Pigmented Follicular Cyst in an Intradermal Nevus

İntradermal Nevüs İçerisinde Pigmente Folikül Kisti

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Özet

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Abstract
Pigmented follicular cyst (PFC) is defined as a cyst localized in the dermis, opening to the epidermis by a pore-like structure, and is lined by stratified squamous epithelium. The cyst contains pigmented terminal hair shafts in its lumen. In this report we present an unique case of pigmented hair follicle cyst in an intradermal nevus (IDN). To our knowledge, this is the second case of the medical literature. The mass effect of IDN seems to cause follicular occlusion, and it is suggested that mechanical effect takes place in pathogenesis.

Keywords
Pigmented Follicular Cyst; Pigmented Skin Lesion; Dermal Cyst
Introduction
PFC is a benign, asymptomatic, and mostly solitary dermal cyst and contains pigmented terminal hair shafts in its lumen. The co-existence of adnexial or epithelial cysts and melanocytic nevus are seen often in the literature, but the co-existence of IDN and PFC has been published only once in the literature [1]. We believe that the true incidence will correlate with the awareness of the pathologists.

Case Report
A 34-year-old woman was presented with swelling on the lateral side of her right eye, which was present since childhood but has increased in size in recent weeks. Dermatological examination showed a confined, brown colored papule, 0,7cm in diameter, localized in the lateral canthus of the right eye. The lesion was excised under local anesthesia with a clinical diagnosis of IDN. Histopathological examination of the H&E stained sections showed a cyst with a narrow pore-like opening to the superficial epidermis in an ordinary IDN with maturation in the deep levels (figure 1). The cyst was lined by stratified squamous epithelium and showed epidermal keratinization (figure 2). No atypia was observed in the stratified squamous epithelium. Acute inflammatory cells were seen in the lumen of the cyst and brown pigment was shown in transverse and oblique sections of the hair shaft (figure 3). After melanin discoloration process, the pigment has disappeared and so it was shown to be melanin. After surgical excision of the lesion, the patient did not receive further treatment. Neither complication nor recurrence occurred during 2 years of follow-up period.

Discussion
PFC was firstly defined by Mehregan and Medenica in 1982 [2]. Since than, 17 cases reported. It is generally seen in head and neck (10 of 17 cases) of men (13 men, 4 women). The age of presentation for evaluation ranges between 20 and 63 years (mean 41 years). To date, no pediatric cases have been reported. Clinically, PFC typically presents as a single pigmented dome-shaped or intracutaneous papulonodule with multiple lesions only described in four patients [3,4] The diameter of the lesion ranges between 0,3 and 3cm. The most frequent clinical diagnosis is MN. Microscopically the cyst is lined by stratified squamous epithelium and shows epidermal keratinization. Sometimes the cyst contains a narrow pore-like opening to the surface epidermis. Characteristically, multiple pigmented hair shafts and laminated keratin are present within the cyst cavity [3,6] The pigmented hair shafts, a characteristic pathologic finding for pigmented follicular cyst, may sometimes degenerate into a pigmented substance, as described by Iwahara et al [7]. The marked weathering of the hair shafts seen in this case was thought to be the result of frequent inflammation and secondary infection of the cyst. The coexistence of adnexial and epithelial cysts and melanocytic nevus are seen often in the literature. Cohen and Rapini described the clinicopathologic manifestations of 93 cases and reviewed the features of the 69 previously reported nevi with cysts in the world literature. Variants of nevus with cyst include lesions in which the nevus is a congenital, “dysplastic,” blue, or Spitz nevus. The cysts may be sebaceous cysts, hidrocystomas, dermoid, or trichilemmal cysts [8]. Although pathogenesis of PFC is not exactly known, it is suggested that occlusion of the follicular infundibulum plays an initiator role in these lesions [8]. The presentation of epidermoid cysts frequently in IDN, less frequently in compound nevus and rarely in junctional or blue nevus supports this obstruction hypothesis. Because the obstruction phenomenon occurs most often when there is an increase in the number and activity of nevus cells in the upper dermis, as is the case with intrader-
mal and compound nevus. Junctional nevus with cells irregularly arranged at the dermoepidermal junction, and blue nevus with cells in deeper dermis are less likely to occlude a follicular orifice. The co-existence of IDN and PFC has been published only once and to our knowledge; this is the second case in of the medical literature [1]. Nevoid component of these two cases do not have malignant findings, such as cytologic atypia, mitotic activity or invasion, which are characteristic for nevoid melanomas [9]. In conclusion, PFC is a benign, asymptomatic, and mostly solitary lesion and no treatment modality except excisional biopsy has been reported. When multiple and symptomatic, pigmented follicular cysts can represent a therapeutic challenge. Surgical excision of multiple cystic lesions is time-consuming and carries the risks of increased intraoperative hemorrhage, greater loss of normal tissue, and scarring. In contrast, CO2 laser therapy provides a bloodless surgical field, more precise lesion ablation, and minimal scar formation [5].

Competing interests
The authors declare that they have no competing interests.

References

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