Mammary Duct Ectasia in a Man with Liver Disease, End Stage Renal Failure, and Adjacent Arteriovenous Fistula

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ABSTRACT

Mammary duct ectasia is a rare finding in males. We report a case of mammary duct ectasia in a 58 year old male with liver failure and end stage renal failure. We discuss radiology findings of mammary duct ectasia as well as potential risk factors and management options for symptomatic male mammary duct ectasia.

CASE REPORT

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A 58 year old male was referred to a general breast surgeon by his primary care physician with a complaint of gynecomastia with heavy bloody nipple discharge from the left breast for several months. Nipple discharge was often soaking through shirts within a few minutes of donning them. The patient's medical history included seven years of end stage renal failure requiring hemodialysis through an arteriovenous (AV) fistula, hepatitis C infection, hypertension, coronary artery disease, and seizures. His social history included use of alcohol, tobacco, and recreational intravenous drugs (primarily heroin).

Physical examination revealed bilateral gynecomastia with a left breast predominance, an indistinct 1-2 cm mass in upperouter quadrant of the left breast, and abundant sanguinous discharge from a single duct of the left nipple. Examination of the left arm revealed an AV fistula surgically created for hemodialysis anterior to the head of the humerus that was patent with easily auscultated bruit and palpable thrill. The patient exhibited a moderately distended abdomen dull to percussion with caput medusa sign. The patient demonstrated mild confusion and hepatic encephalopathy. Laboratory examination is summarized in table 1. Our patient fell within the Child-Pugh liver failure class B and National Kidney Foundation stage 5 of chronic kidney disease [1,2].

Radiographic evaluation included a digital mammogram (Figure 1) and galactogram (Figure 2) followed by ultrasound examination (Figure 3). The mammogram revealed diffuse gynecomastia with extremely dense fibroglandular breast parenchyma limiting the sensitivity of mammography. There was marked dilation of veins leading from the breast into the body wall. Galactogram demonstrated a markedly dilated ductal system indicative of mammary duct ectasia. Multiple

small filling defects were suggestive of blood clots, intraductal papillomas, or less likely, air bubbles. Ultrasound of the left breast also identified a dilated ductal system with internal debris and no solid masses.

Due to copious and disruptive nipple discharge as well as difficulty achieving full mammographic visualization of the breast tissue, the patient underwent left total mastectomy with sentinel lymph node biopsy. The mastectomy was performed without immediate complication and the patient was discharged that same day with a JP drain at the surgical site. The patient's post-operative course was complicated by a hematoma at the mastectomy site and readmission 13 days post-operatively with fever, altered mental status, and purulent drainage around the JP drain. The patient was treated with JP drain removal and antibiotics and was discharged after resolution of his wound infection and encephalopathy.

Pathological examination of the left breast (Figure 4a-e) confirmed benign pathology with gynecomastia, duct ectasia, and chronic periductal inflammation. Pathological examination of the dissected sentinel lymph node revealed a benign lymph node.

The patient was well at one month of follow-up.

DISCUSSION

Mammary duct ectasia most commonly occurs among multiparous women in the fifth or sixth decade of life. It is principally characterized by chronic inflammation and fibrosis around a mammary duct clogged with debris. Its prime clinical and radiological significance is as a mimic for malignant disease of the breast [3]. Mammary duct ectasia accounts for 1-2% of symptomatic breast complaints in women but is a very rare finding in men [4]. Mammogram findings suggestive of mammary duct ectasia include subareolar ductal dilation without a coexisting mass, typically with calcified inspisated secretions within the ducts. Dilated subareolar ducts which are present bilaterally are also suggestive of duct ectasia over a malignant process. MR and US evaluation of duct ectasia typically reveals branching, fluid filled tubular structures below the nipple. As in this case, with US, inspissated secretions can often produce structures mimicing an intraductal papilloma [5]. In MR evaluation, papillary carcinomas and papillomas usually produce a more limited segment of T1 contrast enhancement than duct ectasia, and the presence of a contrast-enhancing mass superficial to the dilated ducts is much more suggestive of papilloma or papillary carcinoma [6].

There exists significant lexical confusion within the literature regarding the conditions of mammary duct ectasia and periductal mastitis [7]. Historically, mammary duct ectasia and periductal mastitis were viewed as separate stages of the same disease process and most early case reports regard the diseases synonymously, but more recent research suggests two separate disease processes. This discussion includes case reports of adult males with both periductal mastitis and

mammary duct ectasia. Periductal mastitis is strongly associated with cigarette smoking, infection, and abscess formation. On the contrary, mammary duct ectasia is not associated with smoking or sepsis. Rather, mammary duct ectasia seems to result from aging, involution of the duct outlet, and inspissation of debris within the duct. The small number of reported cases in males makes it difficult to authoritatively categorize the diseases in men.

The authors' review of the literature revealed 13 previous case reports of males with mammary duct ectasia/periductal mastitis [7-14]. In the available case reports of male mammary duct ectasia/periductal mastitis, gynecomastia was mentioned as a finding in 2 of 13 cases, [10,13] specifically excluded in 1 case, [7] and not mentioned in the other 10 cases. Immune compromise is a common theme in 4 of 13 case reports: two cases report HIV infection, [11] one case reports Behçet's disease, [12] and one case reports recent cisplatin and gemcitabine chemotherapy for lung cancer [7].

This report is unique in describing male duct ectasia in the setting of an adjacent AV fistula in the ipsilateral arm or with comorbid hepatic or renal failure. One report discusses a patient that, "was hepatitis B immune from a previous infection [11]." The elevated venous pressure and flow from the neighboring AV fistula likely contributed to the volume of sanguinous nipple discharge expressed from the left breast and perhaps played a primary role in the occurrence of male mammary duct ectasia, but the unilateral, single duct discharge in this patient raised clinical suspicion for an underlying invasive mass. The marked gynecomastia found in this case may reflect the development of male mammary duct ectasia secondary to hepatic insufficiency and increased female sex hormones. Our patient presented with three conditions associated with gynecomastia: liver failure, renal failure requiring hemodialysis, and intravenous drug use [15]. This case might also suggest a role for immune compromise shared by 4 of 13 prior reported males with duct ectasia; renal and hepatic failure have both been implicated in immune compromise. Hemodialysis dependent patients have reduced counts of T4+ and T8+ T-lymphocytes, blunted responses to vaccination, immune cells which demonstrate attenuated responses to in vitro assays of immune competence, and they experience a high risk for infections [16,17]. Liver failure has similarly been implicated in altering immune function; monocytes demonstrate altered level of cytokine production depending upon stage of liver failure [18].

The benign pathology of the excised breast and complicated post-hospital course suggest a more conservative treatment approach in future clinical encounters with adult men presenting with gynecomastia, copious nipple discharge, breast mass, and imaging findings consistent with mammary duct ectasia such as local surgery to address nipple discharge. Also, MR evaluation or additional imaging of the asymptomatic breast could have proven a valuable adjunct to the x-ray and US imaging obtained for this patient in guiding a less invasive management approach. However, without more definitive imaging characteristics to guide a localized surgery, mastectomy was advised by this patient's surgeon.

TEACHING POINT

Mammogram and US findings suggestive of mammary duct ectasia include subareolar ductal dilation without a coexisting mass, typically with calcified inspisated secretions within the ducts and dilated subareolar ducts which are present bilaterally are also suggestive of duct ectasia over a malignant process. MR findings of mammary duct ectasia include diffuse contrast-enhancement on T1 images without an enhancing mass between the dilation and duct outlet. Mammry duct ectasia's prime clinical importance is as a clinical and radiographic mimic of malignant breast disease.

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FIGURES

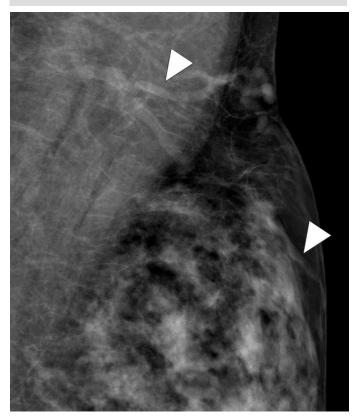


Figure 1: 58 year old man with duct ectasia: mammogram (MLO view) of left breast demonstrating diffuse gynecomastia with dense breast tissue limiting evaluation and marked dilation of veins leading into body wall (arrowheads)

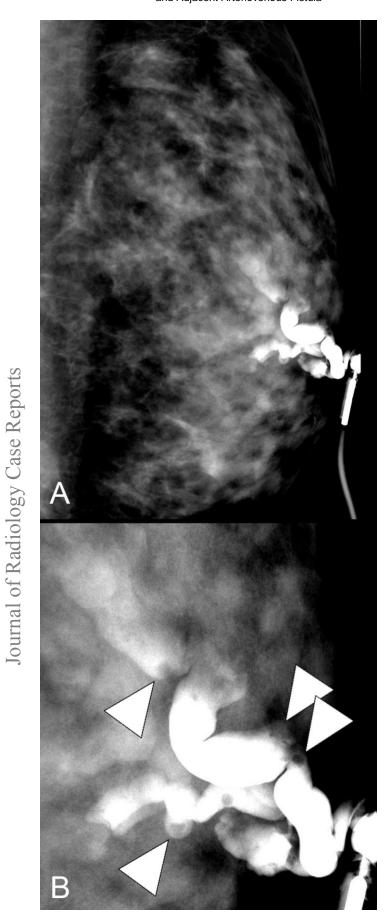


Figure 2: 58 year old man with duct ectasia: galactogram of left breast (a) revealed marked mammary duct ectasia and gynaecomastia (b = magnification).

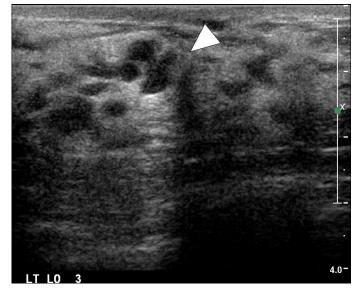


Figure 3: Ultrasound examination of 58 year old man's left breast with duct ectasia demonstrating dilated ducts in the retroareolar region and ill-defined hypoechoic mass suspicious for intraductal papilloma or papillary carcinoma identified by arrowhead (14 MHz, 5 cm linear transducer)



Figure 4a

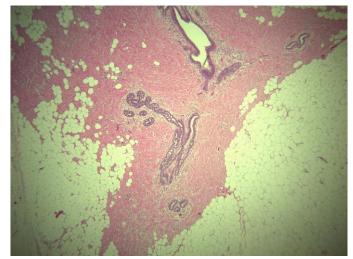


Figure 4b

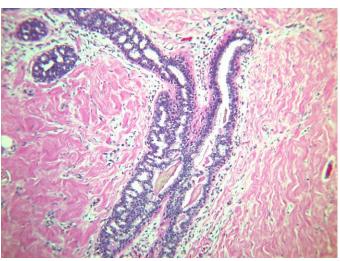


Figure 4c

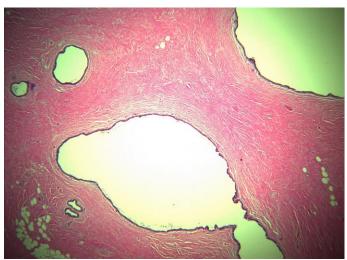


Figure 4d

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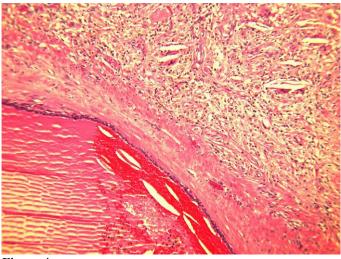


Figure 4e

Figure 4: a. Gross view of dissected breast; ectatic duct marked with arrowhead b. Low magnification view of gynecomastia (H&E; $100 \times$) c. High magnification view of gynecomastia with papillary hyperplasia of duct epithelium (H&E; $400 \times$) d. Low magnification view of mammary duct ectasia (H&E; $100 \times$) e. High magnification view of mammary duct ectasia demonstrating inspissated debris (lower left field), duct wall (middle) and chronic inflammatory and fibrotic changes around duct (upper right) (H&E; $400 \times$)

	Value	Reference range	
Creatinine	7.96 (†)	0.70-1.20 mg/dL	
Blood urea nitrogen	37 (↑)	9-20 mg/dL	
Ammonia	45 (↑)	9-33 μmol/L	
ALT	17 (\psi)	21-72 U/L	
AST	23 (↔)	17-59 U/L	
Albumin	4.2 (↔)	3.5-5.0 g/dL	
Protime	16.5 (†)	11.9-14.6 sec	
INR	1.3 (†)	0.9-1.1	
Bilirubin (direct)	<0.2	0.0-0.4 mg/dL	
Bilirubin (indirect)	Undetectable		

Table 1: Lab values of the patient

Breast Imaging:

Etiology	Subareolar breast ducts become filled with inspissated secretions following involution of duct outlet
Incidence	1-2% of symptomatic breast complaints for women, very rare in men
Gender ratio	Symptomatic mammary duct ectasia is overwhelmingly present in females
Age predilection	5 th and 6 th decades of life
Risk factors	Unknown
Treatment	Symptomatic, distinguish from malignant breast disease
Prognosis	Excellent

Table 2: Summary table of mammary duct ectasia

	Mammogram	US	MRI
Mammary duct ectasia	Subareolar ductal dilation,	Inspissated secretions may	T1 contrast enhancement in
	typically with calcified	mimic intraductal tumor. Free-	clumps or heterogeneously
	secretions. Often bilateral.	floating debris particles often	along entire duct [18]
		present.	
Papillary carcinoma	Ill-defined, spiculated hyper-	Ill-defined masses; hypoechoic	T1 contrast enhancement
	attenuating mass	relative to surrounding fat	along lesion and singular
			clump of ductal branching;
			lesion with poorly defined
			margins, spiculation
Papilloma	Retroareolar mass with sharp	Fluid filled ducts.	Similar to papillary carcinoma;
	margins		lesion enhancement often
			homogeneous and borders
			tend to be better defined

Table 3: Differential diagnosis table of mammary duct ectasia

ABBREVIATIONS

AV = arteriovenous US = Ultrasound

MR = Magnetic resonance

KEYWORDS

mammary duct ectasia, arteriovenous fistula, gynecomastia, renal failure, liver failure

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