Lobular capillary hemangioma (LCH), characterized by benign proliferation of capillaries with a lobular architecture, is a common benign vascular tumor of children and adults [1]. The International Society for the Study of Vascular Anomalies includes LCH in the benign vascular tumor group [1]. LCH may occur at all ages; however, LCH is more frequently seen in women and men under 18 years of age [2]. The most common etiologic factors are trauma, hormonal factors, and poor oral hygiene.

The majority of LCH present on the head and neck, commonly involving gingiva, lips, tongue, and buccal mucosa. LCH is rarely located in the nasal cavity [3]. The anterior septal mucosa, tip of the turbinates, and the vestibule are the common nasal cavity sites for LCH [4]. LCH may involve superficial cutaneous, mucosal, and subcutaneous structures. The rare occurrence and deep location of subcutaneous variant makes the clinical diagnosis challenging. We conducted a retrospective chart review to report the clinical, radiologic, and histological features of LCH in a child with a subcutaneous lateral nasal mass.

2. Case Report

A 12-year-old male with past medical history of type II diabetes mellitus presented with a gradually enlarging mass on the left lateral nasal wall for 8 months (Figure 1). The patient hit his nose on his brother’s head a year ago. The child had no nasal obstruction, no epistaxis, no recurrent sinus infections, no anosmia nor hyposmia, and no snoring. There was no family history of childhood cancers. On physical examination, the 1.5×1.5 cm mass was a firm, nontender, immobile, subcutaneous nodule, with no color change of the overlying skin. CT imaging documented a hyperdense and nonlipomatous mass involving soft tissue of the left lateral nasal wall. An excisional biopsy was performed. Histologic evaluation showed subcutaneous lobular capillary hemangioma. Subcutaneous lobular capillary hemangioma, although uncommon, should be considered in the differential diagnosis of lateral nasal wall mass in children.
3. Discussion

In the present study, subcutaneous LCH was documented in a child with a history of trauma to the nose. While no direct causal relationship has been identified, there is an association with estrogen exposure and trauma. Five percent of pregnant patients experience a new onset LCH, especially of the gingiva, hence the term “pregnancy tumor” [5]. Exogenous hormone therapy has also been implicated as a risk factor. Additionally, receptors to the beta isoform of estrogen have been identified on various vascular tumors, including hemangiomas, possibly supporting a hormonal etiology [6]. Prior local trauma is also frequently reported in the patient history, as was in this case; however, it is not considered necessary for development [5].

Histologically, LCH consists of a benign proliferation of capillaries arranged in lobules separated by fibrous septa [7]. Superficial lesions may ulcerate and be associated with inflammation and edema, while the deeper lesions often lack inflammation. The historical terminology, pyogenic granuloma, is a misnomer as the mass is neither infectious or pus producing nor granulomatous. Therefore, the current terminology of LCH is recommended. While usually not needed, immunohistochemistry for endothelial markers CD31 and Factor VIII may be helpful.

The variants of LCH include oral mucosal, satellite, intravenous, dermal, and subcutaneous. Subcutaneous, as described in this case, is the rarest form of LCH and lacks the distinct friable, raised, easily bleeding appearance that is characteristic of the more common counterpart. A review of 106 LCH cases at a single institution found an incidence of 3.4% for the subcutaneous variant [5]. The clinical differential diagnosis of subcutaneous LCH includes a wide variety of subcutaneous pathologies, including but not limited to vascular malformation, Kaposiform hemangioendothelioma, and infantile hemangioma. The present case must also take into account the differential diagnosis for a child with a nasal sidewall mass. The clinical differential diagnosis of lateral nasal wall mass includes nasal dermoid, encephalocele, neoplasm, vascular malformation, and other various reported entities such as foreign body inclusion cyst and heterotopic glial tissue [8, 9]. Surgical treatment with complete excision is the mainstay of treatment and also
provides definitive diagnosis. Recurrence, occurring in 16%, is due to incomplete excision of the mass [4, 10]. No malignant change has been reported [4].

In conclusion, subcutaneous mass can manifest as a lateral nasal wall mass in children, and excision of the mass forms the mainstay of treatment. Prompt diagnosis and treatment of subcutaneous LCH is essential to prevent complication or sequela.

Disclosure

An earlier version of this manuscript was presented in part at the Annual Meeting of American Society of Pediatric Otolaryngology, April 20–22, 2018, in National Harbor, Maryland, USA.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

References


