Acta Histochemica xxx (2015) xxx–xxx

Contents lists available at ScienceDirect

Acta Histochemica

journal homepage: www.elsevier.de/acthis

Head and neck solitary infantile myofibroma: Clinicopathological and immunohistochemical features of a case series

Rodrigo Nascimento Lopes [a], Fábio de Abreu Alves [a], André Caroli Rocha [a], Thales M. Suassuna [b], Luiz Paulo Kowalski [c], Jurema Freire Lisboa de Castro [d], Danyel Elias da Cruz Perez [d,*]

[a] Department of Stomatology, A. C. Camargo Cancer Center, São Paulo, São Paulo, Brazil
[b] Department of Oral and Maxillofacial Surgery, Hospital Geral de Areias, Recife, Pernambuco, Brazil
[c] Department of Head and Neck Surgery and Otorhinolaryngology, A. C. Camargo Cancer Center, São Paulo, São Paulo, Brazil
[d] School of Dentistry, Oral Pathology Section, Federal University of Pernambuco, Recife, Pernambuco, Brazil

ARTICLE INFO

Article history:
Available online xxx

Keywords:
Infantile myofibroma
Differential diagnosis
Head and neck
Immunohistochemistry
Mandible

ABSTRACT

Infantile myofibroma is a rare mesenchymal benign tumor mostly found in the head and neck region. The aim of this study was to describe a small case series of head and neck solitary infantile myofibroma, emphasizing the importance of the histopathological and immunohistochemical features, and the potential diagnostic challenges. For the study, clinical and imaging data were obtained from the medical records. All cases were histologically reviewed, and immunohistochemical analyses were performed to confirm the diagnosis. Four cases of head and neck solitary infantile myofibroma were identified. All patients were females and presented a mean age of 3 years old (ranging from 2 to 6 years). The site of the tumors were the mandible, right cheek, subcutaneous tissue adjacent to basal cortical of the mandible and upper anterior gingiva. No symptoms, such as pain or paresthesia, were reported. Computerized tomography revealed well-delimited tumors. All tumors were positive for vimentin and alpha-smooth muscle actin. All patients underwent surgical excision and no signs of recurrence were observed after long-term follow-up. In summary, head and neck solitary infantile myofibromas are rare and present excellent prognosis. The correlation between clinical, histopathological and immunohistochemical features are essential for a correct diagnosis.

Introduction

Myofibroma is a benign myofibroblastic tumor composed of eosinophilic spindle-shaped cells arranged in fascicles and around blood vessels (Beck et al., 1999; Rubin and Bridge, 2002 Vered et al., 2007). According to the WHO classification of soft tissue tumors (Rubin and Bridge, 2002), the term myofibromatosis should be restricted to multicentric tumors, while myofibroma seems to be more appropriate for solitary lesions. The solitary form is the most frequent and it appears as a locally invasive neoplasia (Beck et al., 1999; Sugatani et al., 1995). The tumor predominantly affects children (Sugatani et al., 1995; Urs et al., 2014), but may develop later in infancy or adults (Vered et al., 2007). Most tumors affect the head and neck region, mainly the skin and subcutaneous tissue.

The diagnosis of myofibromas is based on the histopathological and immunohistochemical features. The myofibroblastic nature is supported by the immunohistochemical expression of smooth muscle markers, such as alpha-smooth muscle actin and specific muscle actin. The local surgical excision is the treatment of choice. The prognosis is related with extension and location of the tumor. Previous studies (Lingen et al., 1995; Oudijk et al., 2012; Urs et al., 2014) have shown a low recurrence rate, however if central nervous system is involved or there are multiple visceral tumors the prognosis is poor (Sugatani et al., 1995). Head and neck solitary infantile myofibromas are rare, with most cases reported as single case reports. The present study reports a small series of solitary infantile myofibroma affecting the head and neck region, including a mandibular case, emphasizing the clinical, radiological, histological and immunohistochemical features.

Abbreviations: BFH, benign fibrohistiocytoma; CT, computerized tomography; IM, infantile myofibroma-myofibromatosis; MM, myofibroma-myofibromatosis; SFT, solitary fibrous tumor; WHO, World Health Organization.

* Corresponding author at: Universidade Federal de Pernambuco, Departamento de Clínica e Odontologia Preventiva, Área de Patologia Oral, 4ª Travessa Professor Artur de Sá s/n, Cidade Universitária, CEP: 50740-521, Brazil.
E-mail address: danyel.perez@ufpe.br (D.E.d.C. Perez).

http://dx.doi.org/10.1016/j.acthis.2015.02.001
0065-1281/

Table 1
Immunohistochemical profile of the tumor cells.

Antibody*	Clone	Dilution	Result
Vimentin	V9	1:400	Positive
Alpha-smooth muscle actin	1A4	1:100	Positive
S-100 protein	Polyclonal	1:2500	Negative
Desmin	D33	1:50	Negative
CD34	QBEnd 10	1:50	Negative
Pan-cytokeratin	AE1/AE3	1:500	Negative
Ki-67	MIB-1	1:100	Positive (<5%)

*All antibodies are from Dako, Glostrup, Denmark.

Material and methods

Between 2000 and 2013, the records of the patients with solitary infantile myofibroma, obtained from the archives of the Department of Stomatology, A.C. Camargo Cancer Center, Sao Paulo, Brazil, and Oral Pathology Laboratory, Federal University of Pernambuco, Recife, Brazil, were selected for the study. Clinical data such as age, gender, location and size of the tumor, type and time of complaints, mobility and tooth displacement were obtained from the medical records. In the same way, imaging findings (cortical thinning, root resorption and tooth displacement), treatment and outcome were also recorded. The study has been approved by the Local Ethics Committee.

All cases were histologically reviewed. In order to confirm the diagnosis, immunohistochemical analyses, using antibodies such as vimentin, alpha-smooth muscle actin, S-100 protein, desmin, CD34 and cytokeratin AE1/AE3 were performed. Additionally, the cell proliferative rate was also evaluated by Ki-67 (Table 1). For immunohistochemical reactions, 3-μm histological sections were obtained from the paraffin-embedded tissue blocks and mounted on slide-coated glass slides. Antigen retrieval was performed in a pressure cooker for 4 min using a 10 mmol $L)^{-1}$ citrate buffer (pH 6.0), following by a washing step with phosphate-buffered saline. Incubations with the primary antibodies were performed for 18 h at 4 °C. After this, the tissue sections were incubated with Post Primary Block for 30 min at 37 °C (NovoLink Max Polymer, Novocastra, Newcastle, UK), followed by application of diaminobenzidine tetrahydrochloride (Dako, Glostrup, Denmark) as the chromogen. Slides were counterstained with Carazzi's hematoxylin. Positive and negative (omission of primary antibodies) were included in all reactions.

Results

Clinical findings

The clinical features are summarized in Table 2. All patients were females with an age ranging from 2 to 6 years. The sites of the tumors were the mandible, right cheek, subcutaneous tissue adjacent to basal cortical of the mandible and upper anterior gingiva. The cases 2 and 3 presented a short time of complaint (4 months), whereas the case 4 showed a longer time of duration (16 months). The case 1 was identified in a routine radiographic examination performed after a facial trauma (fall from her height). During the anamnesis, in case 2, the patient's mother also reported that the child suffered a facial trauma before the lesion development. Tumor size ranged from 2.5 to 5 cm. The cases 3 and 4 appeared as mobile nodule, whereas, in the case 2, the nodule presented reduced mobility. The case 1, an intraosseous tumor, was not mobile for obvious reasons. The clinical appearance varied according to the site of the tumor. In case 1, a slight mandibular swelling was observed (Fig. 1), whereas a submucosal and subcutaneous nodules were observed in cases 2 and 3, respectively. The case 4 appeared as an exophytic nodule in the upper anterior gingiva. No symptoms, such as pain or paresthesia, were reported. In case 4, the lesion caused displacement of the deciduous maxillary central incisors.

Imaging findings

In case 1, the bone window computerized tomography (CT) scan demonstrated a well-defined, osteolytic, unilocular and hypodense lesion in the left body of the mandible, which caused enlargement and disruption of the buccal cortical bone (Fig. 2). The soft tissue window CT showed a solid lesion without invasion of the adjacent soft tissues. In cases 2 and 3, CT scan demonstrated a well-circumscribed, homogenous nodule with soft tissue density, but without bone invasion (Fig. 3). No imaging exams were recorded in case 4. In addition, no signs of multicentric masses were observed.

Treatment and outcome

All lesions were fully excised. Due to patients age, a frozen section biopsy was performed under general anesthesia in cases 1 and 2, which indicated a spindle-cell neoplasm. In case 2, the frozen section examination showed a neoplasm infiltrating the peripheral tissues without cellular atypia. It was not possible to establish the nature, namely benign versus malignant, of the tumor. In the same way, due to patients age, in cases 3 and 4, a surgical excision was performed under general anesthesia and the specimen sent for histopathological analysis. During the surgery, all but one (case 2) were well-circumscribed tumors and were easily excised. The case 2 appeared with irregular margins, raising the suspicion of malignancy.

After treatment, the patients are under periodic follow up. No clinical and imaging signs of recurrence were observed after a mean time of 33 months of treatment (ranging from 12 to 60 months).

Histopathological and immunohistochemical features

Histological examination of all cases showed a basic common theme: a spindle cell tumor showing alternating hypercellular and hypocellular areas with large amount of collagen fibers. The

Table 2
Clinical, treatment and outcome features of cases of head and neck infantile solitary myofibromas.

Case	Age (years)	Site	Size (cm)	Time of complaints (months)	Clinical presentation	Treatment	Follow up time (months)	Outcome
1	02	Mandible	5.0	Routine radiographic examination	Slight mandibular swelling	Surgical excision	48	NED*
2	02	Right cheek	3.0	4.0	Submucosal nodule	Surgical excision	60	NED
3	06	Subcutaneous, adjacent to basal cortical of the mandible	3.0	4.0	Subcutaneous nodule	Surgical excision	12	NED
4	02	Upper anterior gingiva	2.5	16	Exophytic nodule	Surgical excision	12	NED

* NED, no evidence of disease.

Please cite this article in press as: Lopes RN, et al. Head and neck solitary infantile myofibroma: Clinicopathological and immunohistochemical features of a case series. Acta Histochemica (2015), http://dx.doi.org/10.1016/j.acthis.2015.02.001