Case Report

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Lipoblastoma of the Tongue: A Rare Case Report

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Abstract

Lipoblastoma, a rare benign tumor originating from white fetal adipose tissue, occurs primarily in infancy and early childhood. Lipoblastoma commonly occurs in the trunk and upper and lower extremities and is less common in the head and neck, mediastinum, and retroperitoneum. No case of lipoblastoma of the tongue has been reported to date. Here, we report a case of lipoblastoma presenting as an exophytic mass in the tongue of a 3-year-old boy. To the best of our knowledge, this is the first case of lipoblastoma of the tongue reported in the literature.

Keywords: Lipoblastoma; Tongue; Benign tumor

Introduction

Lipoblastoma is a neoplasm of embryonic adipose tissue that commonly occurs in children younger than 3 years old. Histologically, this exclusively benign tumor, which has a favorable prognosis, consists of fat lobules of variable maturity, multivacuolated lipoblasts, fibrocapillary networks and myxoid stroma. Despite its rarity, lipoblastoma accounts for more than 15% of benign tissue neoplasms in children and most commonly occurs in the trunk (64%), followed by the extremities [1]; it is less common in the head and neck, mediastinum, and retroperitoneum. Even rarer locations include cardiac sites [2,3] and the labia [4]. Lipoblastoma of the tongue has not yet been reported in the literature. Here, we report this unusual presentation of lipoblastoma in the tongue of a 3-year-old boy. To our knowledge, this is the first case of lipoblastoma of the tongue reported to date.

Case Report

We report the case of a 3-year-old boy presenting with an exophytic, recurrent mass in the tongue one year after surgical resection. When the boy was 1 year old, his family members noticed a nodule 0.5 × 0.2 cm in size on the left side of his tongue, with no swelling or numbness. The nodule increased gradually to 3.0 × 3.0 cm after one year, and a total left-tongue nodular resection was performed at another hospital. The initial resection specimen showed tongue mucosa erosion and granulation tissue composed of a large number of newborn capillaries, lymphocytes, plasma cells and neutrophils **(Figure 1A).** In another area, adipose lobules with focal myxoid stroma segmented by coarse fiber were observed **(Figures 1B and 1C).** Mature adipose cells, spindle-shaped cells and stellate cells with a mucous background, coarse collagen fibers, and vacuolated lipoblasts were observed at high magnification **(Figure 1D),** and the lipoblasts were positive for S-100 **(Figure 1E)** and p16 **(Figure 1F).** These findings confirmed a diagnosis of lipoblastoma.

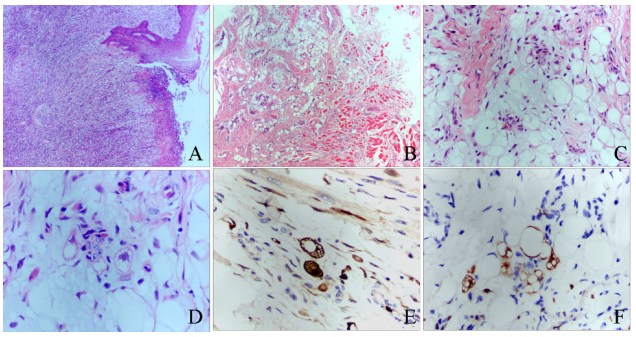

Figure 1 Histological findings for the initial resection specimen from the tongue, (A-F) H&E, (A) Low-power view showing the tongue mucosa erosion and granulation tissue (×40); (B) Low-power view showing adipose lobules segmented by coarse fiber and adipose lobules with focal myxoid stroma (×40);(C) Medium-power microscopy showing lobules of spindle-shaped cells and stellate cells as well as mature and immature adipocytes, with fibrocapillary networks and myxoid stroma (×200); (D) High-power microscopy showing vacuolated lipoblasts (×400); (E-F) Immunohistochemical staining (×200); (E) Staining with an antibody against S-100 showed cytoplasmic and nuclear positivity in tumor cells; (F) Staining for p16 revealed cytoplasmic and nuclear positivity in tumor cells.

The patient recovered well after the operation. One year later, the family found a gradually growing mass in the tongue at the original location, suggesting recurrence. The boy was then referred for assessment. The physical examination was normal except for a 3.0 × 3.5 cm, exophytic, well-circumscribed soft mass in the left tongue **(Figure 2).**

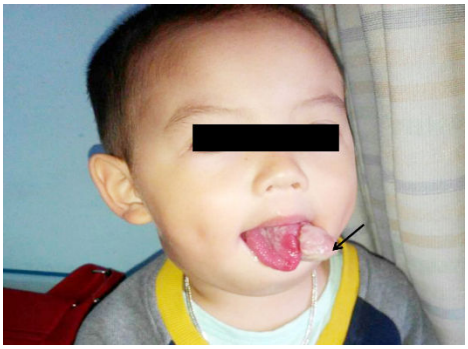

Figure 2 An exophytic mass in the left tongue, 3.0 × 3.5 cm in size, found on examination (arrow).

A combination of sharp dissection and electrocautery was used to excise the mass. Serial sections revealed a yellowish-tan to reddish-tan; well-circumscribed mass measuring 3.0 × 3.5 cm. Microscopically, the pathological morphology was similar to that of the initial resection specimen. Mucosal erosion and granulation tissue were observed. In another area, we observed a multilobulated adipocytic lesion with fibrocapillary networks and myxoid stroma located in the skeletal muscle of the tongue **(Figure 3A).** The capsule was not obvious, and the lobules were composed of mature adipose cells, spindle-shaped cells and stellate cells with myxoid change **(Figure 3B)** and vacuolated lipoblasts **(Figure 3C);** varying degrees of differentiation were observed, with no atypical or malignant features. Within the tumor cells, staining for the immunohistochemical markers p53 and MDM2 was negative, whereas staining for S-100 and p16 was positive, and the proliferation index Ki-67 was 2%. These findings confirmed recurrence of the lipoblastoma. The patient recovered well after the second resection.

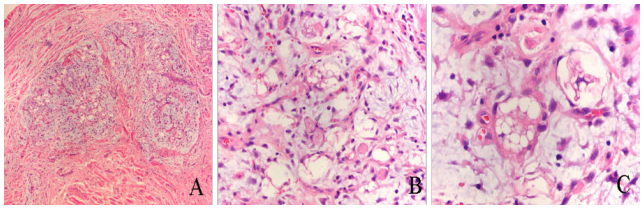

Figure 3 Histological findings for recurrent lipoblastoma in the tongue, (A-C) H&E; (A) Low-power view showing a multilobulated adipocytic lesion with fibrocapillary networks and myxoid stroma located in the skeletal muscle of the tongue in another area (×40); (B) Medium-power microscopy showing lobules of spindle-shaped cells and stellate cells as well as mature and immature adipocytes, with fibrocapillary networks and myxoid stroma (×200); (C) High-power microscopy showing vacuolated lipoblasts (×400).

Discussion

Lipoblastoma, a rare benign tumor arising from embryonic white fat, occurs nearly exclusively in infants and young children and is characterized by a 3:1 male predominance. The most common locations are the trunk and extremities, whereas other locations are rare. The occurrence of lipoblastoma of the tongue has not been reported to date. Here, we present a recurrent case of lipoblastoma of the tongue. In this case, the initial resection and recurrent resection specimens exhibited certain histopathologically similar features, such as tongue mucosa erosion, granulation tissue, lobules separated by fibrous septa and composed predominantly of spindle-shaped cells, and stellate cells and vacuolated lipoblasts with myxoid areas and no atypical or malignant features. The tumor cells were positive for p16; p16 immunostaining has been reported as positive in a few cases of lipoblastoma [5].

The differential diagnosis of lipoblastoma must rule out myxolipoma, myxoid liposarcoma, hibernoma, infantile fibromatosis and lipofibromatosis. Histopathologically, myxoid liposarcoma is extremely similar to lipoblastoma. Both lesions may contain lipoblasts and spindle cells deposited in a myxoid stroma with a prominent plexiform capillary network. Myxoid liposarcomas tend to be less well demarcated; have a less well-developed lobular growth pattern; often contain areas of microcyst formation; may have areas of focal hypercellularity, nuclear atypia or hyperchromasia; and typically show FUS gene rearrangement in FISH analysis. Lipoblastomas lack nuclear atypia and feature PLAG1 rearrangement. The typical age at which the two lesions occur also differs. Lipoblastoma is a tumor of infancy and early childhood, whereas myxoid liposarcoma occurs in adults. Myxolipomas are benign lipomatous tumors that can contain mucoid substances; some of these lesions can possess an abundance of thin- and thick-walled blood vessels, as was observed in the case described here, and they can be differentiated from lipoblastoma by the absence of lipoblasts. Hibernoma, which features microvesicular brown fat and lacks immature lipoblasts or mesenchyme, is rare in children. Infantile fibromatosis is also a rare type of lesion. Microscopy showed that in the present case, the lesion was ill demarcated; was composed of bland spindle cells with scanty pale, amphophilic cytoplasm; had collagenous stroma; was focally densely hypocellular; and had collagenous areas and a few mostly thin-walled vessels but lacked mature adipose cells and immature lipoblasts. Lipofibromatosis is a rare pediatric soft-tissue neoplasm that shows an admixture of adipose tissue and fibroblastic elements and that mainly involves the hands and feet. Microscopy reveals abundant adipose tissue traversed by bundles of spindled fibroblast-like cells. A significant part of the tumor is composed of fat, but it lacks immature lipoblasts.

Conclusion

Although lipoblastomas have the potential for local invasion and rapid growth, they are associated with a favorable prognosis. There is no risk of metastasis or malignant

transformation. Surgical excision is the treatment of choice, and adjuvant therapy is usually not necessary. However, a follow-up period of 2–5 years is recommended, as the recurrence rate is 14–25%. Indeed, our case exhibited recurrence one year later.

References

1. Coffin CM, Lowichik A, Putnam A (2009) Lipoblastoma (LPB): A clinicopathologic and immunohistochemical analysis of 59 cases. Am J Surg Pathol 33: 1705-1712.
2. Sharma P, Shakya U, Sayami G, Rajbanshi BG (2016) Lipoblastoma: An unusual tumour of the left ventricle. Eur J Cardiothorac Surg 49: e147-148.
3. Dishop MK, O'Connor WN, Abraham S (2001) Primary cardiac lipoblastoma. Pediatr Dev Pathol 4: 276-280.
4. Childress KJ, Hicks J, Wu H, Cottrill CM (2016) Lipoblastoma of the Labia: A case report. J Pediatr Adolesc Gynecol 29: e97-e99.
5. He M, Aisner S, Benevenia J, Patterson F, Aviv H, et al. (2009) p16 immunohistochemistry as an alternative marker to distinguish atypical lipomatous tumor from deep-seated lipoma. Appl Immunohistochem Mol Morphol 17: 51-56.