Case Report / *Olgu Sunumu*

J Breast Health 2013; 9: 218-20
DOI: 10.5152/tjbh.2013.37

Inflammatory Pseudotumor of the Breast: A Rare Case

Memenin İnflamatuvar Psödotümörü: Nadir Bir Olgu

Ebubekir Gündeş[1], Adil Kartal[1], Faruk Aksoy[1], Hatice Toy[2]

[1]Necmettin Erbakan Üniversitesi Meram Tıp Fakültesi, Genel Cerrahi Anabilim Dalı, Konya, Türkiye
[2]Necmettin Erbakan Üniversitesi Meram Tıp Fakültesi, Patoloji Anabilim Dalı, Konya, Türkiye

ABSTRACT

Inflammatory pseudotumor of the breast (IPB) is a very rare condition. Literature reports a limited number of cases. Its pathogenesis is also a controversial issue. Within the scope of our study, a 65-year-old female patient, who had resection because of inflammatory pseudotumor of the breast, was evaluated. The patient presented to our clinic with complaints of a painless mass in her right breast. Imaging guided tru-cut biopsy was performed. The pathological findings following the tru-cut biopsy were consistent with inflammatory pseudotumor. Local excision was performed. There was no recurrence during the 24-month follow-ups.

Key words: Breast, inflammatory, pseudotumor

ÖZET

Memenin inflamatuar psödotümörü (MIP) çok nadir bir durumdur. Literatürde sınırlı sayıda olgu bildirilmiştir. Patogenezi tartışmalı bir konudur. Memenin inflamatuvar psödotümörü ön tanısıyla rezeksiyon yapılan 65 yaşında bir kadın hasta irdelendi. Sağ memesinde ağrısız kitle yakınması ile kliniğimize başvurdu. Görüntüleme eşliğinde tru-cut biyopsi yapıldı. Tru-cut biyopsi sonrası patolojik bulgular inflamatuvar psödotümörle uyumluydu. Lokal eksizyon işelemi uygulandı. Yirmi dört aylık takiplerinde nüks saptanmadı.

Anahtar sözcükler: Meme, inflamatuar, psödotümör

Introduction

Inflammatory pseudotumor of the breast (IPB) was first reported by Petlinato in 1988 (1). There are very few cases reported in the literature and these studies are case reports. The pathogenesis of this tumor is controversial (2, 3). IPB may be detected at every age. Clinically, it manifests itself as a palpable, mobile, and a slightly sensitive mass (3). Its imaging characteristics are non-specific and a definitive diagnosis can be confirmed histopathologically (4). Its treatment is local excision. There is a low possibility of recurrence. Local re-excision procedure can be performed on these patients.

Case Report

A 65-year-old female patient presented to our clinic with complaints of a mass in her right breast. The examination revealed a palpable, mobile, and a slightly sensitive mass of about 5x5 cm. Both axillae and the left breast were normal. Her history had no unusual characteristics.

The mammography revealed a high-density nodular opacity with matching borders with the glandular tissue in the posterolateral area contoured to the lobule that was smooth in shape and located in the upper outer quadrant of the right breast (Figure 1). Ultrasonography of the right breast showed a cyst of about 1.5 cm that was about 12 cm away from the areola at the 11 o'clock position and a lesion of 2x 2.5 cm with cystic areas having heterogeneous-smooth contours which could not be clearly differentiated as solid or cystic in the lateral vicinity. This was interpreted as BRADS III.

Local excision procedure was performed on the patient as the result of the imaging guided tru-cut biopsy which indicated inflammatory pseudotumor of the breast (IPB). Pathological analysis results showed that the homogenous lesion was 4.5x3, 5x2.5 cm in size. There was a lesion eliminating the normal structure of the fibrolipomatous breast stroma localized just beneath the skin as seen in the sections. It was observed that the lesion was composed of mononuclear cell infiltration that also included intensive plasmocytes forming a germinal center in some parts and fibrotic fields in others (Figure 2). Immunohistochemical staining results were: LCA (+) (Figure 3), synaptophysin (-), LMW-cytokeratin (-), CD 15 (-), CD 30 (-). CD 20 was (+) (Figure 4) and it was observed that it was intensively stained with KI-67 at germinal centers. Pathological diagnosis was inflammatory pseudotumor of the breast. No recurrences were detected during the 24-month post-op follow-ups.

This case was presented as a poster at the 11th National Breast Diseases Congress, 5-9 October 2011, Antalya, Turkey.
Bu olgu, 11. Ulusal Meme Hastalıkları Kongresi'nde poster olarak sunulmuştur, 5-9 Ekim 2011, Antalya, Türkiye.

Address for Correspondence/Yazışma Adresi:
Ebubekir Gündeş, Necmettin Erbakan Üniversitesi Meram Tıp Fakültesi, Genel Cerrahi Anabilim Dalı, Konya, Türkiye
Phone / Tel.: +90 332 223 61 21 e-mail / e-posta: ebubekir82@hotmail.com

Received / Geliş Tarihi: 12.07.2012
Accepted / Kabul Tarihi: 11.08.2012

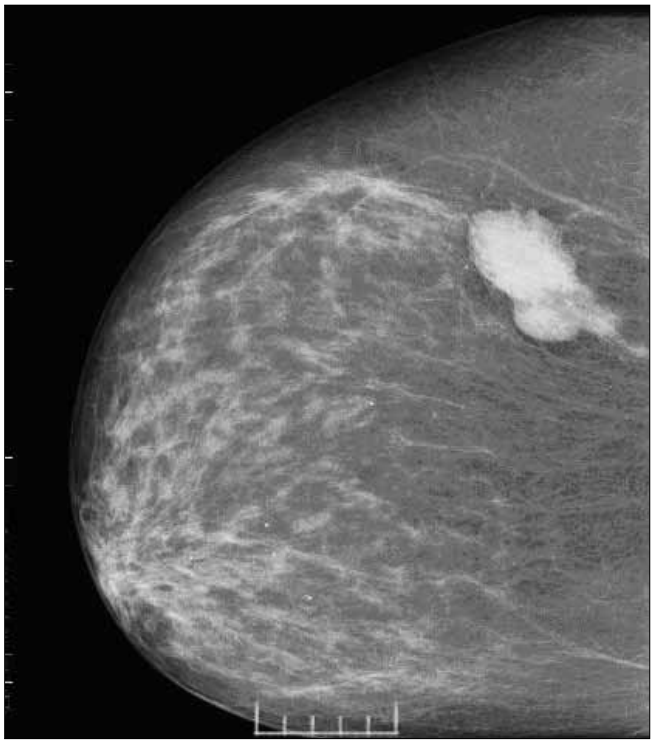

Figure 1. Nodular opacity with smooth, lobular contours in the upper outer quadrant as shown by mammography

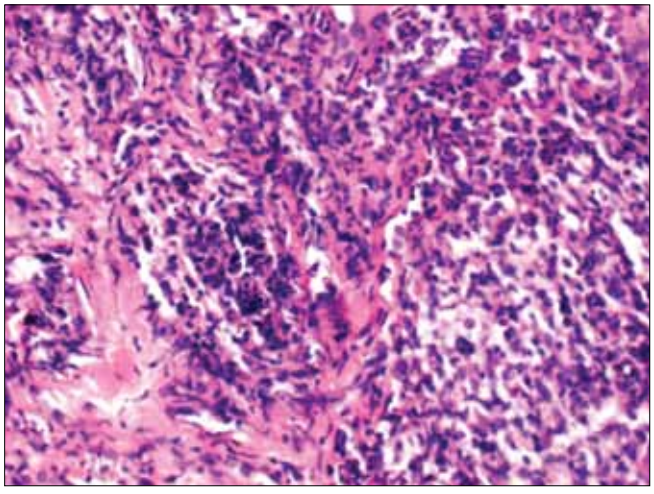

Figure 2. Mononuclear cell infiltration also including intensive plasmocytes that form a germinal center by intensification and fibrotic fields in parts (Hematoxylin-Eosin Staining X 200)

Discussion and Conclusions

The inflammatory myofibroblastic tumor of the breast is a rare, (1-8) low grade, neoplastic lesion (1, 3, 4). It was first reported by Pettinato in 1988 (9). It should not be confused with malignities with high grades clinically, radiologically, and pathologically (1, 6). This significant differentiation might be difficult (1). A couple of cases were reported in the literature and these are in the form of case reports (1, 3, 6). Caggin et al. (10) reported 84 extra pulmonary inflammatory pseudotumors (IP) and among these 48 were female cases, with only one being a breast IP.

The pathogenesis of this tumor is controversial and, although the etiology of this entity is still yet to be known, some researchers argue

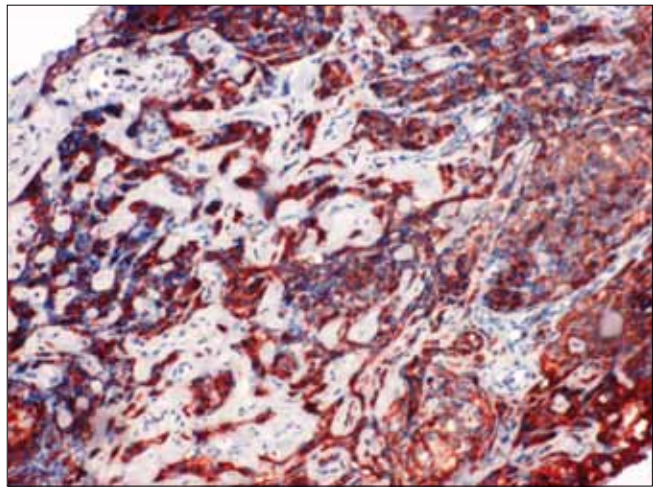

Figure 3. LCA positive tumor cells (x100)

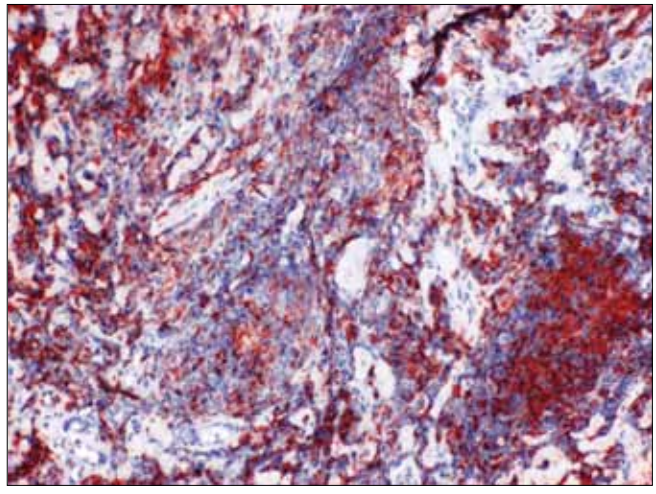

Figure 4. CD20 positive tumor cells (x100)

that an inflammatory response to aberrant reactive or local cytokines underlie its etiology (4-6). Cases with vein invasion, local recurrence, and even metastases were reported (5, 8). Recent studies have found cytogenic clonal anomalies (3, 5, 6) and anaplastic lymphoma kinase expression, which all indicate a neoplastic cause (4). Again, recent studies have stated that an inflammatory pseudotumor might be a disease related to IgG4 (6).

In most of the case reports found in the literature the masses have been reported to be clinically palpable, mobile, and sensitive and most of them were conjoining the skin above them (3). Although IPB's are more common in children and young adults, it might be seen at every age (3, 4). Since its first report in 1988, there have been 23 cases in literature to the best of our knowledge. The average age of these patients was 47 (13-79). Pseudotumor localization area was the right breast in 13 patients, while it was the left in 10. The mean mass size was found to be 2.8 (1-4.5) cm. Our case was a 65-year-old female patient who presented to our clinic with complaints of palpable mass. Pseudotumor localization of our patient was the right breast and the mass size was 4.5 cm.

It is important to have pre-op ultrasonography guided biopsy to prevent unnecessary mastectomies. Imaging characteristics are non-specific (2, 4).

Definitive diagnosis is achieved histopathologically (1-6). The radiological evaluation of our patient revealed a heterogeneous lesion with smooth contours which also contained cystic areas. Tru-cut biopsy was used in the pre-operative diagnosis.

Inflammatory pseudotumor of the breast is composed of mature plasma cells, lymphocytes, histiocytes, and fusiform cells (1, 6, 7). Three major types have been defined: myxoid/vascular, fusiform celled, and hypocellular fibrous type (7). Early lesions are rich in inflammatory cells and mature lesions have more collagen (3, 7). Ultrastructural and immunohistochemical studies demonstrated that the fusiform cells were myofibroblasts (2, 6). In the fusiform celled type of IPB, generally SMA, desmin, and sometimes cytokeratine are positive (7).

The histopathological differential diagnosis has a very wide spectrum. This might be in the form of benign and malignant lesions (6, 7). The differential diagnosis of IPB tumors also covers benign lesions like nodular fasciitis, fat necrosis, and fusiform celled metaplastic carcinoma of the breast, fibromyxoid sarcoma, malignant fibrous histiocytoma,z inflammatory fibrosarcoma, extramedullary plasmacytoma and fibromatosis (1, 3, 6).

Reported recurrences should not dissuade us from surgical excision ofsz tumors (3). Nevertheless, close follow-ups are needed following surgery because of the possibility of recurrence (1, 3-5). All the cases reported in the literature had local excision procedures and only two cases had recurrences. These two cases underwent re-excision procedures. The average disease-free follow-up period of 18 patients reported in literature was found to be 19 (4-85) months. Our patient also had the local excision procedure and no recurrences were observed during the 24-month follow-up period.

Consequently, this situation is very rare and has non-specific imaging findings. Pre-operative diagnosis can be achieved by ultrasonography guided tru-cut biopsies. Pre-operative diagnosis prevents unnecessary mastectomies. Its treatment is wide local excision. Close follow-ups are needed regarding recurrence.

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

Peer-review: Externally peer-reviewed.

Informed Consent: Written informed consent was obtained from patients who participated in this case study.

Author Contributions
Concept - E.G.; Design - E.G.; Supervision - F.A.; Funding - E.G., A.K.; Materials - E.G.; Data Collection and/or Processing - E.G., D.E.; Analysis and/or Interpretation - E.G.; Literature Review - F.A.; Writer - E.G.; Critical Review - F.A., A.K.; Other - E.G.

Çıkar Çatışması
Yazarlar herhangi bir çıkar çatışması bildirmemişlerdir.

Hakem değerlendirmesi: Dış bağımsız.

Hasta Onamı: Yazılı hasta onamı bu olguya katılan hastalardan alınmıştır.

Yazar Katkıları
Fikir - E.G.; Tasarım - E.G.; Denetleme - F.A.; Kaynaklar - E.G., A.K.; Malzemeler - E.G.; Veri toplanması ve/veya işlemesi - E.G., D.E.; Analiz ve/veya yorum - E.G.; Literatür taraması - F.A.; Yazıyı yazan - E.G.; Eleştirel İnceleme - F.A., A.K.; Diğer - E.G.

References

1. Park SB, Kim HH, Shin HJ, Gong G. Inflammatory pseudotumor (myoblastic tumor) of the breast: a case report and review of the literature. J Clin Ultrasound 2010; 38: 52-5. (PMID: 19802887)
2. Kim SJ, Moon WK, Kim JH, Cho N, Chang CM. Inflammatory pseudotumor of the breast: a case report with imaging Findings. Korean J Radiol 2009; 10: 515-8. (PMID: 19721838) [CrossRef]
3. Akbulut M, Gunhan-Bilgen I, Zekioglu O, Duygulu G, Oktay A, Ozdemir N. Fine needle aspiration cytology of inflammatory myofibroblastic tumour (inflammatory pseudotumour) of the breast: a case report and review of the literature. Cytopathology 2007; 18: 384-7. (PMID: 17944956) [CrossRef]
4. Ilvan S, Celik V, Paksoy M, Cetinaslan I, Calay Z. Inflammatory myofibroblastic tumor (inflammatory pseudotumor) of the breast. APMIS 2005; 113: 66-9. (PMID: 15676017) [CrossRef]
5. Haj M, Weiss M, Loberant N, Cohen I. Inflammatory pseudotumor of the breast: case report and literature review. Breast J 2003; 9: 423-5. (PMID: 12968967) [CrossRef]
6. Sastre-Garau X, Couturier J, Derre J, Aurias A, Klijanienko J, Lagace R. Inflammatory myofibroblastic tumour (inflammatory pseudotumour) of the breast. Clinicopathological and genetic analysis of a case with evidence for clonality. J Pathol 2002; 196: 97-102. (PMID: 11748648) [CrossRef]
7. Khanafshar E, Phillipson J, Schammel DP, Minobe L, Cymerman J, Weidner N. Inflammatory myofibroblastic tumor of the breast. Ann Diagn Pathol 2005; 9: 123-9. (PMID: 15944952) [CrossRef]
8. Sari A, Yigit S, Peker Y, Morgul Y, Coskun G, Cin N. Inflammatory pseudotumor of the breast. Breast J 2011; 17: 312-4. (PMID: 21410588) [CrossRef]
9. Pettinato G, Manivel JC, Insabato L, De Chiara A, Petrella G. Plasma cell granuloma (inflammatory pseudotumor) of the breast. Am J Clin Pathol 1988; 90: 627-32. (PMID: 3177278)
10. Coffin CM, Humphrey PA, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor: a clinical and pathological survey. Semin Diagn Pathol 1998; 15: 85-101. (9606801)