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Punch Biopsy Results in Misdiagnosis of Pilomatrixoma

Punch Biyopsi Pilomatriksomada Yanlış Tanıya Neden Olmaktadır

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Abstract

Punch biopsy results in misdiagnosis of clinically unsuspected giant pilomatrixoma located over the parotid gland. This study presents a case of pilomatrixoma that was misdiagnosed as a malignant epithelial tumor using punch biopsy. A 25-year-old male was admitted to our clinic for the evaluation of a mass measuring 7×8×8 cm located over the parotid gland. The patient had previously undergone punch biopsy at another clinic because of a lesion in the parotid gland. Punch biopsy revealed a malignant epithelial tumor. The patient underwent excisional biopsy at our clinic. After the biopsy, the residual skin defect was treated using full-thickness skin grafts. The facial nerve and parotid gland were preserved during the biopsy. Histopathological examination of the excisional biopsy material revealed pilomatrixoma. Punch biopsy may result in misdiagnosis of skin lesions in the parotid gland. A differential diagnosis for benign tumors such as pilomatrixoma is essential prior to an aggressive surgical intervention of the parotid gland.

Keywords: Pilomatrixoma, parotid tumors, malignant transformation, punch biopsy

Öz

Klinik olarak parotis üzerindeki bölgede dev pilomatriksomadan şüphelenilmediği için punch biyopsi ile yanlış tanılar konulabilmektedir. Punch biyopsi sonucu malign epitelyal tümör tanısı ile gönderilen olgunun değerlendirilmesi amaçlanmıştır. Parotis bölgesi üzerinde yaklaşık 7x8x8 cm boyutlarında kitle lezyonu ile müracaat eden 25 yaşındaki erkek hasta kliniğimizce değerlendirildi. Başlangıçta parotis bölgesi lezyonları düşünülen ve diğer klinikçe yapılan punch biyopsi sonucu malign epitelyal tümör olarak rapor edilmiş olan hastaya kliniğimizce eksizyonel biyopsi ameliyatı yapıldı. Ameliyat sonrası oluşan cilt defekti tam kalınlıkta cilt grefti ile onarıldı. Ameliyatta fasiyal sinir ve parotis bezi korundu. Eksizyonel biyopsi sonucu pilomatriksoma olarak rapor edildi. Parotis bölgesi cilt lezyonlarında, punch biyopsi yanıltıcı sonuçlara neden olabilmektedir. Parotis bölgesine yapılabilecek agresif cerrahi işlemlerden önce dev pilomatriksoma gibi iyi huylu tümörlerin ayırıcı tanısının yapılması gereklidir.

Anahtar Sözcükler: Pilomatriksoma, parotid tümörleri, malign transformasyon, punch biyopsi

INTRODUCTION

Pilomatrixoma is a benign lesion that originates from hair follicles and are usually located in the head and neck region.[1] It has bimodal peak presentation during childhood and in individuals aged >60 years.[2] In >50% of cases, pilomatrixoma occurs in the head and neck region (neck and frontal, temporal, periorbital, and periauricular regions).[3] Clinically, this lesion is a firm, solitary, painless, and slow-growing nodule. The lesions are generally <1 cm in diameter, and those that measure >5 cm in diameter are quite rare.[4] Despite their benign character, an inflammation at an older age must be considered as malignancy.[5]

We report a case that can possibly be confused with parotid malignancies, and because of its close location to the parotid gland was diagnosed as malignant epithelial tumor by punch biopsy performed in an external dermatology clinic before the patient applied to our clinic with a massive lesion.

CASE PRESENTATION

A 25-year-old male was admitted to our Plastic surgery clinic with an ulcerated mass over his left cheek. He had no history or any sign of a trauma or an infectious disease. The mass had rapidly grown within a five-month period. Punch biopsy was performed for the

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mass at an external dermatology clinic, and the pathological examination results reported the mass to be a malignant epithelial tumor (Figure 1). Physical examination revealed a painless, ulcerated mass over the periauricular portion of the parotid gland. The mass invaded the skin and the subcutaneous tissues, and measured 7×8×8 cm in size (Figures 2a-c). The mass was firm but unfixed to the deeper tissues on palpation and was bleeding because of surface ulceration. Neurological examination revealed intact facial nerve with all branches. Sonographic appearance of the mass showed a sharply differentiated border with deep structures and calcifications inside the tumor. There was no pathological lymphadenopathy of the neck lymphatic nodes. Magnetic resonance imaging (MRI) suggested a lobulated contoured lesion with exophytic extensions from the fatty tissue, an intermediary plan between the muscle and parotid gland plans, and striction of skin and subdermal tissues at the anterior aspect of the parotid gland (Figure 3). The other systemic examination results were normal. After obtaining his permission the patient was operated under general anesthesia. The facial nerve was preserved during the resection of the mass. A full-thickness skin graft harvested from the right supraclavicular area was used for reconstructing the defect area after resection, followed by tie-over dressing (Figures 2d, e). The histopathological examination revealed calcified and keratinized zones with basophilic and shadow cells (Figure 1). Thus, the diagnosis was confirmed to be pilomatrixoma. The patient had previously undergone punch biopsy, and the diagnosis was malignant epithelial tumor. The specimen was immunohistochemically stained with p53 and Ki67 to exclude any malignancy. Because of the positive staining with p53 and Ki67, the patient was closely followed up for a recurrence risk. The facial nerve examination of the patient was normal in the post-operative period. The patient was discharged from the clinic on post-operative day one. The graft dressing was removed on post-operative day five. No complication was encountered during the follow-up period.

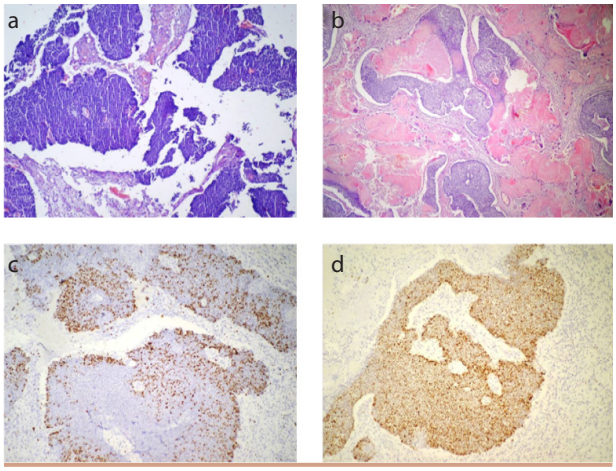

Figure 1. a-d. Histopathological findings, (a) the cell groups are observed as small basaloid cells, with atypical mitosis (H&E stain; original magnification, ×100); (b) two different cell groups are observed in histopathological sections. The cell is les of basaloid and shadow cells (H&E stain; original magnification, ×40); (c) a high Ki67 proliferation index is observed at the periphery of the basaloid cells (Ki67 stain; original magnification, ×100); (d) positive p53 is observed in basaloid cell groups (p53 stain; original magnification, ×100)

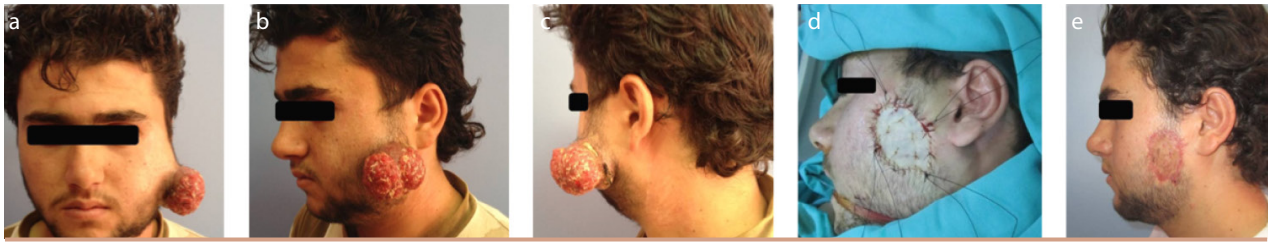

Figure 2. a-e. A 25-year-old male with a tumor over the left parotid gland, measuring 7×8×8 in size, was diagnosed as pilomatrixoma. (a) Pre-operative anterior image, (b) Pre-operative lateral image, (c) Intra-operative grafting image, (d) Image in the first month post-operative

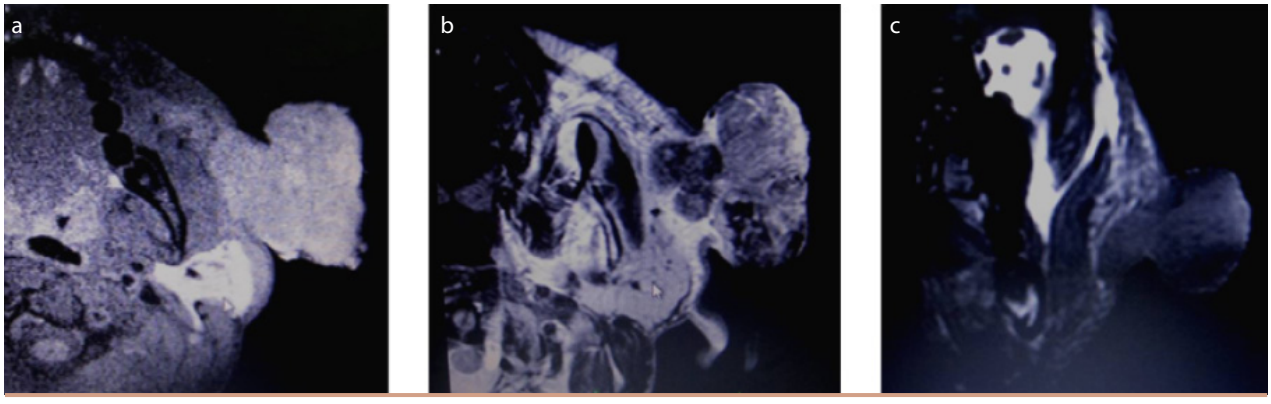

Figure 3. a-c. Pre-operative MR images. (a, b) axial sections, (c) coronal sections

DISCUSSION

Because of the varying clinical presentation of pilomatrixoma, it is usually misdiagnosed. Pilomatrixomas are superficial, firm, slow-growing, painless, and sometimes ulcerated dermal nodules.[6] There may be pain during the inflammation and ulceration periods. Most of the lesions are irregular, measure 0.5–3 cm in diameter, and have a lobular appearance because of calcified content. The tumor most commonly occurs in the head and neck region (40%–70%), followed by upper extremities, trunk, and lower extremities.[7] In the head and neck region, the most involved area is the cheek with 18%, followed by periorbital and scalp regions.[8]

In the literature, the reported number of cases of giant pilomatrixoma (>5 cm in diameter) is 30, of which <10 cases have parotid gland involvement.[4,5,9] Because of the rare presentation of pilomatrixoma in the parotid gland, its consideration during differential diagnosis decreases. Thus, it is misdiagnosed during laboratory examinations; in particular, the diagnostic errors increase in histopathological evaluations by fine-needle aspirations.[10] During clinical examination the most important thing for the diagnosis of pilomatrixoma is to suspect.

The patient was admitted to our clinic following a diagnosis of a malignant epithelial tumor by punch biopsy performed at another dermatology clinic. The biopsy material may have been obtained from the zone where atypical mitosis was dominantly observed and where only basaloid cells existed, thus resulting in misdiagnosis (Figure 1a). The exact diagnosis was obtained after the total resection of the tumor. Benign [pleomorphic adenoma, papillary cystadenoma lymphomatosum (Warthin's tumor), monomorphic adenoma, oncocytoma, and hemangioma] and malignant (mucoepidermoid carcinoma, malignant mixed tumors, actinic cell carcinoma, and squamous cell carcinoma) tumors derived from the parotid gland must be considered during differential diagnosis. Imaging techniques must be used for differential diagnosis of parotid tumors.[11] Sonography is a basic technique that can demonstrate the associations and extensions of the lesion with deeper structures and calcifications inside the tumor.[8] We used both sonography and MRI in this study. Because of the data obtained from the patient, parotidectomy and lymph node dissection were not performed.

Histological examinations revealed that the tumor had a fibrous capsule derived from the subdermal layer and that originated from the outer layer of the hair follicle. Diagnosis depends on the existence of increased mitotic activity and the so-called shadow and ghost cells at the basaloid germinal center of the nodes (Figure 1b).[12] These cells are considered as remnants of underived ectodermal keratinocytes by the time they tend to calcify. The infiltrative features (perineural and perivascular invasion), solitary cell necrosis, and distinguishable mitotic features may be indicators of malignant transformation.[13] The misdiagnosis of the pleomorphic adenoma that contains squamous metaplasia, carcinoma, and basal cell carcinoma may depend on the fine-needle aspiration biopsy.[7,14] Our case was also misdiagnosed as malignant epithelial tumor by punch biopsy. The diagnosis was ensured by the total excision of the tumor (Figure 1b). For the evaluation of the malignant transformation and recurrence, immunohistochemical staining with p53 and Ki67 was performed (Figure 1c, d). The patient was closely followed up because of the positive staining of both p53 and Ki67.

The most important issue for the treatment of pilomatrixoma is excision with clear surgical borders.[2,7,8] Because the tumor has benign features, the surgical borders must be planned according to the location of the lesion, the association with the esthetic units, and the closure techniques, and the surgical boundaries must be planned at least 1–2 cm.[8] However, to confirm the pathological diagnosis and surgical boundaries, esthetic closure may be postponed in order to select either a basic or a complex technique. We chose the most basic technique for the closure of the defect, grafting. We preserved all branches of the facial nerve. Some authors state that superficial parotidectomy should be included in the treatment.[4] However, we suggest that the diagnosis should be confirmed by conservative solutions and also recommended that parotidectomy must be avoided because of the lack of malignancy evidence.

CONCLUSION

Pilomatrixoma should be considered for the differential diagnosis of tumors in the parotid gland. Pathologically, punch and aspiration biopsies may result in misdiagnosis, therefore, because pilomatrixoma is a benign tumor, radical decisions must be postponed until a definite diagnosis.

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