

spondylitis.⁵ Hajjaj-Hassiouni *et al* described two cases of ankylosing spondylitis with the nephrotic syndrome where amyloid was demonstrated in a rectal biopsy specimen,⁶ Geffriaud *et al* followed up six patients with systemic AA amyloidosis and ankylosing spondylitis.⁵ Renal failure eventually occurred in all six between three and 31 years (with a mean of 19 years) after the onset of rheumatological symptoms.

Amyloidosis is a well recognised cause of the nephrotic syndrome, and end stage renal failure is the cause of death in 13–68% of cases of systemic AA amyloidosis.^{1 5 7–10} AA amyloidosis may present many years after the onset of the underlying inflammatory disorder, which may appear to be clinically inactive at the time of presentation.⁹ In a recently published series from the Mayo Clinic the mean time elapsed between onset of symptoms attributable to inflammatory disease and a tissue diagnosis of AA amyloid was 18.3 years.¹

Our patient developed a nephrotic syndrome 44 years after the onset of symptoms attributable to ankylosing spondylitis. At the time of writing he retains adequate renal function. Progression of his renal disease has thus been slower than in the six patients in Geffriaud's series.⁵

Immunostaining of amyloid with specific antisera to the components of amyloid fibrils is

not routinely performed in many pathology departments including, until now, our own. In the authors' opinion this case shows that immunostaining of amyloid should be routinely adopted to avoid overlooking occult or apparently quiescent underlying disease.

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Lobular carcinoma in a mammary hamartoma

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Abstract

Mammary hamartomas are uncommon breast lesions, sometimes presenting as mammographic abnormalities which require pathological clarification. Previous cases have all been benign. A unique case of mammary hamartoma containing atypical lobular hyperplasia (ALH), lobular carcinoma in situ (LCIS), and foci of microinvasive lobular carcinoma is presented. The need for adequately sampling macroscopically innocuous breast lesions is emphasised.

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Case report

A 59 year old woman had a 5 cm, well defined opacity incorporating a fatty component, which was located in the upper, inner quadrant

of the left breast, detected at mammography (fig 1). A soft, round, mobile mass was palpable and this was considered to be benign clinically. A fine needle aspirate showed very few, small clusters of benign epithelial cells. Because of the patient's anxiety, an excision biopsy was performed.

Pathology

The specimen measured 6 × 5 × 3 cm and contained an ovoid well circumscribed mass measuring 3 cm in maximum diameter. It had a yellow and white variegated cut surface and was firm and rubbery in consistency. Microscopically, the lesion was focally surrounded by a thin layer of fibrous tissue containing occasional compressed ducts. The overall pattern consisted of individual and clustered lobular units separated by dense, hyalinised fibrous tissue which were separated by abundant fatty

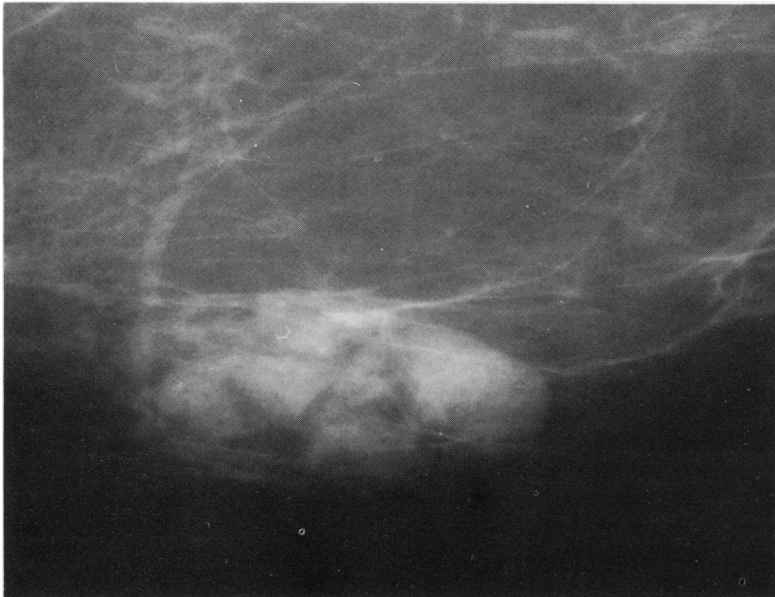


Figure 1 Circumscribed radiographic lesion with translucent areas.

tissues (fig 2). The normal, loose intralobular tissue was completely replaced by paucicellular, fibrous tissue. In those places where the fat approximated the epithelial elements, the ducts and acini retained a thin investing layer of fibrous tissue. The entire specimen was blocked, and sections from eight of the 14 lesional tissue pieces contained foci of lobular neoplasia. Throughout the eight sections 24 lobules (in six sections) showed atypical lobular hyperplasia (ALH), four ducts (in three sections) showed pagetoid spread, and four lobules (in three sections) showed lobular carcinoma in situ (LCIS). Two of the three sections showing foci of lobular carcinoma in situ displayed adjacent foci of microinvasive lobular carcinoma (fig 3). These two microinvasive carcinomas were present in two sections containing only one and two lobules, respectively, distended by LCIS. All foci of lobular neoplasia comprised clustered lobules in each section. A focus of atypical ductal

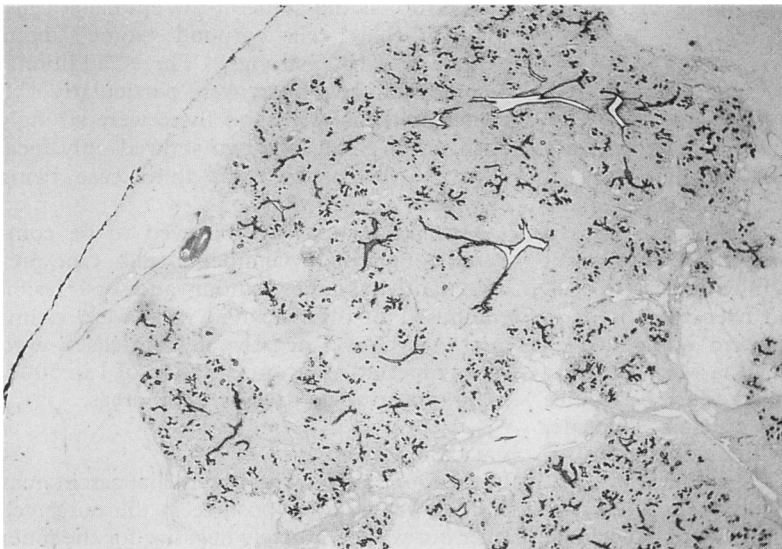


Figure 2 Lobules in dense fibrous stroma and interlobular adipose tissue.

hyperplasia was present in the small amount of attached fatty breast tissue.

Discussion

Arrigoni described the characteristic macroscopic and microscopic features of a breast lesion which he termed mammary hamartoma.¹ These lesions are well circumscribed and both clinically and radiologically may resemble a fibroadenoma. Histologically, they are composed of acinar units, arranged in a lobular pattern within a hyalinised, fibrous stroma and containing variable amounts of fat in the interlobular regions. In addition to these typical features, this case also contained foci of ALH and LCIS with two foci of microinvasive lobular carcinoma. Although epithelial hyperplasia is reported as occurring only rarely,² neoplastic lesions have not been documented in mammary hamartomas before as far as we know.^{3,4} It seems likely that its occurrence in this case, as in fibroadenomas, is coincidental and certainly not unexpected. Nevertheless, it emphasises the need for adequate sampling of macroscopically innocuous lesions as just over half the sections (57%) contained lobular neoplasia and only two (14%) contained microinvasive carcinoma.

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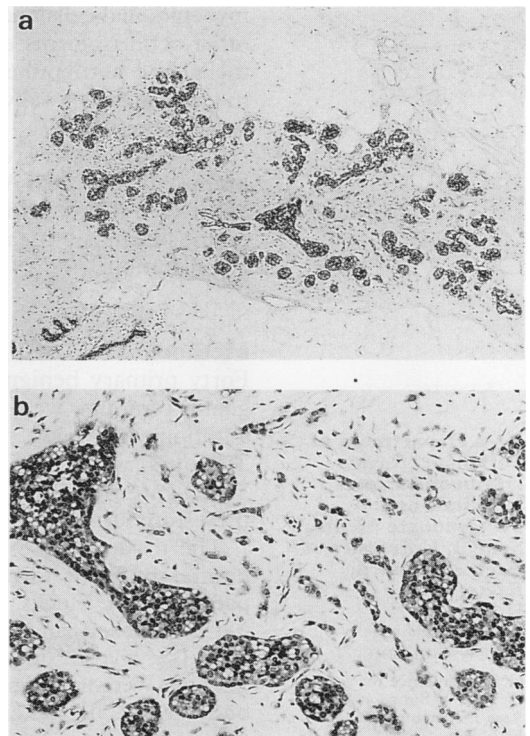


Figure 3 (A) Area of LCIS with invasive lobular carcinoma; (B) higher magnification of invasive component.