Arioua *et al.*
The Egyptian Journal of Otolaryngology (2023) 39:112
https://doi.org/10.1186/s43163-023-00473-5

The Egyptian Journal of Otolaryngology

CASE REPORT Open Access

Parotid pilomatricoma: a case presentation and literature review

Abdelilah Arioua[1,2*], Anouar Benameur El Youbi[1,2], Naouar Ouattassi[1,2], Abdellatif Oudidi[1,2] and Mohamed Noureddine El Amine El Alami[1,2]

Abstract

Background Pilomatrixoma is a rare benign tumor of the hair follicle. It is more common in children and women. The cervical and facial region are the most frequent location for this type of tumor. Due to its variable clinical presentations, it can sometimes be misdiagnosed as a malignant tumor of surrounding tissues, as described in our case.

The diagnosis confirmation is pathological.

Case presentation We report the case of a 34-year-old patient, who presented a swelling in the right parotid region gradually increasing in size for two years, with no facial asymmetry. Clinical examination revealed a rounded, well-defined mass, fixed to the superficial plan, without any cervical lymph nodes. The MRI showed a tumor that seems to be evolving from the superficial lobe of the right parotid, suggesting a pleomorphic adenoma. Total excision of the tumor without margins was performed, preserving the parotid tissue and the facial nerve. The postoperative course was normal; and the pathological study confirmed the diagnosis of pilomatrixoma.

Conclusion Pilomatrixoma is a rare benign tumor but it remains a differential diagnosis of large tumors of the neck and parotid. It is mainly a pathological diagnosis, The treatment of choice still complete surgical excision.

Keyword Pilomatrixoma; Parotid tumor; Calcifying epithelioma; Cervical surgery

Background

Pilomatricoma, or calcifying epithelioma of Malherbe, is a rare benign tumor that develops from the cells of the hair follicles of the skin. It was first described in 1880 by Malherbe and Chenantois, but in 1961 the name "pilomatricoma" was proposed by Forbes and Helwig [1, 2]. Review by according to the literature, the pilomatrixoma occurs in children and young adults below 30 years of age [3].

In general, pilomatricoma tumors can be solid, partially solid or cystic, and may contain deposits of calcium, which explains why it's called "calcifying epithelioma of Malherbe" [1]. The preferred approach for treatment is surgical excision with clear margins. This also reduces the risk of malignant transformation [4].

We report the case of a parotid location of pilomatrixoma; initially making it difficult to differentiate from a parotid gland tumor. The final diagnosis was confirmed by histology.

Case presentation

A 34-year-old patient, with no particular medical history, presented a swelling in the right parotid region evolving for 2 years, gradually increasing in size, with no facial asymmetry (Fig. 1). Clinical examination revealed a rounded, firm, painless, well-defined mass, fixed to the superficial plane and mobile to the deep plane, measuring about four centimeters in size. The overlying skin was red without other inflammatory

*Correspondence:
Abdelilah Arioua
arioua.abdelilah@gmail.com
[1] ENT Head & Neck & Maxillofacial Surgery Department, Hassan II University Hospital, Fez, Morocco
[2] Anatomy, Surgery and Anesthesiology Laboratory, University of Sidi Mohammed Ben Abdellah, Fez, Morocco

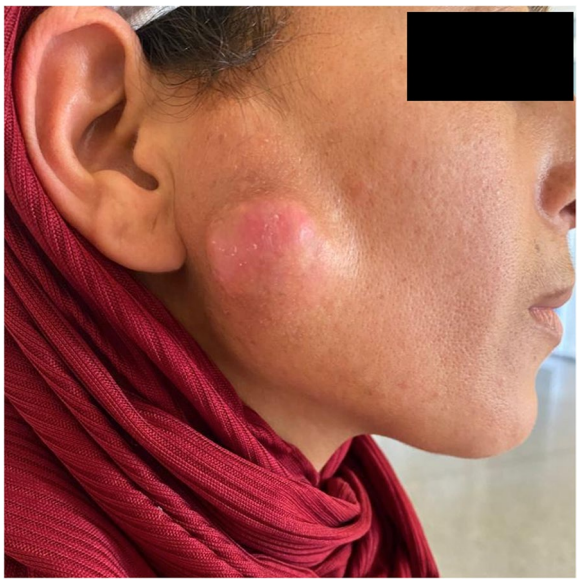

Fig. 1 Mass in the right parotid region

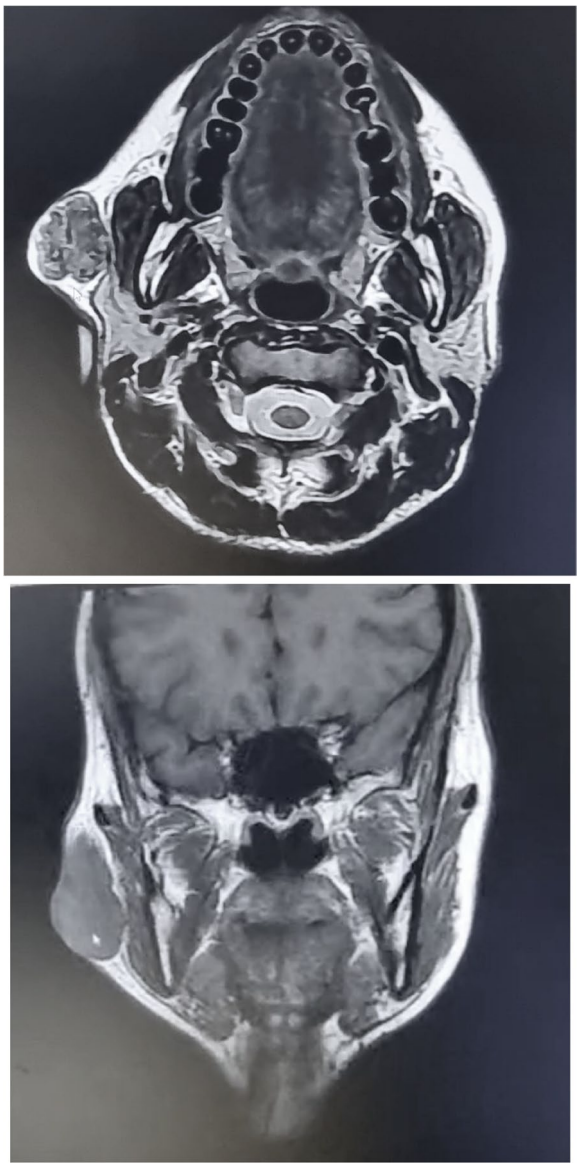

Fig. 2 MRI: tumor with an extension to the right parotid superficial lobe

signs. There was no trismus; or compression of the oropharynx. There were no cervical lymph nodes.

The patient had an MRI that showed a tumor in the right parotid region, with a low T1 and T2 signal, non-restrictive to diffusion, enhancing homogeneously after gadolinium injection, well-encapsulated, measuring 4.6 cm in size, and seems to be evolving from the superficial lobe of the right parotid. Apparent diffusion coefficient (ADC) was calculated to1.1, suggesting a pleomorphic adenoma (Fig. 2).

The patient was informed about the surgery procedure. Our approach initially consisted in performing a superficial parotidectomy. Given the tumor size and its fixity to the skin, we performed a modified incision instead of the classic Lazy-S incision, taking all the infiltrated skin. Total excision of the tumor without margins was performed, the parotid tissue and the facial nerve was not dissected. We didn't perform an intraoperative frozen section examination.

The postoperative course was good; facial motility was normal, and the patient returned home 2 days after surgery. The pathological study confirmed the diagnosis of pilomatricoma.

Histologically, it is a tumor proliferation made up of mummified epithelial masses and basophilic cell clusters without abnormal mitosis. Calcifications are present, with no signs of malignancy (Fig. 3).

Discussion

Pilomatricoma, also known as Malherbe's calcifying epithelioma [1], is a rare benign tumor with an incidence reported between 0.5% and 1.6% [5]. It arises from the matrix of the hair follicle and occurs before the age of 30 in 60% of cases, with 40% of tumors developing before the age of 10, and the highest incidence observed between 8 and 13 years of age [6]. It is more common in females, with a female-to-male ratio of approximately 3:2 [6]. Our reported case confirms the female predominance; and occurs in an adult patient.

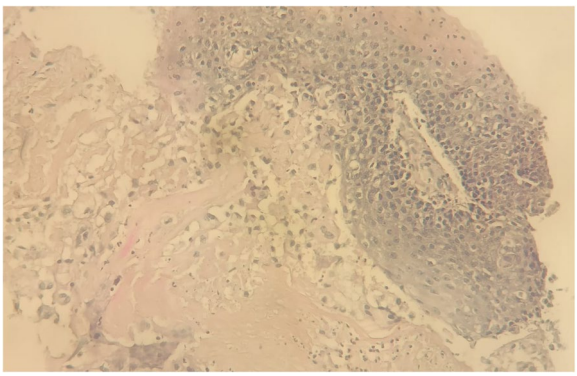

Fig. 3 Pilomatricoma: tumor proliferation made up of mummified cell masses and non-atypical basophilic cells (HES * 100)

Over 75% of pilomatrixomas, which are most commonly found on the head and neck, are located on the scalp, face, neck, or arms [7]. The parotid region is a frequent location for these tumors [7].

Pilomatricomas in the parotid region can be mistaken for parotid neoplasms [8]. Other common differential diagnoses for pilomatricomas in the head and neck have been described in the literature, including sebaceous cyst, ossifying hematoma, giant cell tumor, chondroma, dermoid cyst, foreign body reaction, degenerative fibroxanthoma, and cutaneous osteoma [9]. Diagnosis mainly relies on histopathological examination, with a high rate of diagnostic error [5]. Clinical examination of parotid pilomatricomas reveals a solid or cystic, slow-growing, mobile, and painless mass [8]. The overlying skin may have a reddish or bluish coloration, sometimes it looks like a vascular or malignant tumor infiltrating the cutaneous tissue. Deep extension may have been close to the superficial lobe of the parotid [8]. CT scan or MRI can differentiate preauricular pilomatricomas from parotid tumors, it also can evaluate the extension of voluminous tumors and their exact location near to noble structures [7].

Surgical excision of pilomatricomas is necessary because they do not regress spontaneously. The margins of excision should be clear, although margins of 1 to 2 cm recommended by some authors are likely excessive due to the benign nature of the disease. Malignant transformation is rare, with less than 20 reported cases, and usually occurs in older individuals. After a complete excision, pilomatricomas rarely recur, with reported rates ranging from 0 to 3% [4]. Therefore, surgical excision remains the treatment of choice, but long-term follow-up is essential [10].

Conclusion

Pilomatricoma is a rare benign tumor arising from the matrix of the hair follicle. It is mainly a pathological diagnosis, but it remains a differential diagnosis of large tumors of the neck and parotid. The treatment of choice is complete surgical excision of the tumor. Recurrence and malignant degeneration of the lesion are uncommon.

Acknowledgements
Not applicable

Authors' contributions
AA was involved in diagnosis procedures, surgery, and manuscript drafting. ABE was involved in literature review and drafting of the manuscript. NO and AO were involved in surgery and manuscript revision. MNA reviewed the manuscript for insightful remarks. All authors read and approved the final manuscript.

Funding
Not applicable.

Availability of data and materials
The datasets generated and/or analyzed during the current study are not publicly available due to patient's data confidentiality but are available from the correspondent author on reasonable request.

Declarations

Ethics approval and consent to participate
The IRB of our institution approved our study and the patient signed the consent to participate to the study. Our IRB is CEHUF (comité d'ethique hospital-universitaire de Fès). Email.Comite.ethique.fes@usmba.ac.ma.

Consent for publication
An informed consent for publication purpose was obtain from the patient. Written consent is available.

Competing interests
The authors declare that they have no competing interests.

Received: 7 May 2023 Accepted: 23 June 2023
Published online: 14 July 2023

References

1. Malherbe A, Chenantais JE. Note sur l'epitheliome calcifie des glandes sebacees. Progres Med Par. 1880;8:826–8. [Google Scholar]
2. Forbis R, Helwig EB (1961) Pilomatrixoma (calcifying epithelioma). Arch Dermatol 83:606–618
3. Behnka N, Schulte K, Ruzicka T, Megahed M (1998) Pilomatrixoma in elderly individuals. Dermatology 197:391–393. https://doi.org/10.1159/000018040
4. Giant pilomatricoma involving the parotid: case report and literature review: Gerhard S Mundinger , Derek M Steinbacher, Justin A Bishop, Anthony P Tufaro. Division of Plastic, Reconstructive, and Maxillofacial Surgery, Johns Hopkins Hospital, 601 N. Caroline Street, Baltimore, MD, USA. gsm@jhmi.edu
5. J. Dang, J. Sun, W. Wang et al., Pilomatricoma in the right parotid region: a case report and review of the literature, Asian J Surg, https://doi.org/10.1016/j.asjsur.2022.12.029
6. Friedrich W. Moehlenbeck, MD (1973) Pilomatrixoma (Calcifying Epithelioma) A Statistical Study Arch Dermatol;108(4):532–534.https://doi.org/10.1001/archderm.1973.01620250020004
7. Pilomatrixoma in the parotid region : J Laryngol Otol. 2001;115 12:1026–1028.https://doi.org/10.1258/0022215011909693

8. Brandner MD, Bunkis J (1986) Pilomatrixoma presenting as a parotid mass. Plast Reconstr Surg 78(4):518–21. https://doi.org/10.1097/00006534-198610000-00016
9. Head and neck pilomatrixoma : M Rotenberg, O Laccourreye, R Cauchois, L Laccourreye, M Putterman, D Brasnu. DOI: https://doi.org/10.1016/s0196-0709(96)90010-1
10. Birman MV, McHugh JB, Hayden RJ, Jebson PJ (2009) Pilomatrixoma of the forearm: a case report. Iowa Orthop J 29:121–123

Publisher's Note