Pilomatricoma in the Upper Extremity After Vaccination: A Case Report

Zeynep Tuzlalı, Burak Kaya, Serdar Mehmet Gültan
Department of Plastic Reconstructive and Aesthetic Surgery, Ankara University School of Medicine, Ankara, Turkey

Abstract

Pilomatricoma (also known as pilomatrixoma) is a rare benign skin tumor arising from hair follicle stem cells; it is also known as calcifying epithelioma of Malherbe. A definitive diagnosis can be made by histopathological examination. Surgical excision of the lesion is recommended for its treatment. The lesion can often be misdiagnosed when differential has not been considered. In the literature, there are few reports of pilomatricoma cases occurring after vaccination. A 21-month-old male patient was admitted to our clinic with a mass on his left arm that occurred after vaccination. The lesion was completely excised. The diagnosis of pilomatricoma was made after histopathological examination. As there were no predisposing factors other than vaccination, we concluded this to be a complication of the vaccination. The pathogenesis of pilomatricoma is unclear, but inflammation and trauma may promote neoplastic development. We report this case to draw attention to this rare lesion and to the possible etiopathogenetic association of vaccination with pilomatricoma.

Keywords: Pilomatricoma, vaccination, calcifying epithelioma of Malherbe, Malherbe tumor

INTRODUCTION

Pilomatricoma (pilomatrixoma), also known as calcifying epithelioma of Malherbe, is a rare benign skin tumor arising from hair follicle stem cells. The lesion was first described in 1880 by Malherbe and Chenantais as a calcified tumor derived from the sebaceous glands. In 1961 Forbis and Helwig proposed the name pilomatricoma. It was later described that the tumor cells resemble the matrix cells in the hair follicle and could therefore be arising from the matrix cells at the base of the hair. Pilomatricoma is an asymptomatic benign tumor which typically presents as a slowly growing, solitary, subcutaneous or intradermal nodule. It is more common among females and usually presents during the first two decades of life. Its etiology is not fully known, but trauma and infection are considered to have a role. It has been reported to possibly be an outcome of a disruption in the cycle of hair follicles. The recommended treatment is surgical excision. While definitive diagnosis can be made with histopathologic examination, pilomatricoma should be considered in the presence of calcification in imaging. However, diagnostic value of radiological imaging is considered to be low. Presence of a rigidly palpable mass is pathognomonic.

In this study, we present the case of a 21-month old male patient that developed pilomatricoma after vaccination to the arm, and review the correlation between pilomatricoma and vaccination in reference to the literature.

CASE REPORT

The 21-month old male patient who was brought to our clinic was evaluated for a rigid, well-defined, mobile hyperemic lesion of 3x3 cm to his lateral left arm. The lesion had emerged after a vaccination at 12 months of age. The patient had Bacillus Calmette-Guérin (BCG) vaccination when he was two-months old and experienced no complications such as discharge, redness or swelling. At the
age of 12 months he had measles-mumps-rubella (MMR), pneumococcal conjugate (PCV), and chickenpox vaccines combined. One week after the shot he developed redness at the injection site which started to swell and harden one month later. The patient had no fever during this course. He did not have any family or trauma history. His vascular and neuromotor examinations were normal, no systemic pathologies were identified. Physical examination showed no axillary lymphadenopathy, and hematologic and biochemical test results were within normal limits. Initially, fat necrosis or subcutaneous abscess was considered. In radiographic imaging, the mass was observed to be a well-circumscribed nodular lesion (Figure 1). CT imaging showed a lobulated, well-circumscribed low-density mass with dense peripheral calcification of about 30x28x26 mm localized to the cutaneous-subcutaneous plane in the middle section of his left arm (Figure 2).

Written consent was obtained from the patient’s family. The patient was taken to surgery under general anesthesia. An incision was made over the mass so as to include the intact skin. Obtuse and sharp dissection was performed and the mass was released from its surrounding tissues (Figure 3). Preserving the adjacent anatomical structures, the mass and part of the intact skin were removed in whole (Figure 4). The macroscopic tissue specimen, which was observed to be well-defined, yellowish-white, rigid and capsular (Figure 5), was sent for pathologic examination. After hemostasis was achieved, cutaneous and subcutaneous tissues were primarily closed. No problems were observed in his controls and the patient was discharged with recommendations. Complications or recurrence were not observed in the early postoperative period. Microscopic examination showed that the material was composed entirely of tumorous tissue. The tumor was identified to consist of groups of basaloid epithelioid cells and wide keratin layers formed through the trichilemmal keratinization of these cells. Ker-
atin layers were seen to consist of “ghost cells” and the case was diagnosed as Pilomatricoma (Figure 6).

DISCUSSION

Pilomatricoma (pilomatrixoma), also known as calcifying epithelioma of Malherbe, is a rare benign skin tumor arising from hair follicle stem cells. Typically, it occurs as an asymptomatic cutaneous or subcutaneous nodule in the head and neck region or the upper limbs. In our case, the tumor was not localized in the head and neck region, and showed no symptoms other than swelling. While its actual incidence is not known, there are studies that report an incidence rate of 3.03% to 0.1%. Several clinical types of pilomatricoma, such as bullous, giant, perforating, multinodular have been described. It has been reported to be associated with the myotonic dystrophy gene, the polyomavirus, Gardner’s syndrome, xeroderma pigmentosum, Turner’s syndrome, sarcoidosis and basal cell nevus syndrome. The tumor is typically asymptomatic, and grows slowly over months to years. Clinically, pilomatricoma presents as solitary, painless, dermal or subcutaneous masses. Lesions can present as soft and cystic masses in the early phase, but characteristically develop into rigid and well-circumscribed masses over time.

While its etiology is not exactly known, trauma and infection are considered to have a role in the development of this type of tumor. In our case, we believe the tumor to be secondary to the trauma and increased inflammation caused by the injection.

The lesion is typically seen before the age of 20. In our case, the 21-month-old male patient falls within this range. Epidermal cyst, calcified lymphadenopathy, ossified or calcified hematomata, foreign bodies, and other benign or malignant soft tissue tumors should be considered for differential diagnosis. In our case, presence of calcification, as revealed by direct radiography, suggested pilomatricoma. It can be treated with surgical excision, as we did in our case. Definitive diagnosis of these tumors can be reached through histopathologic evaluation. In examination, the tumor is seen to consist of basaloid cells, ghost (shadow) cells, keratin filaments, cornified material, and giant cells.

Tumors arising from vaccination scars can be induced in response to trauma, long-term inflammation, delayed wound healing response, scar formation, or the weakened agents used for vaccination. Prophylactic vaccines rarely cause skin complications. Bullous pilomatricoma has been reported to develop after Hepatitis A vaccination in the injection site. In the literature, there are several cases of pilomatricoma reported to occur in the injection site after a BCG vaccine, an influenza vaccine or following intramuscular injections. But we have not come across any reports about a pilomatricoma case that occurred following a combination vaccine (Measles-Mumps-Rubella [MMR], Pneumococcal Conjugate [PCV], and Chickenpox) administered at the age of 12 months. The presented case differs from other pilomatricoma cases in that the tumor has developed following a combination vaccine.

Tumors that arise after vaccination can be thought to be induced by traumas and persistent inflammation associated with the injection. Pirouzmanesh et al. report that inflammation was seen in 40.8% of their pilomatricoma cases in microscopic examinations. A trauma that can damage the hair follicles during vaccination can also lead to missuppression in apoptosis and to pilomatricoma formation. In our case, we, too, believe that the post-vaccination inflammation has triggered the formation of the tumor. Hematoma subsequent to an injection can also accelerate the growth of pilomatricoma tumors. The early redness and swelling at the injection site suggests the presence of an unnoticed hematoma following vaccination.

To summarize, pilomatricoma can be confused with other types of lesions, given its rarity and unknown characteristics. Therefore, it should be considered as a possibility in cases presenting with swelling after vaccination. In this regard, soft tissue tumors, in particular, should be eliminated by differential diagnosis.

Informed Consent: Written informed consent was obtained from patient’s parents who participated in this study.

Peer-review: Externally peer-reviewed.
Author contributions: Concept - Z.T., B.K.; Design - Z.T.; Supervision - S.G.; Resource - Z.T.; Data Collection and/or Processing - Z.T., B.K.; Analysis and/or Interpretation - B.K.; Literature Search - Z.T., B.K.; Writing Manuscript - Z.T.; Critical Reviews - B.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

REFERENCES