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CASE REPORT

Pleomorphic lipoma of the face: case report

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Pleomorphic lipoma is an uncommon variant of lipoma, which microscopically may resemble a liposarcoma. However, it is a slow-growing and well-circumscribed lesion with a benign behavior. Therefore, recognizing this entity is extremely important to perform the proper treatment. We present an additional case of pleomorphic lipoma in the face, which to our knowledge seems to be the 11th case reported in the English-language literature.
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Keywords: pleomorphic lipoma; immunohistochemistry; fine needle aspiration biopsy; differential diagnosis

Introduction

Lipomas are relatively common soft tissue mesenchymal neoplasms affecting more frequently trunk and proximal portions of the extremities (Neville *et al*, 2002). Occurrence in head and neck region correspond to about 15–20% of all lipomas (Gnepp, 2001).

There are several histological subtypes such as: fibrolipoma, spindle cell lipoma, infiltrating lipoma, angiolipoma, myxoid lipoma, atypical lipoma, and pleomorphic lipoma (Enzinger and Weiss, 2001; Fregnani *et al*, 2003).

Pleomorphic lipoma (PL) was first reported in the late 1970s by Enzinger (1977). Because of the microscopically bizarre features, it is important to correlate the cytological and histological findings with the clinicopathological data, to avoid a misdiagnosis of a malignant counterpart.

Although lipomas in general are frequent in the head and neck region, there are few cases of the pleomorphic variant reported in the face (Table 1). We describe the clinicopathological and immunohistochemistry findings of an additional case of pleomorphic lipoma of the face.

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Case report

A 65-year-old male patient was referred to the Orocentro, Piracicaba Dental School, UNICAMP for evaluation of a facial swelling present for about 30 months. On clinical examination a 3 cm, semi-fixed, smooth surface and asymptomatic mass located in median right facial region was observed (Figure 1). The patient related a 10-year tobacco use (three cigarettes per day), but he had quit 20 years ago. Intraorally, no alterations were noted. A fine needle aspiration biopsy (FNAB) was performed and revealed a paucicellular smear, with peripheral blood cells, multivacuolated stroma with variable size, compatible with mature adipocytes and some multinucleated cells (Figure 2). Based on these features, a diagnosis of lipoma was suggested. Surgical excision of the lesion was performed under local anesthesia and grossly a well-circumscribed, yellow soft tissue fragment measuring $3.2 \times 3.0 \times 4.0$ cm was obtained. Histopathological examination showed predominantly adipose and fibrous connective tissue with large pleomorphic and multinucleated cells, which were characterized by hyperchromatic nuclei arranged in a floret-like pattern that were often overlapping (Figure 3a,b). The diagnosis of pleomorphic lipoma was established and immunohistochemical study was performed demonstrating that the floret cells were positive for CD34 (Figure 4a) and negative for CD68 (Figure 4b). The patient is in follow-up for about 8 months without signs of recurrence (Figure 5).

Discussion

PL is an uncommon variant of lipoma, which was first described by Enzinger (1977). Although its appearance may be similar to a sclerosing or pleomorphic liposarcoma (Enzinger and Weiss, 2001), they usually show a non-aggressive pattern, with average time from the onset to diagnosis of 3 years and 3 months (Digregorio *et al*, 1992). It affects more commonly males (4:1) between 50 and 70 years of age, and is typically found in the posterior neck, shoulder and back region (Digregorio *et al*, 1992; Enzinger and Weiss, 2001). To the best of our knowledge, there are only 10 cases of

Table 1 Pleomorphic lipomas of the face reported in the English-language literature

Author	*No. of cases*	*Age*	*Gender*	*Size (cm)*	*Follow-up (months)*
Azzopardi *et al* (1983)	2	56	Male	3	42
		68	Male	2.5	12
Walker and Jones (1986)	1	79	Female	6	NA
Bryant (1987)	1	60	Male	1.9	NA
Digregorio *et al* (1992)	1	46	Male	1	108
Allen *et al* (1998)	1	65	Male	1.5	NA
French *et al* (2000)	4	20	Female	NA	–[a]
		42	Female	NA	–[a]
		67	Male	1	120
		71	Male	NA	252
Present case	1	65	Male	4	8

NA, not available.
[a]Follow-up less than 3 months.

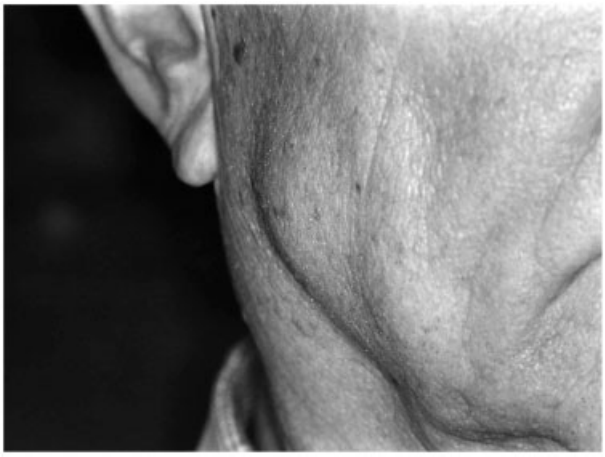

Figure 1 Clinical photograph showing an asymptomatic swelling in the right cheek

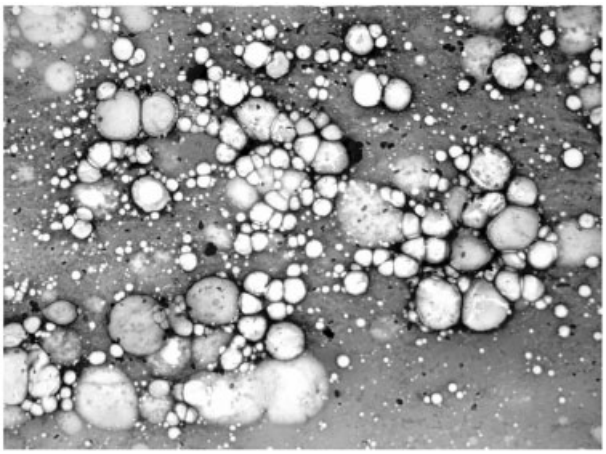

Figure 2 Photomicrograph of FNAB demonstrating mature adipocytes, hyperchromatic oval to spindle pleomorphic cells with scant cytoplasm and some fibroblasts (Diff-Quik, ×100)

pleomorphic lipomas in the face reported in the English-language literature, being the eleventh in the present case. However, the four cases reported by French *et al* (2000) were classified as spindle cell/pleomorphic lipoma with no further specification (Table 1).

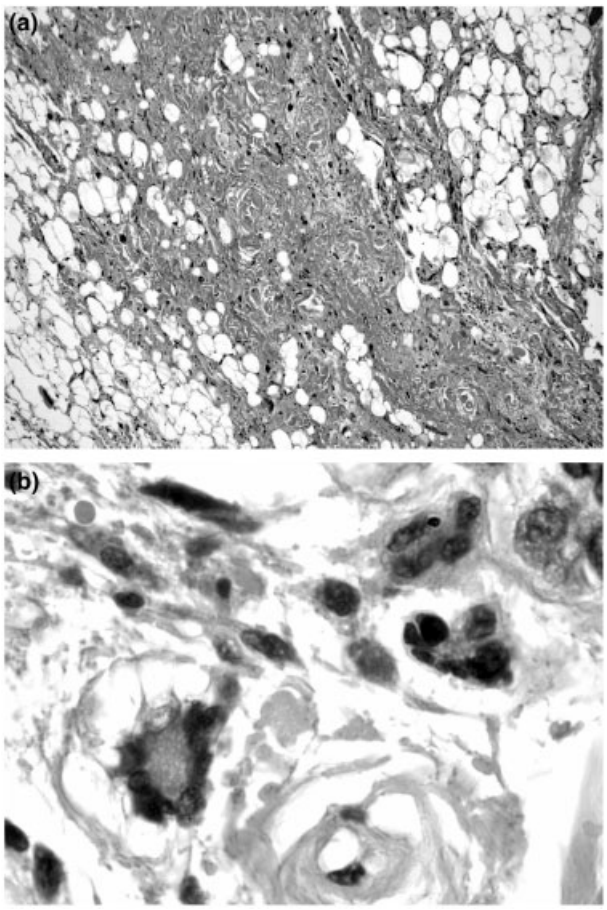

Figure 3 (**a**) Photomicrograph demonstrating adipose and fibrous connective tissue with large pleomorphic and multinucleated cells (H&E, ×50). (**b**) Floret-like multinucleated giant cells with scant cytoplasm and peripherally arranged nuclei (H&E, ×1000)

The use of FNAB for diagnosis of head and neck masses is increasing. Several authors have reported FNAB for diagnosis of pleomorphic lipoma in different anatomical sites, especially in head and neck region (Skoog *et al*, 1999; Yencha and Hodge, 2000). However, the final diagnosis of pleomorphic lipoma must be established based on the clinical, cytological and mainly on histological features, once pleomorphic cells seen in aspirates could lead to a misdiagnosis of a malignant lesion.

Differential diagnosis of PL includes benign and malignant soft tissue neoplasms. Spindle-cell lipoma (SCL) clinically appears as PL, usually affecting head and neck regions in a similar age group, but histologically floret cells are not seen in SCL (Enzinger and Weiss, 2001). Among malignant neoplasms, well-differentiated liposarcoma (WDL) should be considered. The distinction between them is based on particularly clinicopathological features. Comparing the anatomic affected sites, WDL occurs more commonly in extremities and retroperitoneum whereas PL has a superficial involvement (Enzinger and Weiss, 2001). It is important

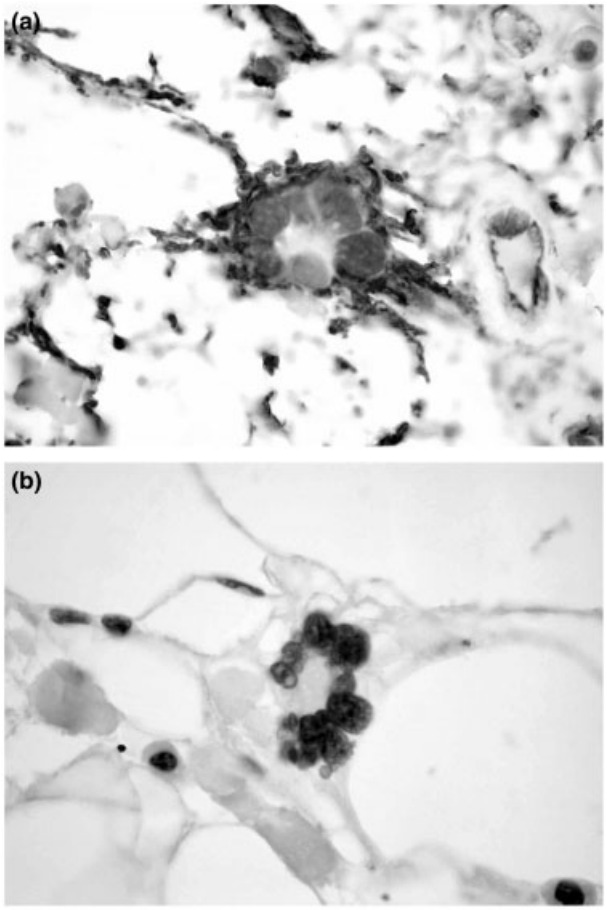

Figure 4 Immunohistochemical staining showing: (**a**) positive for CD34 antigen and (**b**) negative for CD68 antigen

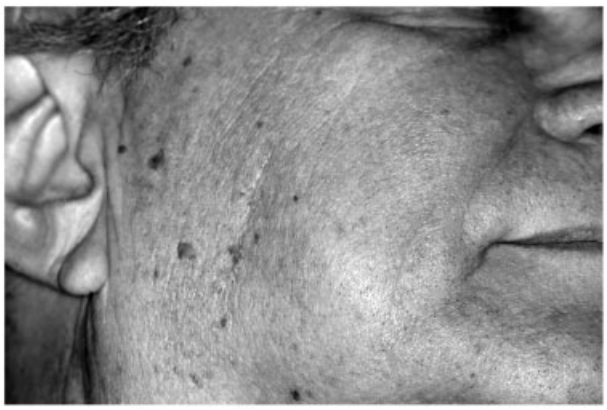

Figure 5 Follow-up photograph of 8 months after surgery

to emphasize that PL of the head and neck is most commonly found in a subcutaneous location, and this information could be crucial in the differential diagnosis (Enzinger and Weiss, 2001). Cytogenetically, PL as well as SCL usually presents chromosome aberrations of 16q and 13q, being these alterations distinct to that seen in atypical lipoma/well-differentiated liposarcoma (Rubin and Fletcher, 1997; Enzinger and Weiss, 2001).

Grossly, PL presents usually as a well-delimitated, lobulated, soft mass, with shape varying from spherical to ovoid, and surface color being yellowish-gray (Shmookler and Enzinger, 1981; Daniel *et al*, 2003). Histologically, the presence of scattered, bizarre giant cells that presents a floret-like pattern is characteristic, but not pathognomonic, once these cells can be present in well-differentiated liposarcomas and pleomorphic liposarcomas (Enzinger and Weiss, 2001). Regarding lipoblasts, it is present in about half of the cases of PL (Shmookler and Enzinger, 1981). However, we did not find lipoblasts in our case. Lipoblasts can also be seen in liposarcomas, but the difference between PL and liposarcomas is the relative proportion of lipoblasts to floret cells, being lipoblasts in a very distinct minority, in contrast with liposarcomas in which lipoblasts outnumber any tumor cells of floret type (Azzopardi *et al*, 1983).

Immunohistochemistry is an auxiliary tool for PL diagnosis. PL, as SCL, is positive for human progenitor cell antigen (CD34). Lipoblasts and normal adipocytes are CD34 negative, while floret cells and atypical spindle cells show strong positivity for CD34 (Suster and Fisher, 1997; Yencha and Hodge, 2000; Daniel *et al*, 2003). It is also known that floret cells are CD68 negative, as in our case (Beham *et al*, 1989). As in SCL, spindle cells and floret cells stain negative for S-100 protein and lipoblasts and lipocytes stain positive (French *et al*, 2000; Enzinger and Weiss, 2001).

Treatment of PL is complete local excision. Despite its malignant histological appearance, PL usually does not show recurrence. The association of clinical data with histological findings has a crucial role for the correct diagnosis of PL, avoiding a possible misdiagnosis of the lesion as being a malignant neoplasm.

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