

Pediatric unilateral facial swelling



Awatef Draz, BDS, PhD,^a Wael Elias, BDS, MS,^b Azza El-Sissi, BDS, PhD,^a and Min-Ling Liu, MD^c
King AbdulAziz University, Saudi Arabia and George Washington University, Washington, DC, USA.
(Oral Surg Oral Med Oral Pathol Oral Radiol 2017;123:519-523)

CLINICAL PRESENTATION

A 10-year-old, previously healthy, Afghani female presented with a 3-week history of rapidly progressive, painless left facial swelling to the Oral Surgery Department at our institution. She denied any fevers, constitutional symptoms, or recent trauma or travel. Her immunizations were up-to-date, and she had no history of tuberculosis exposure. She reported severe pain, which started 2 days before her presentation to us. Extraoral clinical examination revealed a left-sided facial swelling extending from the infraorbital area to the upper lip, with left facial droop and erythematous overlying skin with evidence of overlying telangiectasia (Figure 1A). A large 4 × 4-cm protruding mass was evident. Further intraoral examination revealed a nontender, red, firm mass involving the left maxillary alveolar ridge and the entirety of the left maxilla and crossing over to the contralateral side of the hard palate (Figure 1B). Grade III mobility of teeth #22 through #27 on the left side was evident. A complete physical examination was unremarkable for any lymphadenopathy or hepatosplenomegaly.

DIFFERENTIAL DIAGNOSIS

Facial swelling is a common yet challenging clinical entity in the pediatric population owing to a very broad differential.¹ A clear understanding of the varying clinical presentations, facial anatomy, and pathologic etiologies helps narrow down this differential considerably. Cases of pediatric facial swelling are usually categorized based on presentation into 4 distinct groups: (1) acute swelling with inflammation, (2) nonprogressive, (3) slowly progressive, and (4) rapidly progressive swellings.²

Acute swelling with inflammation was among our top considerations, as it is the most common cause of pediatric facial swelling.¹ However, the inflammation accompanying this acute swelling is usually due to sinusitis, lymphadenitis, or odontogenic infections. Our patient did not have any preceding tooth or sinus infection, lymphadenitis, constitutional symptoms, or fevers. Moreover, she had an insidious onset, rather than the acute onset associated with such infections, steering us away from this category.

Absence of pain at the onset of swelling as well as purulent discharge led us to consider other categories. Next we considered the slowly progressive swelling category of etiologies. A number of conditions are known to cause slowly progressive lesions, including neurofibroma, lymphatic or vascular malformation, and hemangiomas or osseous disease, such as fibrous dysplasia. Fibrous dysplasia produces characteristic deformities with extensive maxillary involvement, resulting in slowly progressive malar surface protrusion and usually causing loss of the nasomaxillary angle and a feline facial appearance. Cherubism, an autosomal dominant inherited condition with characteristic mandibular swelling, usually affects both sides of the face, making this a less likely explanation for our patient.^{3,4}

Capillary hemangioma of infancy was not fitting for a diagnosis in our patient. Capillary hemangiomas are usually bright red in color and, while protuberant, they are compressible. The location can be facial, scalp, or back and anterior chest. Only 20% of patients have them visible at birth, and they are 5 times more common in females than males. Our patient lacked a history of such lesion at birth, but more importantly, she did not have the initial phase of enlargement, known as the proliferative phase, noted in the first 2 to 10 months of life, followed by the spontaneous regression/involution.

Our patient was unlikely to have a vascular malformation, as those are present in 90% of patients at birth, but unlike capillary hemangioma, they are seen equally in males and females. Our patient developed the swelling in the past 3 weeks only. Vascular malformation grows in proportion with the growth of the infant and lacks any regression, as seen in capillary hemangiomas. Venous malformation appears as well-defined avidly enhanced masses with occasional phleboliths seen.⁵ Lymphangiomas were not fitting either,

^aProfessor of Oral and Maxillofacial Pathology, Department of Basic Oral and Diagnostic Sciences, Faculty of Dentistry, King AbdulAziz University, Jeddah, Kingdom of Saudi Arabia.

^bAssistant Professor and Chairman of Oral and Diagnostic Sciences Department, Department of Basic Oral and Diagnostic Sciences, Faculty of Dentistry, King AbdulAziz University, Jeddah, Kingdom of Saudi Arabia.

^cAssociate Professor of Pathology, Pathology and Laboratory Service, Veterans Affairs Medical Center, and George Washington University, Washington, DC, USA.

Received for publication Aug 1, 2015; returned for revision Apr 26, 2016; accepted for publication May 5, 2016.

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2212-4403/\$ - see front matter

<http://dx.doi.org/10.1016/j.oooo.2016.05.007>

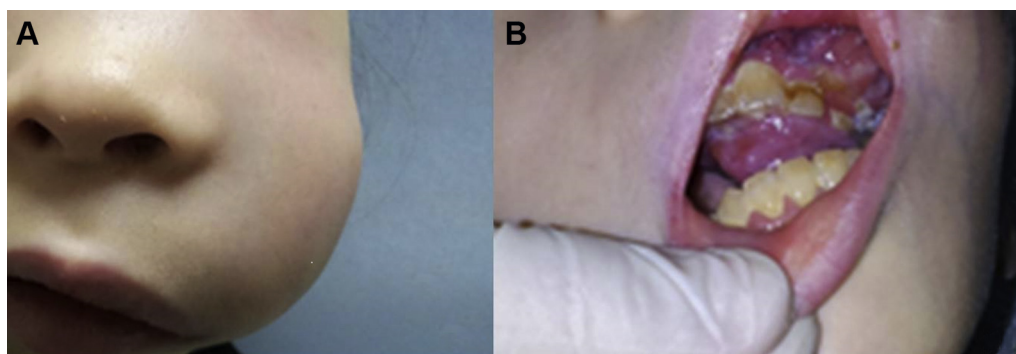


Fig. 1. **A**, Significant left facial swelling extending from the infraorbital region to lip with a facial droop and overlying erythematous skin. **B**, Intraoral view showing an erythematous exophytic swelling obstructing the left maxillary vestibule and the left side of the oral cavity.

given that they present as neck masses at birth.^{6,7} Nevertheless, cavernous lymphangiomas are not uncommon in the oral cavity. Unlike the case presented, however, oral lymphangiomas typically involve the tongue and clinically have a pebbly surface.

Causes of nonprogressive swelling were considered next, namely congenital midfacial masses, including frontoethmoid cephaloceles, nasal gliomas/nasal dermoid, and epidermoid cysts.⁸ All have the same manifestations clinically, as they present with a broad nasal bridge, glabellar swelling, and hypertelorism.⁹ Imaging by computed tomography and magnetic resonance imaging is usually diagnostic, with computed tomography showing bony defects of the craniofacial junction and magnetic resonance images better delineating the relationship of the mass to the brain.¹⁰ While congenital causes for nonprogressive swelling are usually present shortly after birth, occasional cases may not be immediately evident at birth and may take several years to manifest; acute infections of those lesions could lead to rapid growth.

We had reluctantly considered a congenital etiology. Our patient had a unilateral rather than a midline swelling and had a progressive course without inflammatory signs and symptoms, making a congenital etiology a less likely cause, and this case therefore did not fit into the nonprogressive category.

Finally, a rapidly progressive lesion, such as malignant neoplasm, became the most likely diagnosis. While most neoplasms cause a gradual change in facial features, aggressive malignancies tend to cause rapid changes. A hematoma secondary to trauma may also cause a rapidly progressive swelling. However, there was no history of trauma in our patient to justify that diagnosis. A malignant neoplasm extending from the maxillary sinus causing a unilateral facial swelling, such as rhabdomyosarcoma, metastatic neuroblastoma, osteosarcoma, Ewing's sarcoma, Langerhans's histiocytosis, lymphoma, and myeloid sarcoma was

considered. The latter two usually have a rapid growth and can cause constitutional manifestations as fevers, chills, and weight loss, none of which were present in our patient. Bone marrow suppression secondary to malignant leukemia or lymphoma clones and a resultant severe thrombocytopenia may be manifested by petechial skin rash, oral purpura, and an increased bleeding tendency. The latter is an important issue to consider when planning a biopsy of a rapidly progressive facial swelling by the surgeon. Similarly, other malignancies arising from contiguous structures were also considered as a cause of the rapidly progressive unilateral facial asymmetry and swelling that was evident in this patient. Such malignancies include palatal neoplasms like mucoepidermoid carcinoma of salivary glands with possible intraoral findings of fungating mass and tissue destruction with necrosis and ulceration.

In this case, we favored an aggressive neoplastic etiology, given the rapidly progressive presentation in the context of absent history of trauma, prior infection, or inflammatory manifestations.

DIAGNOSIS AND MANAGEMENT

Diagnostic imaging was indicated in our patient, as she lacked acute inflammatory manifestations, making an infectious process less likely. Additionally, the decision to pursue imaging was supported to facilitate surgical planning to rule out malignancy. A cone-beam computed tomography scan was done (Figure 2), revealing a mass originating from the maxillary sinus encroaching on and involving the palatal aspect of the maxilla.

Aggressive tumors in the maxilla, such as non-Hodgkin's lymphomas, frequently cause discrete foci of alveolar damage, loss of lamina dura with destruction of dental crypts, and widening of the periodontal ligament on imaging studies.¹¹⁻¹⁴

In the absence of definitive clinical manifestations, an incisional biopsy of the hard palate mass was performed

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