

What is the clinical utility of histological diagnosis between pilomatrixoma and trichilemmal cyst in children?

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Abstract

Pilomatrixoma and trichilemmal cyst are two benign tumors of hair follicles that can affect children. We performed a retrospective study to histologically differentiate the two tumors to determine if this has clinical utility. For 5 years, we conducted a retrospective study on the patients who underwent surgery and evalua-

ted some parameters (site, recurrence and scar outcome) categorizing them based on their histological type. We treated 71 patients, with an average age of 7.6 years. Most of the lesions were localized on the scalp, head and neck. Histological examination diagnosed 55 pilomatrixomas and 16 trichilemmal cysts. Five patients with pilomatrixoma developed an unaesthetic scar (4 in subscapular region), while the recurrence was present in 10 cases, of which 6 were trichilemmal cysts (37,5% of 16 cases). Both types of lesions healed successfully, however trichilemmal cysts are more susceptible to recurrence.

Introduction

Pilomatrixoma (PM), also known as the calcific epithelioma of Malherbe, is a benign tumor of hair follicles that can affect children. This tumor can be located in all regions where there are hairs but mainly prefers the region of the head and neck. Clinically, it looks like a solitary, slow-growing, hard, dome-shaped cyst. It can cause pain especially when it is in regions where bones are present, and in some cases, it can become infected and cause bad scars. Multiple formations are rare and are usually associated with syndromes. Other skin lesions, such as dermoid cyst, epidermoid cyst or Trichilemmal Cyst (TC), may resemble PM, and a differential diagnosis cannot be made without histology. The purpose of our study is to determine if this type of tumor exhibits distinct behavior in terms of scarring and recurrence, and whether, consequently, it is beneficial to perform a histological diagnosis of the lesion.

Materials and Methods

Between 2018 and 2023, we conducted a retrospective study of patients with skin lesions suspected of being PM. We collected information about age, sex, localization, size, clinical and ultrasound presentation, previous infections, recurrence, treatment and histological findings.

The indication for surgery was a previous injury infection, pain at the site of the injury or a continuous trauma to the area (such as hair or bone protrusions). Before the surgery, each patient had an ultrasound. Surgical excision was performed on all patients under general (sedation) and local anesthesia. An elliptical incision was made oriented along the lines of relaxed skin tension. Transdermal continuous suture was used to treat the wound, except for sites with hair (the scalp) where an interrupted suture was used. A histological examination was performed after removing the skin margin and all lesion. All patients were discharged the same day of surgery. None required antibiotics. Pain control was

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Ethics approval: the Ethics Committee of the University of Campania “Luigi Vanvitelli” (0007953 of 06/04/2020). The study is conformed with the Helsinki Declaration of 1964, as revised in 2013, concerning human and animal rights.

Informed consent: all patients’ guardians participating in this study signed a written informed consent form. Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

Availability of data and materials: all data generated or analyzed during this study are included in this published article.

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achieved with the administration of paracetamol if necessary. A clinical and ultrasound follow-up was conducted at 6, 12 and 24 months after surgery. The wound was assessed by a pediatric surgeon at the 24-month follow up, and each was assigned a score according to the Vancouver Scar Scale (VSS) (vascularity, pliability, pigmentation, height): wounds with a score above 5 were considered bad.

The study was conducted according to the Helsinki principles and was approved by the Ethics Committee of the University of Campania "Luigi Vanvitelli" (0007953 of 06/04/2020).

Results

We collected data from 71 patients over a 5-year period. Of these, 40 were female (57%) and 31 were male (43%). The mean age at presentation was 7.6 years (range 1-17); the mean age at surgery was 8.5 years (range 2 - 17). The size of the excised lesions ranged from 0.5 cm to 3.2 cm. The location of the lesions was as follows: 15 lesions on the scalp, 15 on the head, 15 on the neck, 5 on the dorsal region, 10 on the subscapular region, and 11 on the upper limb (Table 1). Surgery was indicated in 25 cases with previous infections that were treated with antibiotics, 20 cases of pain

at the site of the lesion, and 26 patients who experienced ongoing trauma due to the anatomical position of the scalp and head. Because there are no distinguishing features between the two types of lesions, clinical and ultrasound differential diagnosis was not possible. Each patient was discharged on the same day as the surgery, except for 4 cases that showed eating difficulties (vomit) and were discharged later: the mean discharge time was 12 hours (range 6 - 24). Histological examination was carried out in all cases and allowed the diagnosis of PM (Figure 1) in 55 cases and

Table 1. Localization of the lesions after histological diagnosis.

Localization	Pilomatrixoma (recurrence)	Trichilemmal cyst (recurrence)
Head	11 (2)	4 (1)
Neck	15	0
Scalp	3	12 (5)
Dorsal region	5	0
Scapular region	10 (1)	0
Superior limb	11	0

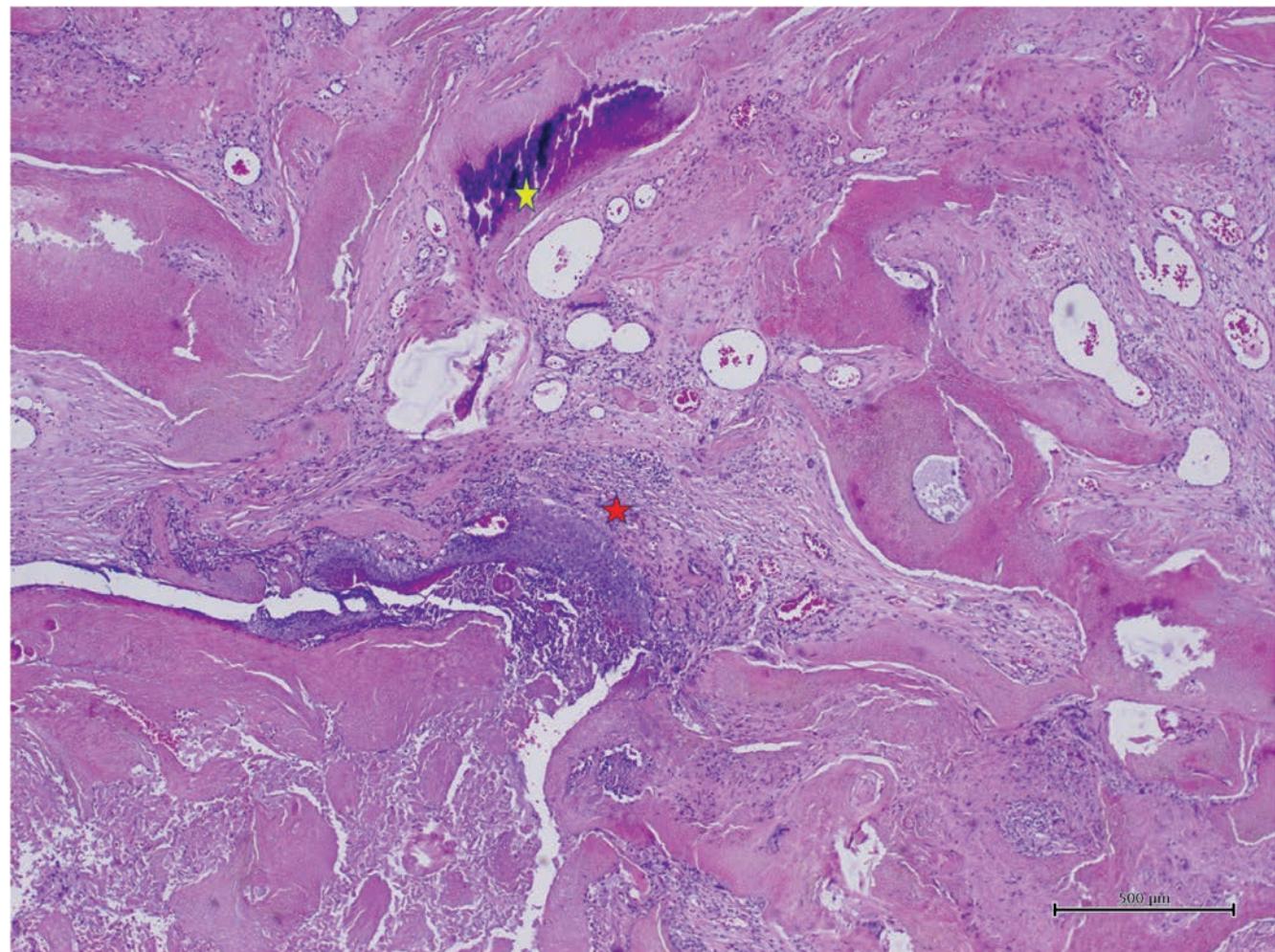


Figure 1. Histological image of pilomatrixoma: low-power view showing sheets of ghost (shadow) cells, with focal basaloid cells (red star) and calcification (yellow star) (hematoxylin and eosin, original magnification 40x).

TC (Figure 2) in 16. PM had a mean size of removed lesions of 2.3 cm (ranging from 1 to 3.2) and TC had a mean size of 1.2 cm (ranging from 0.5 to 1.8). In all cases, margins were free of lesions reported. At 7-day follow-up, no wound dehiscence or infection was observed. The 24-month follow-up was conducted to evaluate the scar and any recurrence. Our patients did not experience pain or itch from the bad scar in any case. All patients diagnosed with TC have reported a good scar, while 5 cases (9.1%) diagnosed with PM had a scar with VSS score > 5 (4 cases of hypertrophic scar and 1 case of neck cheloid). At follow up of 24 months recurrence occurred in a total of 10 cases (14.1%). Our analysis showed that 4 recurrences occurred in patients with PM (7.2% of all PM), with one developing a new lesion in a different location. Additionally, 6 cases were found in patients with TC (37.5% of all trichilemmal cysts), all at the site of the intervention (Table 2).

Discussion

The PM is a benign tumor that develops from hair follicle matrix cells due to a disorder in their growth cycle.¹ It typically

appears as a slow-growing domed solitary mass that can be moved to deep planes. Clinical features of PM include a reddish or bluish color and the 'tent sign', where the skin is stretched over calcification.² In syndromic contexts, such as myotonic dystrophy or Gardner's syndrome, these tumors can take various forms and can be multiple.³

PM is more common in females and typically localizes in the head and neck region, although it can occur anywhere on the body where there are hairs, as observed in our series. Clinically, PM is usually asymptomatic but can cause pain in trauma areas. Several skin tumors have the same characteristics, but the TC is the main clinical imitator, and it also comes from the hair follicle, in particular from the isthmus. TC are most commonly located on the scalp (90% of cases), followed by the face and trunk,⁵ and rarely genitalia, groin, palms and fingertips.⁶

TC are uncommon in pediatric age, in literature few cases of patients under 10 years of age with a diagnosis of TC have been reported.⁷⁻¹⁰

Similar to PM, this cyst is located in the subcutaneous layer and has a higher incidence in females. It may also present foci of calcification, and have a malignant variant (proliferating trichilemmal tumor).⁹ This variant, although considered biologically benign, can be locally aggressive. In rare cases, malignant transformation has been reported, evidenced by regional or distant metastases.¹⁰⁻¹² The proliferating trichilemmal tumor can also occur in children.^{13,14}

The ultrasound features of both types of cysts are similar, showing internal hyperechogenic foci of calcifications and posterior shading. Calcifications are more pronounced in PM. Hyperechogenic areas around the tumor indicate local adipose inflammation. PM is characterized by the presence of a peritumor hypoechogenic edge, indicating a connective tissue capsule.¹⁵ The definitive diagnosis can only be confirmed through histopathological examination.

PM presents as subcutaneous nodules consisting of two layers of cells: an inner layer of ghost cells or shadow cells (without nuclei) and an outer layer of basaloid cells. Calcium deposits are present within the nodules, which are surrounded by a connective tissue capsule.¹⁶ The thickness of the cell layer depends on the maturation stage of the lesion. The TC consists of a single layer of pale cells that become keratinized with calcium deposits.¹⁷ Surgery is curative for both conditions. PM rarely recurs in the same position after complete excision, but TC are more likely to recur due to their multicentric origin.¹⁸ In our case studies, recurrence occurred in 37.5% of cases of TC, but only in 7.2% of patients with PM.

These symptomatic skin lesions should be treated surgically, as they may be associated with inflammation/infection, cosmetic disfigurement, and rarely neoplastic transformation. After surgery, the appearance of the scar is important, especially in pediatric age, as children are still growing and the psychosocial effects are significant. It is also necessary to consider the possibility of recurrence, for cases of CT, in which, despite the removal of the cystic wall, the risk of recurrence remains high.

The risk of wound infection is similar for both lesions and does not depend on the use of antibiotics, but rather on the site of the lesion.¹⁹ As for scalp lesions, it has been widely shown that the presence of hair around the surgical incision does not increase the risk of infection, unlike shaving which can produce microlesions that hinder the healing process. If shaving is necessary, it should be done as little and as close to the surgery as possible. A successful healing process can be ensured with the appropriate suture techniques, a good distribution of elastic tension, and the use of a moisturizing dressing.²⁰ A lot of attention should be paid to the scarring outcome because at certain sites, such as the scalp, it can lead to

Table 2. Data on scar and recurrence by histological type.

	Pilomatrixoma (%)	Trichilemmal cyst (%)
Histological diagnosis	55	16
Scar		
VSS score <5	51 (92.7)	16
VSS score >5	4 (7.3)	0
VSS score >10	1 (1.8)	0
Recurrence same site	3 (5.4)	6 (37.5)
Recurrence same site	1 (1.8)	0

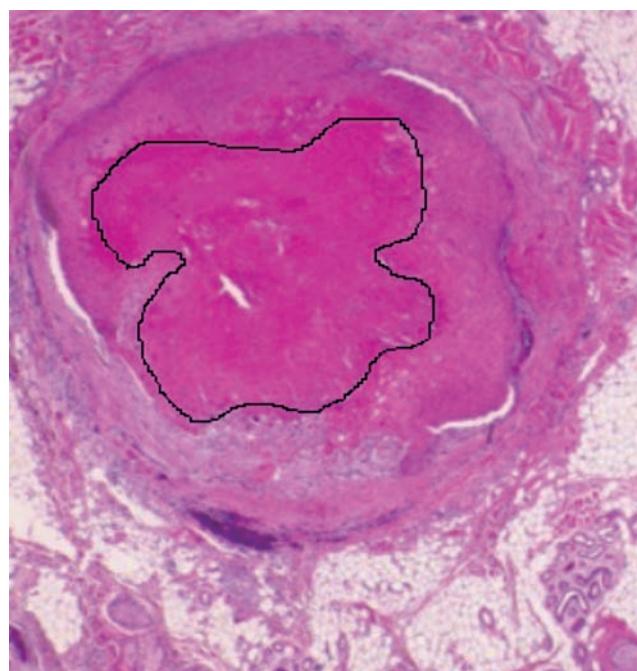


Figure 2. Histological image of trichilemmal cyst: cyst full of keratin (black line) (hematoxylin and eosin, original magnification 20x).

hair loss and create psychological problems in patients, especially in females.

In conclusion, pilomatrixomas are skin lesions that can affect children and are often misdiagnosed with other cystic lesions (especially trichilemmal cysts). The treatment is surgical and a histological examination is mandatory because it is the only way to have an accurate diagnosis. Histologically differentiating the two lesions is important for follow up. In fact, according to our experience, PM carries a greater risk of unaesthetic scar while TC has a higher risk of recurrence.

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