Fibroepithelioma of Pinkus: variant of basal cell carcinoma or trichoblastoma? Case report

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SUMMARY: Fibroepithelioma of Pinkus: variant of basal cell carcinoma or trichoblastoma? Case report.

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Introduction. Fibroepithelioma of Pinkus (FeP) is a rare tumor that most often affects women aged between 40 and 60 years. Clinically FeP presents as a soft, usually solitary, polypoid or papillomatous well circumscribed tumor of skin color. It is typically located to the trunk and extremities.

Case report. A 75 year old male presented to our Department complaining for the presence of a lesion of the dorsal region. His medical history was free except for several basal cell carcinoma (BCC) surgically excised. Clinical examination revealed a pigmented lesion in the back. The lesion was surgically excised and histopathology showed of a fibroepithelioma of Pinkus.

Discussion. Currently, FeP is considered a rare variant of basal cell carcinoma, with characteristic histopathological features, although this view is somewhat controversial as some authors considered FeP to be a variant of trichoblastoma. The pathogenesis of FeP is still under investigation. It is thought that a mutation in the tumor suppressor gene TP53 might predispose to the development of FeP.

Conclusions. Our case is interesting for two reasons. First, in our case FeP is pigmented. Moreover we present a case of Fep in a patient with a history of BCC, a finding that supports the classification of fibroepithelioma of Pinkus as a variant of basal cell carcinoma.

RIASSUNTO: Fibroepitelioma di Pinkus: variante di carcinoma basocellulare o tricoblastoma? Caso clinico.

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Introduzione. Il fibroepitelioma di Pinkus (FeP) è un tumore raro che colpisce prevalentemente le donne con un picco di incidenza tra la quarta e la quinta decade di vita. Clinicamente si presenta come una lesione, generalmente solitaria, di consistenza molle, di forma polipoide o papillomatosa, ben circoscritta, del colore della cute circostante. Si localizza prevalentemente al tronco e alle estremità.

Caso clinico. Uomo di anni 75. Si presenta alla nostra osservazione per una neoformazione dorsale. La sua storia clinica non era rilevante, ad eccezione di numerosi carcinomi basocellulari già escissi. All'esame obiettivo la lesione dorsale appare pigmentata. La neoformazione viene escissa chirurgicamente e l'esame istologico pone la diagnosi di fibroepitelioma di Pinkus.

Discussione. Attualmente le teorie eziopatogenetiche più accreditate per il FeP sono due. Secondo alcuni autori, il tumore può essere considerato una variante benigna del carcinoma basocellulare (BCC); secondo altri, il fibroepitelioma di Pinkus è un analogo del tricoblastoma. La patogenesi del FeP è anch'essa poco chiara; è stato proposto che la mutazione del gene TP53 potrebbe predisporre allo sviluppo del tumore.

Conclusioni. La nostra osservazione è interessante per due motivi. Innanzitutto per la presentazione clinica peculiare della lesione: in letteratura è presente un solo caso di FeP pigmentato. Inoltre, nel nostro caso il FeP si è sviluppato in un paziente con una lunga storia di BCC e ciò potrebbe supportare l'ipotesi eziopatogenetica attualmente più accreditata, ovvero quella che considera il FeP come variante benigna del BCC.

KEY WORDS: Fibroepithelioma of Pinkus - Basal cell carcinoma - Trichoblastoma. Fibroepitelioma di Pinkus - Carcinoma basocellulare - Tricoblastoma.

Introduction

Fibroepithelioma of Pinkus (FeP) was first described as premalignant fibroepithelial tumor of the skin by Herman Pinkus in 1953 (1). It is an uncommon tumor that has a female preponderance and a peak age between the fourth and the sixth decade (2). Classification still remains controversial and is considered by some authors as a variant of BCC (3), while others categorize it as fenestrat-

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ed trichoblastoma, i.e. a benign counterpart of BCC (2).

Clinically, FeP present as soft polypoid or papillomatous tumors with a strong predilection for trunk and extremities, in contrast to conventional basal cell carcinomas, which are almost always found in sun-exposed sites (2). However, to our knowledge, the tumors are generally indolent and aggressive behavior or metastasis has not been reported (4). Among a number of differential diagnoses, dermal melanocytic nevus, pedunculated fibroma, acrochordon (skin tag), and seborrheic keratosis are most commonly listed.

Histologically it shows narrow bands of basaloid or squamous keratinocytes that terminated in small nubs of peripherally palisaded basaloid cells, surrounded by more abundant stroma, which always form an even border with the underlying dermis. This stroma is rich in fibrocytes that differentiated toward follicular papillae (Fig. 1).

Discussion

Fibroepithelioma of Pinkus (FeP) was first described as "premalignant fibroepithelial tumor of the skin" by Herman Pinkus in 1953. Although FeP is currently accepted as a variant of basal cell carcinoma (BCC), its classification still remains controversial. FeP may also be categorized as a variant of trichoblastoma, a benign counterpart of BCC (5).

Bowen and LeBoit examined 75 cases of fibroepithelioma of Pinkus, and proposed that these lesions represent a fenestrated variant of trichoblastoma. Important histological features of fibroepithelioma of Pinkus supporting this view included the nature of the interface between tumor and dermis and the composition of the adjacent stroma. Cases of fibroepithelioma of Pinkus demonstrated a "blunt interface with the underlying dermis" and did not infiltrate the dermis or subcutaneous tissue as is seen in nodular basal cell carcinoma. The stro-



Fig. 1 - Fibroepithelioma of Pinkus: immunohistological findings.

Case report

A 75-year old Caucasian male was referred to the our Department with a lesion in dorsal region. He had no systemic complaints, no recent weight loss, but a personal history of several BCC.

Physical examination was unremarkable except for a 3x3 cm solitary, soft, pigmented, painless mass in the dorsal region dated about one year ago (Fig. 2).

A wide surgical excision was performed. The final pathologic diagnosis was fibroepithelioma of Pinkus (Fig. 1).

The wound healed with an acceptable postoperative result and clinical follow-up at 8-months showed non other complication or recurrence.



Fig. 2 - Fibroepithelioma of Pinkus of the dorsal region.

ma surrounding fibroepithelioma of Pinkus is rich in fibroblasts, a pattern seen in many trichoblastomas, thought to be indicative of benignity. These fibroblastrich areas demonstrated papillae-like differentiation. Immunohistochemical data was also used to bolster their argument.

Compared with basal cell carcinoma, fibroepithelioma of Pinkus shows diminished p53 and MIB-1 expression. Immunohistochemical stains for cytokeratin 20 also demonstrated retention of Merkel cells, in general a feature of benign trichoblastic tumors and not of basal cell carcinoma (4). Previous studies of fibroepithelioma of Pinkus have also demonstrated the presence of Merkel cells and their absence or rarity in basal cell carcinoma. Lastly, fibroepithelioma of Pinkus frequently occurs on the trunk in contrast to basal cell carcinoma, a lesion most commonly seen in sun-exposed areas of the head and neck, often accompanied by solar elastosis. Indeed, only 15% of basal cell carcinoma occurs on the trunk, versus 81% of fibroepithelioma of Pinkus cases. Others maintain that fibroepithelioma of Pinkus is in fact a subtype of basal cell carcinoma, including Pinkus it self who regarded the lesion as a precursor to basal cell carcinoma (4).

In their recent examination of the histopathologic characteristics of fibroepithelioma of Pinkus, Ackerman and Gottlieb list multiple criteria for regarding it as a basal cell carcinoma variant. Some of these criteria include an asymmetric neoplasm manifesting a fenestrated growth pattern, composition of trichoblasts, foci of clefts between the tumor strands and fibrotic stroma, and the occurrence of fibroepithelioma of Pinkus in continuity with a nodular type basal cell carcinoma. Additionally, there is only limited differentiation toward follicular germs or primitive mesenchyme and the germ-like structures are not associated with rudimentary papillae. Other morphologic features for classifying fibroepithelioma of Pinkus as a variant of basal cell carcinoma include its continuity with pre-existing infundibula and a tendency to extend into the deep dermis or even subcutaneous fat in certain cases. It has been proposed that fibroepithelioma of Pinkus develops from the spread of malignant basal cells along eccrine ducts that serve as a template. The distortion and branching of sweat gland ducts during inflammation and/or in the presence of a neoplastic milieu can result in the characteristic narrow branching and anastomosing strands seen in fibroepithelioma of Pinkus as tumor cells partially replace the ducts (4).

Recently studies of basal cell carcinoma have demonstrated positive expression of androgen receptor by immunohistochemistry. The positive immunostaining ranged from 60% to 78% of basal cell carcinoma cases. In many cases of basal cell carcinoma, androgen receptor expression was focal, involving less than 5% of the tumor mass, but nonetheless felt to represent true positivity (4). Additionally, studies examining benign follicular tumors, such as trichoepithelioma or trichoblastoma, have not demonstrated androgen receptor expression in these tumors. Katona and coll. examined fi-

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broepithelioma of Pinkus for androgen receptor expression to see if this marker might aid in the classification as either a basal cell carcinoma or trichoblastoma. These cases of fibroepithelioma of Pinkus demonstrated androgen receptor expression analogous to basal cell carcinoma. Seventy-seven percent (10/13) of fibroepithelionas of Pinkus showed immunoreactivity for androgen receptor. This was comparable to that seen in the control group of basal cell carcinoma, and significantly higher than that seen in the control groups of trichoblastoma and trichoepithelioma. These results for and rogen receptor positivity in fibroepitheliomas are also quite similar to what has been reported in the literature for androgen receptor expression in basal cell carcinoma and so these are supportive of the hypothesis that fibroepithelioma of Pinkus is a variant of basal cell carcinoma (4).

The pathogenesis of FeP is still under investigation. It is thought that a mutation in the tumor suppressor gene *TP53* might predispose to the development of FeP. Similarly to BCC, the suggestion has been made that mutations in the *PATCHED* gene, which provides inhibitory signal in the Hedgehog pathway, could also lead to the development of FeP. Further studies are needed to further elucidate the genetic predisposition of FeP (5).

Conclusion

Our case is interesting for two reasons. First in our patient FeP is pigmented and, to our knowledge, it has been previously reported in the litterature only by Strauss and coll (6). Moreover our patient had a history of BCC, a finding that supports the classification of fibroepithelioma of Pinkus as a variant of basal cell carcinoma.

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