Case Report

## Mammary duct ectasia and periductal mastitis in males

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## **ABSTRACT**

Mammary duct ectasia/periductal mastitis is exceedingly rare in males. Ten cases have been reported in the literature, the last 2 cases were associated with Human immunodeficiency virus. The previously reported cases presented in females with blood stained discharge or subareolar inflammatory process. All cases required surgical treatment, 7 out of 10 cases underwent a form of mastectomy. To the best of our knowledge this disease entity was not reported in patients suffering from Behcet's disease. This article described 2 new cases, one of them is a young man who had associated Behcet's disease. An immune mechanism may be responsible for such association, which was not reported before. The clinical presentation, pathological findings and management of these 2 new cases were outlined followed by literature review.

Keywords: Mammary duct ectasia, mastitis, male breast, Behcet's disease.

Saudi Med J 2001; Vol. 22 (11): 1030-1033

Duct ectasia and periductal mastitis are benign conditions affecting the breast. The first description of both ductal dilatation and periductal inflammation was made by Bloodgood<sup>1,2</sup> in 1923, the term duct ectasia was popularized by Haggenson<sup>1,2</sup> in 1951. Although duct ectasia is the most common cause of nipple discharge in women,<sup>3,4</sup> it is exceedingly rare in males. From 1974 -1996 only 10 cases were reported in the literature.<sup>5-11</sup> To the best of our knowledge the association of duct ectasia with Behcet's disease was not reported. The aim of this article is to report 2 new cases of male duct ectasia and periductal mastitis, one of them is a young man suffering from Behcet's disease, and to review the literature for this rare entity.

**Case Report.** *Patient 1.* A 23-year-old man presented with right breast blood stained spontaneous nipple discharge of one-month duration. There was no previous history of breast problems or trauma. Clinical examination confirmed the discharge and no masses could be detected in both

breasts and axillae. Cytological examination of the discharge was positive for blood but no abnormal cells were detected. Right retroareolar density was seen in mammogram. The patient was a non smoker and no family history of breast cancer. He was a victim of Behcet's disease for the previous 4 years before his presentation with nipple discharge. The diagnosis of Behcet's disease was made for the presence of oral and genital ulcers with arthritis, his older brother had the same manifestations with central nervous system symptoms. The patient has been on colchicine tablets since the diagnosis. The patient underwent excision of the ductal system through curvilinear incision along the upper half of the areola-skin junction.<sup>12</sup> The histopathological examination showed, dilated ducts filled with secretions and macrophages and surrounded by fibrosis and inflammatory cells infiltrate. Benign ductal hyperplasia and apocrine metaplasia were prominent in certain sections. The overall picture was consistent with duct ectasia and periductal mastitis.

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Received 28th April 2001. Accepted for publication in final form 7th July 2001.

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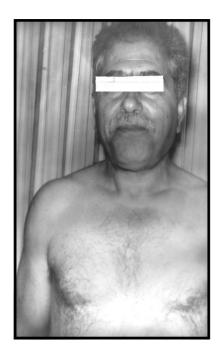


Figure 1 - The right areola/nipple complex was distorted in comparison with left side.

**Patient 2.** A 50-year-old man presented with a right breast subareolar painful mass of 2 weeks duration. Clinical examination revealed a retracted nipple; swollen areola and juxta-areolar area with redness, hotness and ill-defined tender fluctuant mass 3x3 cm (Figure 1 and 2). There were no palpable axillary lymph nodes in both sides and the other breast was normal. He had no family history of breast cancer. A fine needle aspiration was made through the nipple in the outpatient clinic, 5ml of yellow, thick pus was obtained and sent for culture, sensitivity and cytology. The patient was started on cefuroxime 500mg twice daily



Figure 2 - Closer view shows retracted nipple.

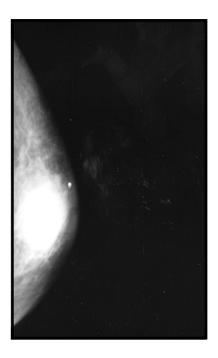


Figure 3 - Mediolateral view mammography shows thickened areolar skin and a retro-areolar dense mass.

metronidasole 500mg 3 times daily and returned to the clinic after 10 days. The culture results were staphylococcus coagulase negative. The cytology report described inflammatory cells with no evidence of malignancy. The patient had significant improvement after aspiration and antibiotics, but an ill-defined indurated slightly tender retroareolar area persisted when he was seen in the clinic. Mammography performed during the 2nd visit revealed thickened areola and retroareolar dense mass (Figure 3). The ductal system along with the indurated area were excised by the same surgeon and with the technique used for the previous case. The histopathological examination revealed, an abscess cavity, some ducts were dilated, elongated and surrounded by mononuclear inflammatory cells. The findings were consistent with periductal mastitis and duct ectasia.

**Discussion.** Duct ectasia/periductal mastitis is a benign disease complex of uncertain etiology. As it is more common in females, pregnancy and lactation were incriminated as a cause of the disease, but the condition was reported in virgins<sup>13,14</sup> and males. Some authors believe it is an involutional change of the breast ductal system due to the aging process.<sup>15</sup> One to 2 thirds of patients are smokers.<sup>16</sup> Cigarette smoke may damage the ductal epithelium by its direct toxic effect8 or indirectly by influencing the blood flow and hormonal action on the duct

epithelium.<sup>13</sup> In duct ectasia and periductal mastitis, there is growing evidence indicating that both aerobic and anaerobic bacteria play a significant role in this condition.<sup>1,14</sup>

The uncertainty of this disease entity is extended to its pathogenesis, the earlier investigators believed that duct dilatation preceded the accumulation of thick fluid in the ducts as a result of hormonal changes and excessive desquamation of ductal These changes are subsequently followed by periductal inflammation when ductal contents leak throw the wall of thin and damaged ducts. 16,17 The concept of duct ectasia/periductal mastitis was challenged by some authors 14,16,18 who stated that the pathological process started as periductal mastitis with subsequent ductal dilatation secondary to destruction of the elastic lamina supports the ducts. Recently Dixon and his colleagues<sup>2</sup> made a further challenge to the pathogenesis of this condition. They stated that duct ectasia and periductal mastitis were not strictly linked pathological processes.

The incidence of duct ectasia/periductal mastitis is highly variable<sup>1,19</sup> (2%-25%). **Browning** associates<sup>20</sup> noted a histologically proven duct ectasia present in 4% of patients who had associated breast symptoms and 8% of patients where duct ectasia was recognized as an incidental finding. Although the disease principally affects women it is exceedingly rare in men. Tedeschi and McCarthy<sup>5</sup> reported the first case of duct ectasia/periductal mastitis in 1974, Downs and associates<sup>6</sup> recorded the last 2 cases in the literature in 1996, their cases were associated with HIV infection, they attributed the condition to impaired immunity and increased susceptibility to repeated nipple infection. The disease was reported in a 3 year-old boy who presented with metachronous, bilateral blood stained discharge.<sup>7</sup> In adults, the youngest patient was 28 year old who had duct ectasia in association with focal epitheliosis and intraductal hyperplasia,8 the remaining 5 adult patients<sup>9-11</sup> reported in the literature (47-62 years old) presented with blood stained nipple discharge or recurrent subaerolar lump. Some patients had a mammary fistula, either spontaneous or after drainage of an abscess. Seven out of 10 patients were treated by a form of mastectomy, either subcutaneous<sup>7,8</sup> or including the areola – nipple Our recent 2 cases had the same presentation as previously reported, they were treated by excision of the ductal system sparing the unnecessary removal of the areola/nipple complex<sup>10</sup> or the unaffected fat in the mammary area. They had no complications or recurrence of symptoms 20 and 21 months after treatment. The 2nd case in this report was similar to others reported in the literature, but he was a heavy smoker. Cigarette smoke could be an etiological factor as in female entity.

The other case in this report is a young man suffering from Behcet's disease, the histological finding of his ductal system showed in addition to duct ectasia and periductal mastitis, a benign ductal hyperplasia and apocrine metaplasia, these findings are inpart similar to the case reported by McClure et

The relationship of ductal pathology and Behcet's disease was not recorded in the literature. Behcet's disease is a multisystem disorder characterized by a recurrent inflammatory reaction. Ocular, articular, vascular, intestinal, pulmonary and neurologic involvement were reported as manifestations of this disease.21 Its etiology is not well understood, most studies proposed that an immunological mechanism plays a significant role in its pathogenesis.<sup>21,22</sup> The coexisting duct ectasia/periductal mastitis and Behcet's disease may suggest immune factors could have an implication to the development of duct ectasia and periductal mastitis. Inspite of one experimental study<sup>23</sup> proposing a possible immune mechanism might be responsible for the development of duct ectasia/periductal mastitis, further studies are required for verifying this postulation.

Acknowledgments. The author would like to thank Professor A. S. Alsroujieh, G. F. Farah (Department of Surgery) and Professor F. Kamal (Department of Pathology) for their criticism and technical help, and thanks to Mrs. Fatima Wahdan for typing the manuscript.

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