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CASE REPORT

Cellular Neurothekeoma of the Upper Lip in an Infant

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Key Words
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Cellular neurothekeoma is an uncommon benign skin neoplasm and also a variant of neurothekeoma. Cellular neurothekeomas usually occur in the skin of the upper trunk, head, or neck of children and young adults; however, they rarely occur in infants or involve the lip. A 6-month-old male infant was incidentally found to have a tumor in the upper lip. The tumor was elastic, nontender, and movable, and the overlying mucosa and skin were normal without discoloration. The tumor was excised from the mucosal side of the upper lip, and a pathological examination revealed cellular neurothekeoma. Cellular neurothekeoma in the lip of an infant without overlying skin discoloration might delay the diagnosis and lead to wrong preoperative diagnosis. No similar case has been reported in the literature.
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1. Introduction

Lip tumors in children are uncommon, and most of them are benign.[1,2] The pathology of lip tumors is variable, with hemangioma as the most common one.[1,2] Cellular neurothekeoma is an uncommon benign skin neoplasm and a variant of neurothekeoma.[3] Cellular neurothekeomas usually occur in the skin of the upper trunk, head, or neck of children and young adults; however, they rarely occur in infants or involve the lip.[3—5] We describe a case of cellular neurothekeoma in the upper lip of a 6-month-old male infant. No similar case exists in the literature. The clinical and pathological characteristics of this patient are discussed, and the literature is reviewed.

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2. Case Report

A 6-month-old previously healthy male infant was found incidentally to have a nodular lesion in the upper lip by his mother. On examination, the patient was well except that a nodule was noted in his upper lip. This nodule was elastic, nontender, movable, and measured 1.0 × 0.7 × 0.5 cm. The overlying mucosa and skin were normal without discoloration. No neck lymph node enlargement was found. No other abnormal physical or laboratory findings were present. The patient had no history of trauma, and the family history of neoplasm was negative. Ultrasonogram revealed a well-defined, hypoechoic, heterogeneous mass. The preoperative diagnosis was an epidermoid cyst. An operation was performed under general anesthesia. We approached the tumor from the mucosal side of the upper lip (Figure 1). The tumor was well-defined, elastic, unencapsulated, and extended to the submucosal muscular layer. The tumor was totally excised. The patient stood the operation well, and the postoperative course was uneventful. Histological examination (Figure 2) revealed that the tumor was composed of hypercellular epithelioid cells with diffuse and fascicular growth patterns. Large, hyperchromatic tumor cells and focal mitoses were seen occasionally. Myxomatous change and some trapped nerve bundles were identified. No atypia was found. Immunohistochemically, the tumor was positive for neuron-specific enolase and smooth muscle actin, but negative for S-100 protein, desmin, and glial fibrillary acidic protein. Mild immunoreactivity of CD34 was noted. The immunoreactivity of NKI/C3 was unclear because no such examination was done. Histologically, the tumor was a cellular neurothekeoma. After a 6-month follow-up, the patient was well, and no tumor recurrence was noted.

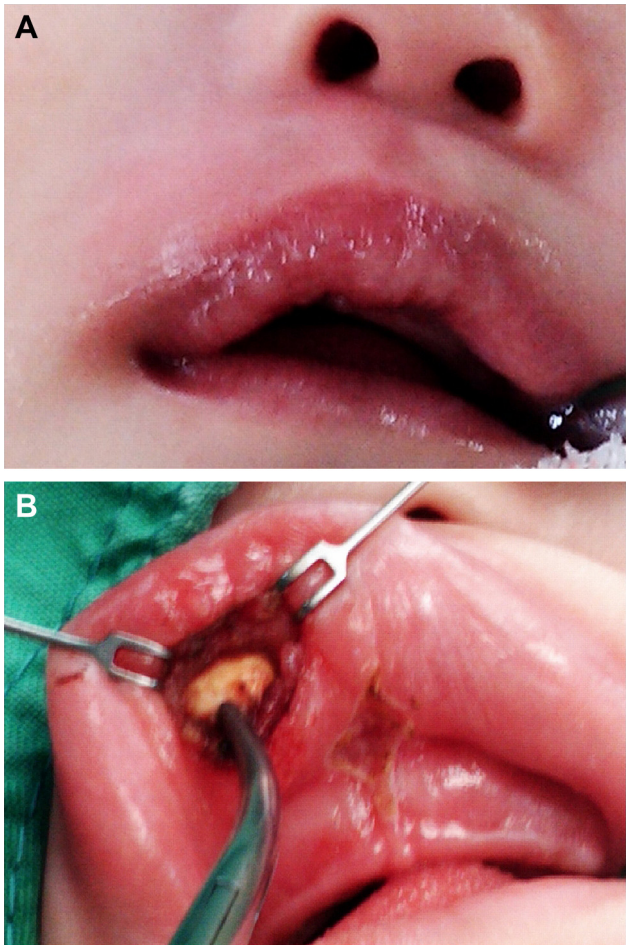

Figure 1 Photographs of the patient. (A) Absence of discoloration of the upper lip. (B) The tumor was excised from the mucosal side of the upper lip.

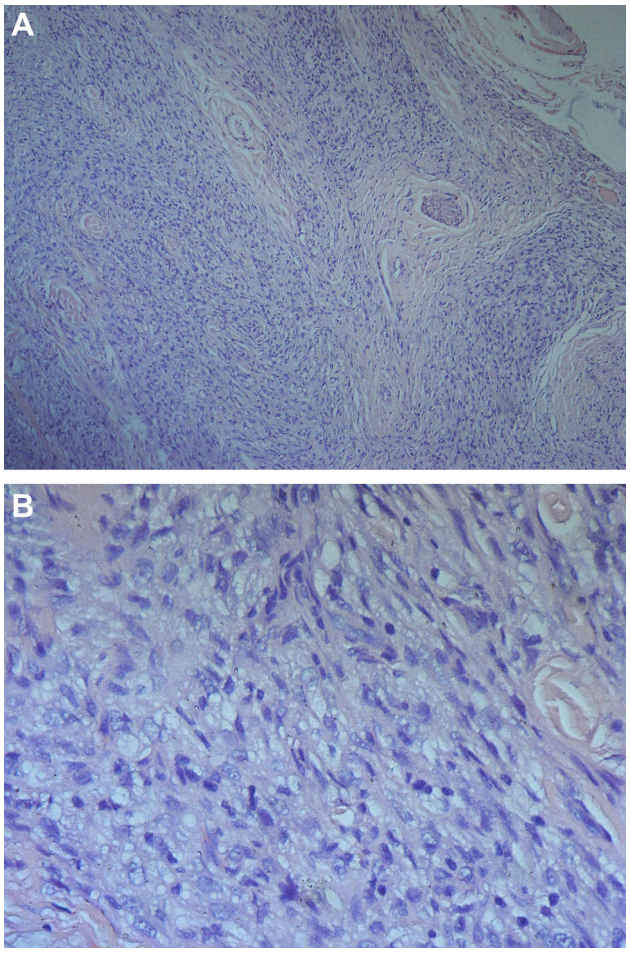

Figure 2 Histopathology of the tumor. The tumor was composed of hypercellular epithelioid cells with diffuse and fascicular growth patterns. [Hematoxylin and eosin staining: (A) 40×; (B) 200×.]

3. Discussion

Soft tissue tumors in children differ from those in adults in frequency, anatomical site, histological types, and prognosis.[6] Tumor occurring in the lip of an infant is extremely rare and difficult to diagnose, especially for a lesion without changes in the overlying skin, as in this case. The pathology of lip tumors includes hemangioma, lymphangioma, hemangiolymphangioma, papilloma, neurofibroma, retention cyst of small salivary glands, venous malformation, eosinophilic granuloma, common

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