: No fracture or focal osseous abnormality identified.

Facial soft tissues: There are soft tissues swelling with abnormal attenuation noted over the malar regions bilaterally involving the anterior buccal space. This extends superiorly to the level below the inferior margin of the orbit. There is abnormal subcutaneous soft tissue over the malar regions bilaterally and some associated skin thickening noted in this region. Findings suggest inflammatory or post- traumatic process such as cellulitis/fasciitis or hematoma, bilaterally. Differential concerns, though remote, would include non-Hodgkin's lymphoma or changes from facial soft tissue augmentation procedures. Masticator spaces are unremarkable bilaterally and no muscle involvement currently identified.

Salivary glands: The parotid glands and submandibular glands have an unremarkable imaging appearance.

Orbit & globes: Soft tissues swelling extends to the inferior margin of the orbit, anteriorly. The post septal orbital soft tissues are otherwise unremarkable. Lacrimal glands are unremarkable. Globes are unremarkable.

Paranasal sinuses and mastoid air cells: Paranasal sinuses are normally aerated.

Visualized intracranial structures: The visualized intracranial structures are within normal limits.

Skull bases and cervical spine: The skull base and partially visualized upper cervical spine are intact and unremarkable.

Surgical Pathology Report*:

Pathologic diagnosis

"Upper lip, incisional biopsy: Dermal acute and chronic granulomatous inflammation with pronounced giant-cell reaction; no evidence of malignancy."

*Upon receipt of this surgical pathology Report, the ENT requested another review of the biopsy slides by a second pathologist.

Pathology Report Addendum [Table 2]:

Interpretation/Result

"The slides show an incisional biopsy of oral mucosa with well-formed granulomata palisading around pale blue to gray areas of necrobiosis, with a few eosinophils in the necrobiotic foci. Small lymphocytes and histiocytes surround the granulomata. No Birefringent foreign material is identified on polarized light microscopic examination. No obvious needle-shaped clefts are identified in the necrobiotic areas. Dr. _____ was contacted by telephone, and there is no known history of injected collagen or other substance [previous history of Juvéderm Voluma injection was not disclosed, yet]. The well-formed granulomata and necrobiosis would be unusual for granulomatous chelitis, although this remains a possibility, as does a reaction to non-birefringent foreign material or endogenous material, and other granulomatous processes. It is unclear how to best classify this granulomatous inflammatory pattern. This case will be sent for review to UCSF Dermatopathology and Oral Pathology Service, and the result will be issued as an addendum to this report.

A literature search on adverse effects of dermal fillers by the patient's former dentist in Chicago, led to a working diagnosis of a foreign body reaction to hyaluronic acid dermal fillers. Following contact and case discussion between the patient's plastic surgeon and the medical consultant for the Allergan Corp, an adverse reaction to Juvéderm Voluma was identified as a probable etiology for the facial Granulomatosa. A series of hyaluronidase injections were prescribed as a treatment and administered to the patient by her plastic surgeon. Six weeks following the initiation of hyaluronidase injections, the patient's facial swelling subsided and is no longer visible [Figure 4].

Discussion

This patient scenario presents a number of confounding obstacles for obtaining a proper diagnosis and potential treatment. The first obstacle was the patient presenting a diagnostic dilemma for facial edema with unknown etiology. Within the diagnostic thinking process of various pathologic causes for facial edema, multiple healthcare providers initially considered a number of entities in the development of a differential diagnosis [Figure 5]. The patients past history, current health status, localization of the edema, absence of any signs or symptoms of infectious agents, autoimmune disease, or neoplasm led to the following differential diagnosis:

- Orofacial Granulomatosis

- Allergy
- Foreign Body Reaction

Allergy as an etiology was ruled out following examination of the patient by an allergist. The lack of a proper history regarding dermal filler injections made the consideration of a foreign body reaction to be unlikely. Orofacial granulomatosis is an uncommon disease encompassing a variety of clinical presentations, when biopsied; reveal the presence of nonspecific granulomatous inflammation. [5] Orofacial swelling, with or without intraoral manifestations is a common presentation at onset. [6] The disorder is idiopathic but appears to represent an abnormal immune reaction to a variety of inciting agents. Orofacial granulomatosis is highly variable and can occur at any age; however, the majority of patients are adults. The most frequent site of involvement is the lips. The labial tissues demonstrate a nontender, persistent swelling that may involve one or both lips. When involving the lips alone, it is called Chelitis Granulomatosa. In addition, similar lesions can be seen in association with a number of systemic diseases, such as Sarcoidosis and Crohn's. [8] The pathologic examination in this case demonstrated well-formed granulomata and necrobiosis which are not the type of granulomas seen classically with chelitis granulomatous. A literature search on Pub Med, using search terms-Foreign Body, Granulomas, Dermal Fillers, Hyaluronic Acid Fillers-found a number of studies reporting genuine granuloma formation following implantation of injectable dermal fillers as a rare complication. Reported incidences ranged from one in 100 patients (1 percent) to one in 5000 (0.02 percent). Foreign body granulomas have been observed several months to years after injection at implantation sites [9-11].

Figure 4: Resolution of facial swelling following hyaluronidase injections.

In the era of evidenced-based practice and technological advances, taking a proper medical history often has been underrated in its efficacy in the diagnostic process. This idea continues to be contrasted with the traditional method of thorough history taking, physical examination, and clinical reasoning about what tests, if any, are needed to reach a proper diagnosis. The later tradition in medicine may take somewhat longer, but remains a cornerstone of clinical practice. [2, 3] In this current scenario, knowledge of the dermal fillers and Botox injections was not ascertained in history-taking from each of the healthcare providers that examined the patient. The patient's history of treatment by a plastic surgeon, one year prior to the onset of facial edema, remained unknown. A further obstacle to obtaining a proper diagnosis in this case could be the lack of a common medical record. If each Medical Record contains sufficient, accurate information that could be used to support a diagnosis and justify treatment, patient outcomes would promote continuity of care among health care providers. [4] In this particular case, the previous treatment

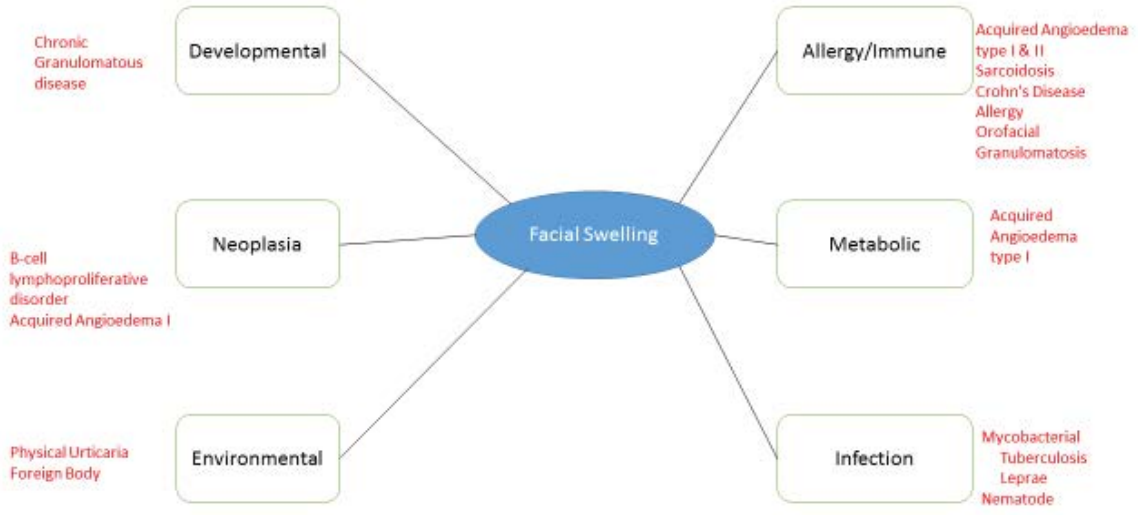

Figure 5: DAMIEN Diagnostic Thinking Map.

record from the patient's plastic surgeon would have been available and of consideration in the diagnostic process and subsequent resolution of the facial granulomatosis.

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Citation: Brown BP (2018) Orofacial Granulomatosa: A Case for Interprofessional Collaboration. Dent Pract 1: 001-005.