Keloid-like Pilomatricoma of the Auricle: A Case Report and Review of Literature

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Abstract
Pilomatricoma is a rare benign skin adnexal tumor arising from hair matrix cells. It usually manifests as a firm-to-hard, well-circumscribed, nontender nodule often mistaken as a dermoid cyst. However, its clinical presentation has been reported to mimic keloid. In addition, although it occurs most frequently in the head and neck area, pilomatricoma is rarely reported on the auricle. Herein, we reported a case of pilomatricoma occurring on the posterior part of a 9-year-old girl's auricle. Unlike classic pilomatricoma, the tumor closely resembled a keloid. Nevertheless, histological examination following excision of the tumor revealed a dermal tumor arranged in solid nests consisting of basaloid and ghost cells along with foreign body reaction, calcification, and ossification, confirming the diagnosis of pilomatricoma. The rarity, atypical clinical presentation, and unique location of this tumor contributed to the misdiagnosis of this case. Therefore, we reviewed previous cases of pilomatricoma of the auricle reported in the literature to provide a comprehensive understanding of this rare entity.

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Introduction

Pilomatricoma, which is also known as pilomatrixoma or calcifying epithelioma of Malherbe, is an uncommon benign skin adnexal tumor arising from primitive epidermal basal cells that differentiate into hair follicle matrix cells [1]. Several studies have reported a low overall incidence of this tumor, estimated to be only 0.001%–0.003% of all dermatohistopathologic samples examined and only approximately 0.12% of all cutaneous neoplasms [2, 3]. The exact pathogenesis is unclear; however, some studies have linked it to a mutation in the CTNNB1 gene, which encodes beta-catenin, a component of cadherin protein necessary for the terminal differentiation of hair follicles [2].

Pilomatricoma can occur at any age, but it is mostly found in children and young adults. In general, it is presented as a painless, firm dermal nodule that attaches to the overlying skin, but it is mobile from the subcutaneous layer. However, diverse clinical variants have been described, including perforating, anetodermic, proliferating, and pigmented types [4, 5]. Consequently, pilomatricoma may mimic various skin conditions, such as epidermoid and dermoid cysts, trichilemmal cysts, and steatocystoma [6]. Diagnosis of pilomatricoma is dependent on histologic findings, which are characterized by islands of tumors in the dermal or subcutaneous layer, encompassing two types of cells, namely, basaloid matrixal cells in the periphery and classic anucleated “ghost” cells toward the center, with variable areas of calcification [7].

Various studies have reported that the head and neck area is the most common location of this tumor, with the preauricular area being the most common site [2, 8, 9]. However, although rarely reported, pilomatricoma cases have been found in unique locations, such as the auricle. Consequently, clinicians are less familiar with this entity. The rarity of this tumor, low incidence of cases, and diverse clinical presentations have led to a high number of misdiagnoses. Studies have demonstrated that the overall preoperative diagnostic accuracy of pilomatricoma could be as low as 1.1% [3].

Here, we reported a unique case of pilomatricoma of the auricle, which was misdiagnosed as a keloid. We also performed a brief review of the literature to evaluate the epidemiological and clinical features of pilomatricoma on this unusual location to increase the awareness of clinicians on this rare entity and minimize misdiagnoses in the future.

Case Report

A 9-year-old girl presented to our dermatology and venereology outpatient clinic with a 2-month history of a lump on the back of her left ear. The lesion was preceded by a small wound that healed and became a lump. Her mother had tried to give a topical antibiotic and topical corticosteroid cream, but the lesion did not regress. Over 2 months, the lump grew bigger, and later, it was accompanied by mild tenderness. No history of trauma was reported; however, her mother admitted that the girl’s father had a history of keloid.

Upon physical examination, a nontender nodule measuring 1.2 cm × 1 cm × 0.3 cm with a bluish-red color and a smooth surface was observed on the posterior part of the antihelix of her left auricle (shown in Fig. 1). The consistency was firm on palpation and free from the underlying structure upon movement. Based on the history and physical examination, a clinical diagnosis of keloid was made, and complete excision with suprakeloidal flap was planned. However, during surgery, the tumor showed a white mass with an irregular surface; thus, pilomatricoma was suspected.

Histopathologic examination demonstrated a well-demarcated dermal tumor arranged in solid nests, which consisted of monomorphic basaloid cells at the periphery and ghost cells
toward the center. Foreign body reaction, calcification, and ossification were also observed (shown in Fig. 2). These findings were consistent with the features of pilomatricoma.

**Discussion/Conclusion**

In 1880, Malherbe and Chenantais first described a case of calcifying benign tumor thought to be originated from the sebaceous glands and labeled it “calcifying epithelioma of Malherbe.” Further studies discovered that this tumor arises from hair matrix cells; therefore,
in 1961, Forbis and Helwing proposed the term “pilomatrixoma” to describe this entity. In 1977, this term was adapted to “pilomatricoma” [10].

A literature review was conducted by performing a PUBMED search using the combination of terms: “(pilomatricoma OR pilomatrixoma OR ‘calcifying epithelioma of Malherbe’) AND (auricle OR auricular OR ear OR pinna).” A total of 60 articles published from 1969 to 2021 were found in the literature search. We included reviews, retrospective studies, and case reports. Articles that were not relevant (i.e., not pilomatricoma and nonauricular area), non-English articles, and articles from which full articles were not retrievable were excluded. After screening, a total of 16 case reports were included in the analysis (shown in Table 1). In addition, two retrospective studies and one review article were included for additional references in the discussion.

Pilomatricoma primarily occurs in the hair-bearing skin, and it has never been reported on the palms or soles [2, 4, 7]. Various studies have demonstrated that pilomatricoma can be found primarily in the head and neck area, comprising around 64%–84.2% of all pilomatricoma cases, followed by the upper extremities with around 22% of cases [2, 4, 5]. Of the head area, the cheek or preauricular region was commonly reported [2, 8, 9]. By contrast, pilomatricoma of the auricle was rarely reported. One retrospective study has shown that pilomatricoma of the auricle may lead to misdiagnoses because of its close clinical resemblance to other skin conditions such as a dermoid cyst compared with cases involving other body parts [6]. In addition, given the rarity of its occurrence in the auricle, clinicians may initially consider other differential diagnoses that are commonly found in this area, such as squamous cell carcinoma, or those that almost exclusively occur in the auricle, such as tumors of the ceruminous gland [11].

Based on the 16 case reports that we retrieved, pilomatricomas were located in various parts of the auricle (shown in Fig. 3). Five cases (31.25%) were located in the earlobe [10, 12–15], followed by the helix [3, 16–18] (4 cases/25%), outer ear canal [19–21] (3 cases/18.75%), antihelix anterior [22, 23] (2 cases/12.5%), helical root [24], and cymba conchae [11] (1 case each/6.25%). None of those cases reported pilomatricoma on the posterior part of the antihelix as in our case.

Epidemiologically, pilomatricoma has been reported to frequently occur in the first 2 decades of life [5]. Similarly, we found that the majority of patients (10 cases/62.5%) with pilomatricoma of the auricle were aged below 20 years. Some studies have suggested a bimodal distribution with the second smaller peak at the age of 50–65 years [7], but only one patient within this age range was reported in our findings. Based on various studies [2, 4, 5, 8], we found a slight female preponderance, with a female-to-male ratio of 1.3:1.

Pilomatricoma is typically painless. However, pain may occur because of associated inflammation or ulceration [13]. In our findings, the majority of pilomatricoma cases on the auricle area were asymptomatic (10 cases/62.5%). Of those, two cases were reported to progress and become painful as the tumor ulcerated or grew bigger [19, 24]. Similarly, our patient reported mild tenderness as the tumor grew in size. Four cases (25%) of pilomatricomas reported tenderness possibly because of concomitant inflammation [12, 15, 18, 23]. Notably, 3 cases (18.75%) of pilomatricoma on the outer ear canal presented with severe pain in the ear and reduced hearing capacity, which may occur before the tumor was evident, and clinicians should be aware of these features [19–21].

Pilomatricoma is usually less than 1.5 cm in diameter [25]. However, cases of giant pilomatricoma have been reported, and this term was applied to tumors with a diameter of more than or equal to 4 cm [26]. Of all case reports that we reviewed, information on the size of the tumor was available for 13 (81.25%) lesions. In line with the literature, we found that the majority (7 cases/43.75%) of pilomatricomas of the auricle measured less than or equal to 1.5 cm, as in our case. Five cases (31.25%) measured between 1.5 and 4 cm, whereas 1 case (6.25%) described a 5-cm giant pilomatricoma [12]. In addition, we found that all cases of pilomatricoma on the
| Reference | Age, years | Sex | Location (s) | Size, cm | Symptom(s) | Preoperative diagnosis | Diagnostic methods | Treatment |
|-----------|------------|-----|--------------|---------|------------|------------------------|-------------------|-----------|
| Yadav [20], 1969 | 45 | M | Outer ear canal | NA | Deafness, recurrent pain | NA | Histopathology, hearing test | Excision |
| Pathak [21], 1973 | 47 | M | Outer ear canal | NA | Deafness, pain | Keratosis obturans | Histopathology | Excision |
| Vinayak et al. [19], 1992 | 9 | M | Outer ear canal | 0.5 | Painless → painful (large) | Pyogenic granuloma | Histopathology | Excision |
| Rao and Ramnarayan [14], 1992 | 5 | F | Earlobe (inferomedial) | 3 | Painless | Dermoid cyst | Histopathology | Excision |
| Sevin et al. [12], 1995 | 8 | F | Earlobe | 5 × 4 × 4 | Tender | NA | Histopathology | Excision with flap |
| Nasse et al. [18], 2007 | 72 | M | Helix | 2.8 × 1 × 0.6 | Tender | NA | Histopathology | Excision |
| Pant et al. [13], 2010 | 9 | F | Earlobe | 2 × 1.3 × 1 | Painless | Dermoid cyst | Histopathology, FNAC | Excision |
| Hassanein et al. [24], 2011 | 8 months | M | Helical root | 1.2 | Painless → painful (large, ulcerated) | Infantile hemangioma | Histopathology, Doppler examination | Excision |
| Li et al. [22], 2011 | 3 | M | Antihelix | NA | Painless | NA | Histopathology | Excision |
| Upile et al. [23], 2012 | 46 | F | Antihelix | 1.4 × 1 | Tender | Pyogenic granuloma | Histopathology | Wide excision with a skin graft |
| Rajeshwary et al. [3], 2013 | 4 | M | Helix | 1 × 2 | Painless | Calcified dermoid cyst | Histopathology, FNAC | Excision |
| Nigam and Singh [16], 2014 | 19 | F | Helix | 1.5 × 1.5 | Painless | NA | Histopathology, FNAC | Excision |
| Dutta et al. [17], 2015 | 24 | F | Helix | 1.5 × 1 | Painless | Keloid | Histopathology | Excision |
| Van Gysel et al. [15], 2016 | 6 months | F | Earlobe (posterior) | 3.5 × 1.5 | Tender (when inflamed) | Hemangioma | Histopathology, USG | Excision |
| Inagaki et al. [11], 2019 | 52 | F | Cymba conchae | 1 | Painless | NA | Histopathology, MRI | Excision with flap |
| Jeong et al. [10], 2021 | 18 months | F | Earlobe (posterior) | 1.5 × 1 | Painless | Tumor | Histopathology | Excision |
| Our case | 9 | F | Antihelix (posterior) | 1.2 × 1 × 0.3 | Painless → mild tenderness | Keloid | Histopathology | Excision |

NA, not available; FNAC, fine-needle aspiration cytology; USG, ultrasonography; MRI, magnetic resonance imaging.
auricle, including our case, were presented as a solitary lesion. Multiple pilomatricomas in individuals have shown an association with familial conditions or genetic syndromes, such as myotonic dystrophy, Gardner syndrome, Turner’s syndrome, Steinert disease, and Rubinstein–Taybi syndrome [10, 27]. Moreover, we found one case of solitary pilomatricoma of the auricle, which was accompanied by myotonic muscular dystrophy in a 5-year-old girl [14]. No other symptoms or signs of the abovementioned conditions were identified in our case.

The clinical presentations of pilomatricoma are diverse. Previous studies have described several clinical variants of pilomatricoma: the most common one is the classic/mass type, followed by more rare variants such as perforating/ulcerated, anetodermic, proliferating, pigmented, and familial types [4, 5]. Classic pilomatricoma is characterized by a firm-to-hard or cystic, well-circumscribed, nontender nodule that adheres to the overlying skin, but it is relatively mobile from the underneath subcutaneous structure. It can have a bluish or reddish discolorization, but it can also appear as skin color or semi-transparent [7]. Perforating pilomatricoma showed ulceration on the nodule from which the inner calcified material extruded [17]. Based on the description of lesion in each article, we found 11 cases (68.75%) that seemed to be of classic types and 4 cases (25%) of ulcerated types. In addition, 1 case was misdiagnosed as a keloid because of clinical resemblance [17], which is similar to our case. Keloid-like pilomatricoma was not clearly described, but it was characterized by excessive growth beyond the original lesion area and reported to comprise only 1.2% of all variants of pilomatricoma [4]. Few reports have included this clinical presentation as a variant of anetodermic pilomatricoma [28, 29].

Given the rarity of this tumor and variability of its clinical presentations, pilomatricoma is often misdiagnosed. Studies have reported a low preoperative diagnostic accuracy, ranging from only 1.1%–55.5% of cases [3, 7]. Based on our findings, 10 cases (62.5%) provided information on the preoperative clinical diagnosis. However, none of these cases were correctly diagnosed as pilomatricoma, including our case. The unusual location of pilomatricoma on the auricle may contribute to misdiagnoses [11]. Of all the cases that we reviewed, pilomatricomas were commonly mistaken as dermoid cysts (3 cases/18.75%), followed by pyogenic granulomas and hemangiomas (2 cases each/12.5%). In our case, pilomatricoma was thought to be a keloid, similar to a case reported by Dutta et al. [17].

Histopathologic examination confirmed the diagnosis in all 16 cases and in our case. In general, pilomatricoma is characterized by the presence of a well-circumscribed tumor in the dermal or subcutaneous tissue, which consists of islands of epithelial cells containing two characteristic cells: nucleated basaloid matrical cells in the periphery and anucleated “ghost”
or “shadow” cells in the center. The surrounding stroma may contain multinucleated foreign body giant cells [7]. The ghost cells are believed to be the remnants of ectodermal keratinocytes, which fail to differentiate into mature hair follicles and later undergo dystrophic calcification, and such cells are considered histologic hallmarks of pilomatricoma [5]. During the development of pilomatricoma, progressions occur over time, manifesting as changes in histologic appearance and clinical presentation of the tumor. In addition, early lesions have been described to be dominated by basaloid cells, and later stages might show remarkable proportions of ghost cells with calcification, contributing to the hard consistency of fully developed tumors on palpation [11]. Basaloid and ghost cells were present in the histological sections of 16 cases (100%) that we analyzed, which is in contrast to a previous retrospective study that reported only 13.7% pilomatricomas in all locations presented with both cells [5]. Moreover, such cells were evident in our case, and we found that calcification was present in only ten other cases (62.5%), possibly corresponding to the tendency of this tumor to be excised early, during which calcification has not been evident [11]. Furthermore, in our case, we identified the presence of ossification, another marker of late lesions, which was found in only two other cases (12.5%) of pilomatricoma of the auricle.

Other diagnostic examinations have been shown to be of limited value. The commonly performed procedure was fine-needle aspiration cytology (FNAC), which will show the triad of basaloid, ghost, and giant cells in the case of pilomatricoma [5]. Based on our findings, three lesions (18.75%) underwent FNAC before it was confirmed by histology but only one (33.33%) correctly pointed out the diagnosis of pilomatricoma. FNAC has been demonstrated to be accurate in only 40% of cases and particularly misleading if the lesion was still at an early stage or if the aspirate was taken from the periphery [5]. Several imaging examinations, such as X-ray, ultrasound, computed tomography, or magnetic resonance imaging, are rarely used and considered important for detecting calcification or differentiating pilomatricoma from malignant tumors such as vascular or lymphatic [5, 7]. However, Hwang et al. [30] demonstrated that ultrasonography could increase the accuracy of clinical pilomatricoma diagnosis from 33% to 76%. Therefore, ultrasonography could be performed in the case of suspected pilomatricoma with doubtful clinical findings prior to surgery [8]. In our case, no other diagnostic examinations were performed because the lesion clinically and closely resembled keloid.

Spontaneous regression has never been reported; thus, surgical excision is the preferred treatment method for pilomatricoma. Most cases of pilomatricoma of the auricle (13 cases/81.25%) were treated with simple excision. One case of giant pilomatricoma on the earlobe [12] and one on the cymba conchae [11] were reported to require skin flaps following excision. At present, no guidelines on the appropriate margins of excision for pilomatricoma have been constructed, and most cases that we found did not include information on the margins. However, Upile et al. [23] reported an excision with a 4–6 mm margin, which may be necessary in the case of ulcerative pilomatricoma or suspected malignant pilomatrix carcinoma. Overall, recurrence rates of pilomatricoma were low, which were reported to be only around 1.4% [5]. Recurrences were frequently due to incomplete resections or rarely due to malignant transformation to pilomatrix carcinoma [4, 5]. Based on our findings, none of the cases of pilomatricoma of the auricle reported recurrences after a follow-up period of 5 months to 2 years following excision. Furthermore, recurrence has not been observed until 2 months after surgery in our case.

Transformation of pilomatricoma to pilomatrix carcinoma is rare, and it was reported to only occur in adults. Pilomatrix carcinoma manifests as a firm, nontender nodule located on the head and neck, which clinically and closely resembles pilomatricoma [7]. Some studies have suggested the proliferating type of pilomatricoma to be a precursor of pilomatrix carcinoma, which is also known as aggressive pilomatricoma [5]. The high mitotic rate of basaloid cells, cellular atypia, central necrosis, and infiltration into the adjacent skin, soft tissue, and vessels were histologic features indicating a malignant transformation of pilomatricoma [5, 7].
None of the cases that we analyzed either reported a malignant transformation of pilomatricoma or described the presence of these atypical histologic features.

**Conclusions**

Pilomatricoma commonly occurs on the head and neck; however, auricles are rarely reported. Although most epidemiologic and clinical features may be similar to those occurring in other areas, pilomatricoma of the auricle has peculiar clinical presentations such as those mimicking keloid and unfamiliar symptoms, including severe ear pain and reduced hearing. Furthermore, given the rarity of the location, pilomatricoma of the auricle seems to be frequently misdiagnosed, particularly as dermoid cysts. Histological examination remains the gold standard for pilomatricoma on any location, which is confirmed by the characteristic findings of dermal tumors composed of basaloid cells, ghost cells, and calcification, albeit not all components should be observed during diagnosis. Complete surgical excision with a clear margin is the preferred method of treatment for pilomatricoma of the auricle. A wide margin is necessary for pilomatricoma cases that may transform into malignant pilomatrical carcinoma.

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**Statement of Ethics**

The authors state that the patient’s parents gave written informed consent to publish this case and any accompanying images. Any information revealing the patient’s identity is avoided. The study has been done according to the Declaration of Helsinki. Ethical approval was not required for this study according to the national guidelines.

**Conflict of Interest Statement**

The authors have no conflicts of interest to declare.

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**Author Contributions**

Shinta Trilaksmi Dewi: manuscript concept and design, data acquisition and analysis, and manuscript writing and finalization. Hanggoro Tri Rinonce: histopathology examination interpretation and consultant and critical revision of the manuscript. Kristiana Etnawati: critical revision of the manuscript. Yohanes Widodo Wirohadidjojo: manuscript concept and design and critical revision of the manuscript.
Data Availability Statement

All necessary data from the study have been included in the manuscript. However, if required, additional findings are available from Dr. Shinta Trilaksmei Dewi upon request.

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