Oral Science International 9 (2012) 26–28

Contents lists available at SciVerse ScienceDirect

Oral Science International

journal homepage: www.elsevier.com/locate/osi

Case report

A case of pilomatrixoma in the cheek in a 7-year-old girl

Koji Kashima*, Kaori Igawa, Koichi Takamori, Izumi Yoshioka, Sumio Sakoda

Department of Oral and Maxillofacial Surgery, Faculty of Medicine, University of Miyazaki, Miyazaki, Japan

ARTICLE INFO

Article history:
Received 25 October 2010
Received in revised form 8 January 2012
Accepted 17 January 2012

Keywords:
Pilomatrixoma
Calcifying epithelioma
Cheek
Ultrasonography
Magnetic resonance imaging

ABSTRACT

Pilomatrixoma, also known as a calcifying epithelioma, is a benign tumor originating from a hair follicle. We present a case of pilomatrixoma in a 7-year-old girl complaining of swelling in the cheek. Clinical examination disclosed a mass 10 mm in diameter and it was totally removed. The microscopic examination showed encapsulated tumors mainly composed of eosinophilic ghost cells and partly with basophilic portions in hyalinized fibrous stroma with calcification. Preoperative ultrasound and magnetic resonance imaging gave us useful information for complete tumor excision that the tumor existed just under the skin and the risk of facial palsy would be relatively low.

© 2012 Japanese Stomatological Society. Published by Elsevier Ltd. All rights reserved.

1. Introduction

Pilomatrixoma, formerly known as calcifying epithelioma, is a benign tumor arising from protoepithelial cells or hair matrix cells, and it frequently occurs in the subcutaneous tissue of face and neck [1]. This lesion is usually reported by dermatologists and plastic surgeons, and not commonly encountered in dental practice [2]. In this article, we present a case of pilomatrixoma in the cheek in a 7-year-old girl.

2. Case report

A 7-year-old girl visited us complaining of swelling in the left side of the cheek. She presented with a 1-month history of a painless, bluish mass. After her first noticing the tumor, it had been growing gradually in size. Her past history and family history were not contributory. Clinical examination disclosed a mass 10 mm in maximum diameter involving the cheek (Fig. 1). The tumor was elastic, slightly hard, bluish, tender to palpation and not fixed to the underlying tissues. There was no regional lymphadenopathy and general physical examination was unremarkable. Neither loss of sensation nor signs of facial paralysis were observed in the cheek. Although there were no signs of odontogenic infection, decayed and loosened deciduous teeth were found in the maxillary first molars on the both sides. A panoramic radiograph showed that the roots were absorbing, which was about to be replaced by the permanent successional teeth. There was no radiopaque consolidation in the parotid and masseteric region. Ultrasonography demonstrated a well-defined, oval, heterogenous hypoechoic subcutaneous mass with echogenic scattered dots and Doppler flow signal did not increase both in the tumor and in the peripheral region (Fig. 2). Magnetic resonance imaging (MRI) demonstrated a well-defined, 8 mm in size, oval mass in the subcutaneous tissue of the right cheek, with homogenous low T1-weighted signal intensity, low-to-intermediate T2-weighted signal intensity, with a lack of continuity with the parotid gland and the masseter muscle (Fig. 3). The authors tentatively diagnosed the mass as a benign tumor in the left cheek. The lesion was removed with the overlying skin under general anesthesia with primary closure of the wound in the usual manner. Grossly the tumor was elastic slight hard, oval mass measuring 7 mm × 5 mm × 5 mm, mottled with white-yellowish color (Fig. 4). Microscopically, the lesion was encapsulated by fibroconnective tissue in the subcutaneous tissue and mainly composed of eosinophilic ghost cells with central unstained shadow in the site of the lost nucleus, and partly with basophilic portions in hyalinized fibrous stroma. Areas of calcification were also noted, accompanied by mild chronic inflammatory infiltration with multinucleated giant cells, fibroblasts, and angiogenesis (Fig. 5). A final diagnosis of pilomatrixoma in the cheek was made. The postoperative course was uneventful and there was no recurrence at follow-up 2 years later.

3. Discussion

A pilomatrixoma is a nodular, subepidermal benign tumor arising from the hair matrix and it is slightly more common in females

* Corresponding author at: Department of Oral and Maxillofacial Surgery, Faculty of Medicine, University of Miyazaki, 5200 Kihara, Kiyotake-cho, Miyazaki 889-1692, Japan. Tel.: +81 985 85 3786; fax: +81 985 85 7190.
E-mail addresses: kojikash@med.miyazaki-u.ac.jp, qq954eh9k@vega.ocn.ne.jp (K. Kashima).

1348-8643/$ – see front matter © 2012 Japanese Stomatological Society. Published by Elsevier Ltd. All rights reserved.
doi:10.1016/S1348-8643(12)00007-9

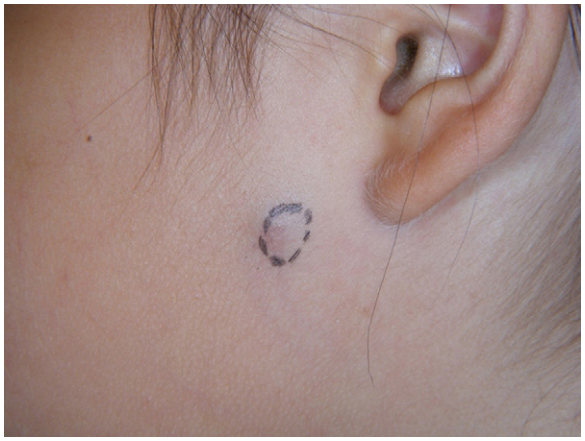

Fig. 1. Initial presentation of the tumor showing a slightly hard, bluish nodule, tender to palpation and not fixed to the underlying tissues. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

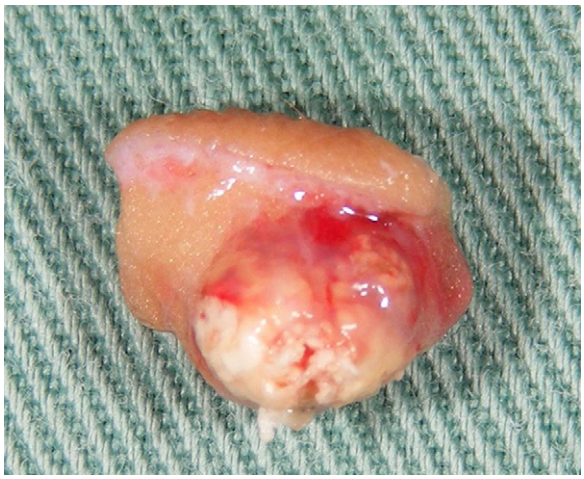

Fig. 4. Macroscopic findings of the excised tumor being elastic slight hard, oval mass measuring 7 mm × 5 mm × 5 mm, mottled with white-yellowish color. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

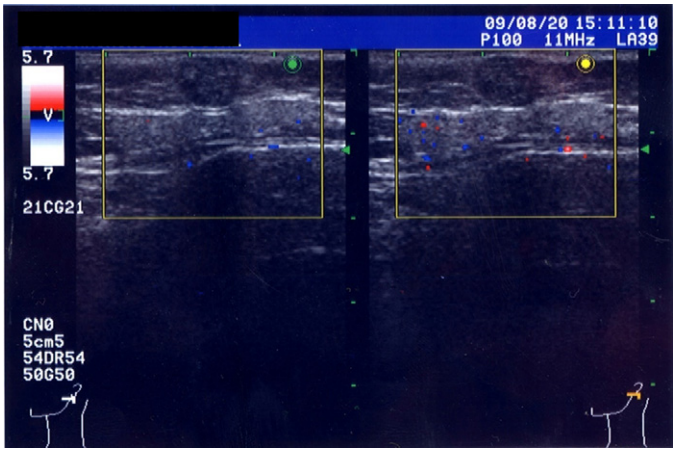

Fig. 2. Ultrasonographic appearance demonstrating a well-defined, oval, heterogeneous hypoechoic subcutaneous mass with echogenic scattered dots, and Doppler flow signal did not increase (arrow).

and usually appears in children, so that 60% of cases are reported in the first two decades of life [1,3]. It occurs predominantly on the head and neck with over 40% of all cases, followed by upper extremities [4]. Our case was a 7-year-old child and it occurred in the cheek.

Despite its frequent occurrence, this lesion is not commonly encountered in dental practice [2] and is misdiagnosed and/or missed in the differential diagnosis [1,4]. Initially, this lesion was called *calcifying epithelioma*, because a sebaceous gland origin was suspected. Later, the term *pilomatrixoma* was given, indicating that the true origin of the tumor is hair matrix cells as well as avoiding a connotation of malignancy [2]. The etiology of this tumor is not completely understood, but there are suggestions that an activating mutation in the β-catenin gene mapped chromosome 3 p22–21.3 plays a major role in the tumor genesis [5].

Although diagnosis of pilomatrixoma can usually be made solely on the basis of clinical features because of its superficial occurrence in the skin, the accuracy is reportedly low, and a correct preoperative diagnosis was made from 28.9% to 49% of the time [6–8]. As for diagnostic imaging, MRI findings of pilomatrixoma are homogenous intermediate T1-weighted signal intensity, heterogenous high T2-weighted signal intensity [9,10]. In computed

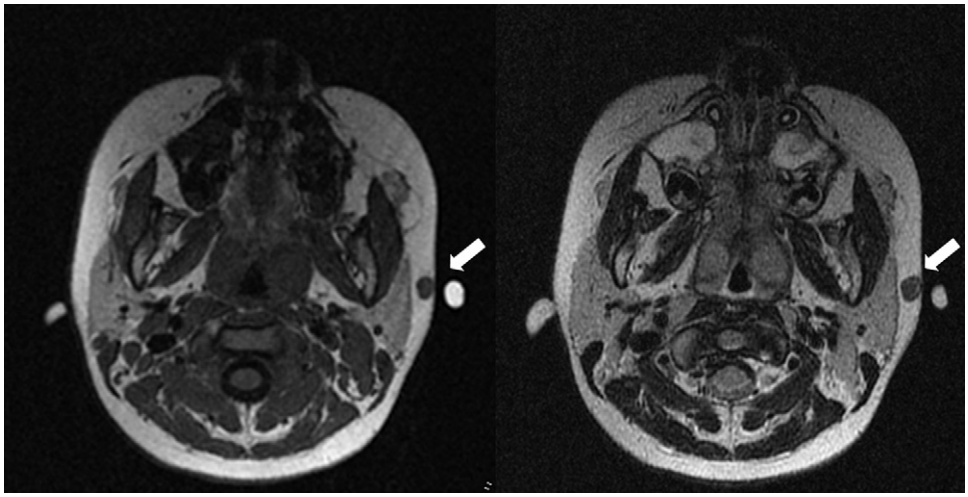

Fig. 3. Magnetic resonance imaging (MRI) demonstrating a well-defined, 8 mm in size, oval mass in the subcutaneous tissue of the right parotid-masseteric region (left: T1-weight signal intensity, right: T2-weighted signal intensity).

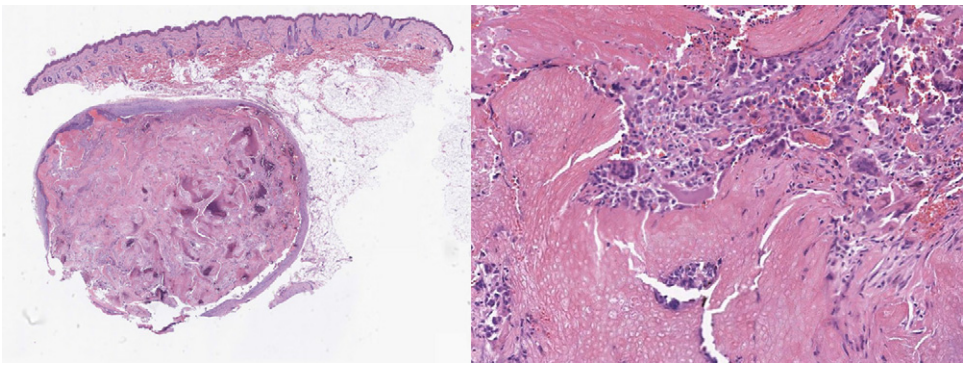

Fig. 5. Histopathologic findings in the specimen (hematoxylin–eosin stain, left: 4×, right: 20×). The tumor was encapsulated by fibro-connective tissue in the subcutaneous tissue and mainly composed of eosinophilic ghost cells and partly with basophilic portions in hyalinized fibrous stroma. Calcification was also noted, accompanied with mild chronic inflammatory infiltration with multinucleated giant cells.

tomographic (CT) scan, it is demonstrated as a noninfiltrating mass containing calcifications located within the subcutaneous tissues [11]. The ultrasound (US) findings demonstrated a well-defined nature of the lesions, internal calcification, and a peripheral hypoechoic rim or a complete echogenic mass with strong posterior acoustic shadowing [12]. Some authors, however, insisted that radiologic imaging is of little diagnostic value for pilomatrixoma to differentiate it from other subcutaneous tumors with any certainty [1,8]. In our case, preoperative US and MRI gave us useful information that the tumor existed just under the skin as well as with the lack of continuity with the parotid gland and the masseter muscle, although we could not diagnose the lesion as pilomatrixoma preoperatively. However, our echogenic pattern of a solid hypoechoic and avascular component informed us that the mass would be a nodule partially calcified with internal calcareous formation.

The characteristic histological appearance of a pilomatrixoma is nests of small basaloid cells that undergo keratinization. And foreign body reaction, calcification, and ossification are common as secondary changes. Although differential diagnosis is varied, pilomatrixoma, first of all, should be differentiated from benign skin regions such as epidermal and dermoid cysts as well as skin malignancies [8]. In the head and neck regions, these regions often present as intraparotid or periparotid tumors and may be confused with benign mixed tumors of the parotid [8]. The other differential diagnosis should include vascular malformation, branchial cyst, hemangioma, inflammatory lesion, calcinosis cuitis, and ossifying hematoma in our fields [3,13].

Once the diagnosis was confirmed, management consists of radical excision. Yoshimura et al. recommended that the lesion should be excised completely, together with the adherent skin [2]. Especially in the cheek, the protection of facial nerve function is important. In our case, preoperative ultrasound findings and MRI allowed us useful information that the tumor existed just under the skin and the risk of facial palsy would be relatively low.

4. Conclusion

We present a case of calcifying epithelioma in a 7-year-old girl. Preoperative US and MRI gave us useful information for complete tumor excision that the tumor existed just under the skin and the risk of facial palsy would be relatively low as well as with the lack of continuity with the parotid gland and the masseter muscle.

References

[1] Duflo S, Nicollas R, Roman S, et al. Pilomatrixoma of the head and neck in children: a study of 38 cases and a review of the literature. *Arch Otolaryngol Head Neck Surg* 1998;124:1239–42.
[2] Yoshimura Y, Obara S, Mikami T, et al. Calcifying epithelioma (pilomatrixoma) of the head and neck: analysis of 37 cases. *Br J Oral Maxillofac Surg* 1997;35:429–32.
[3] Strobl H, Emshoff R. Pilomatrixoma of the cheek: report of case. *J Oral Maxillofac Surg* 1995;53:1355–7.
[4] Machida J, Kawai T, Ogi N. Calcifying epithelioma in the cheek (report of two cases). *Aichi-Gakuin Dent Sci* 1994;7:71–7.
[5] Xia J, Urabe K, Moroi Y, et al. Beta-catenin mutation and its nuclear localization are confirmed to be frequent causes of Wnt signaling pathway activation in pilomatricomas. *J Dermatol Sci* 2006;41:67–75.
[6] Choo HJ, Lee SJ, Lee YH, et al. Pilomatricomas: the diagnostic value of ultrasound. *Skeletal Radiol* 2010;39:243–50.
[7] Kumaran N, Azmy A, Carachi R, et al. Pilomatrixoma-accuracy of clinical diagnosis. *J Pediatr Surg* 2006;41:1755–8.
[8] Danielson-Cohen A, Lin SJ, Hughes A, et al. Head and neck pilomatrixoma in children. *Arch Otolaryngol Head Neck Surg* 2001;127:1481–3.
[9] Hsieh TJ, Wang CK, Tsai KB, et al. Pilomatricoma: magnetic resonance imaging and pathological evaluation. *J Comput Assist Tomogr* 2008;32:320–3.
[10] DeBeuckeleer LH, DeSchepper AM, Neetens I. Magnetic resonance imaging of pilomatricoma. *Eur Radiol* 1996;6:72–5.
[11] Som PM, Shugar JMA, Silvers AR. CT of pilomatrixoma in the cheek. *Am J Neuroradiol* 1998;19:1219–20.
[12] Hwang JY, Lee SW, Lee SM. The common ultrasonographic features of pilomatricoma. *J Ultrasound Med* 2005;24:1397–402.
[13] Hasson O, Shacham R, Nahlieli O, et al. A firm, bluish mass of the cheek in a 17-month-old child. *J Oral Maxillofac Surg* 2002;60:301–4.