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Rare malignacies of the breast diagnosed and treated in the year 2001 at the Chair and Clinic of Oncology of the Poznań University of Medical Sciences

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Cancer is the most common neoplasm of the breast. Other tumours account for approximately 1% of all breast malignancies. Among over 600 patients with breast tumours, of whom as many as 230 were treated surgically in only one year at the 30-bed Department of Surgery of the Oncology Chair and Clinic, we have encountered as many as 5 cases of extremely rare histopathological types of breast malignancies. We present these 5 cases in order of their aggressiveness.

Rzadkie nowotwory piersi rozpoznane i leczone w Katedrze Onkologii AM w Poznaniu w 2001 roku

Najczęściej spotykanym nowotworem gruczołu piersiowego jest rak. Nowotwory inne niż rak stanowią około 1% ogółu złośliwych guzów piersi. Spośród ponad 600 guzów piersi, w tym 230 nowotworów złośliwych operowanych w jednym tylko roku kalendarzowym, w 30 łóżkowym Oddziale Chirurgii Kliniki Onkologii, rozpoznano 5 niezwykle rzadko występujących postaci histologicznych. Nowotwory przedstawiono w umownej kolejności, według agresywności ich przebiegu.

Key words: adenomyoepithelioma malignum, breast malignancies, carcinoma adenoides cysticum, fibromatosis infiltrativa mammae, hyperplasia stromalis pseudoangiomatosa, lymphoma malignum
Słowa kluczowe: adenomyoepithelioma malignum, carcinoma adenoides cysticum, fibromatosis infiltrativa mammae, hyperplasia stromalis pseudoangiomatosa, lymphoma malignum, nowotwory piersi

Carcinoma adenoides cysticum

Carcinoma adenoides cysticum is also referred to as carcinoma basocellulare cysticum, epithelioma muciparum, basalioma, adenocarcinoma cylindromatosum or adenomyoepithelioma [1]. According to the WHO classification the proper name, which directly refers to the malignant character of this tumour, is carcinoma adenoides cysticum. This particular tumour develops in the salivary glands, both large and small. Its most common site is the mucosa of the oral cavity, the palate and the paranasal sinuses. It can also be found in the parotid gland, the submaxillary gland and the additional salivary glands of the palate. The larynx is a more rare localisation [1]. It may also appear within the bronchi [2]. The breast is an extremely rare localisation, although it has been reported in literature [3].

Carcinoma adenoides cysticum develops slowly, sometimes over a few decades. It causes late distant metastases. Metastasising along blood vessels, it reaches the lungs, the brain, the bones and the liver. Metastases to local lymph nodes are less frequent. Postoperative recurrences are common, usually during the first three years after surgery. The tumour tends to infiltrate paraneural spaces, therefore it may often cause nerve paralysis and neuralgia. Multifocal lesions have been reported [4].

On microscopic examination the tumour is well delimitated, however, in about half of the cases, it infiltrates local tissues. On histopathological examination, it possesses a glandular component, which consists of the proliferating glandulae and the instertitium, with an abundance of basal membranes. The interstitium may form a variety of glandulae-derived tubular, trabecular, compact or cribriform structures (which imitate cribriform cancer). PAS staining confirms the presence of basal membranes in the interstitium, and especially within its cribriform structures.

Because carcinoma adenoides cysticum produces the carcinoembryonic antigen (CEA), this particular marker may be used to monitor disease progression.

Prognosis is poor. Approximately 30% of patients die during the first 5 years. Adjuvant therapy is considered

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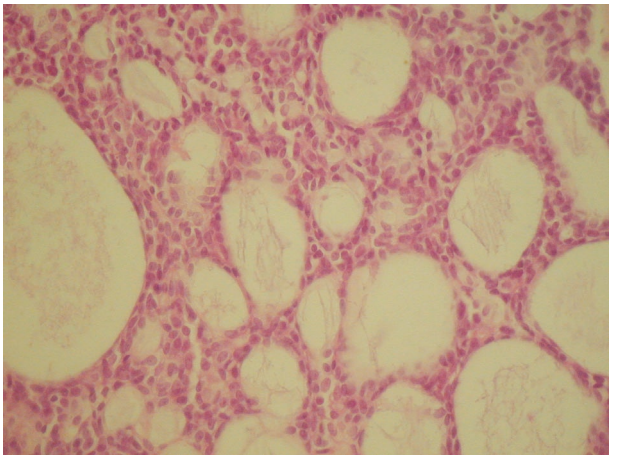

Figure 1. Carcinoma adenoides cysticum Hematoxylin & Eosin (H & E) staining; enlarged 400X

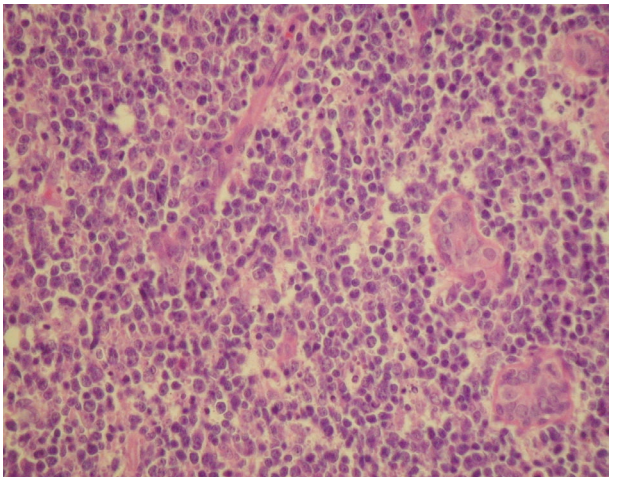

Figure 2. Diffuse large B cell lymphoma H&E staining; enlarged 400X

a necessity, and despite the relative low radiosensitivity high dose irradiation is advised (65-82 Gy) [1].

We have diagnosed carcinoma adenoides cysticum in a 60-year old female patient. She was referred due to a tumour in the left breast. She had been observing the tumour for 8 years, and she claimed it had grown and become painful over the last 18 months. Mammography and fine needle biopsy were performed. PCI provided the diagnosis of cancer cells. The encapsulated tumour was excised whole, within healthy tissue margins and on histopathological examination, was found to be, to our disbelief, fibroadenoma mammae – epithelioplasia florida. The patient remained under very strict follow-up observation. After 10 months, on examination, we found a small tumour – 10 mm in diameter – in the medial end of the postoperative scar. The patient underwent wide excision of the tumour. On microscopic examination, it was found to contain three separate tumours, 10 mm, 6 mm and 2 mm in diameter, containing glandular cells forming cribiform structures. Parts of the lesion had intraductal characteristics, with central necrosis. Atypia of a low grade was also found. The final diagnosis was carcinoma adenoides cysticum et carcinoma ductale *in situ* cum invasione focali. We re-examined the specimen obtained during the first excision and found a focus of this rare malignancy along the excision margin. In view of these findings, we performed radical mastectomy modo Patey. No malignant tissue was found either within the breast or in the 17 excised axillary lymph nodes. During immunohistochemical investigation we found poor positive results for estrogen receptors (+), and positive results for progestogen receptors (+), protein p53 (+), c-erb-2 (+) and cathepsin D (+). After surgery we started tamoxifen therapy. The patient is now under follow-up and has remained recurrence-free for 22 months after mastectomy.

Lymphoma malignum

Lymphatic system malignacies seldom appear as primary tumours of the breast. It is estimated that Non-Hodgkin Lymphomas account for some 0.1% of all breast malignancies [5]. On the other hand, only 1% of lymphomas are diagnosed as primary breast lesions – as such a diagnosis can only be pronounced if there are no other malignancies in any other localisation except for unilateral axillary lymph nodes. B-cell lymphomas appear to be the most common – accounting for some 80% of cases. Due to the extreme rarity of these tumours, the patient groups reported in literature never contain more than 20 subjects. Usually one encounters centroblastic lymphomas (diffuse large B-cell lymphoma, DLCL). Ribrag et al. report a series of 16 patients treated for DLCL primarily localised in the breast [6]. Other lymphomas initially diagnosed as primary breast tumours include MALT (low-grade mucosa-associated lymphoid tissue), diffuse large B cell lymphoma, follicular lymphoma, Burkitt's lymphoma and MCL (mantle cell lymphoma). The histopathological picture of the most common of these – the DLCL – is dominated by a homegenous population of atypical lymphoid cells (characterised, on immunohistochemical examination, by a positive reaction for the lymphoid cell antigen LCA and a negative reaction for cytokeratins). These cells infiltrate the interstitium and, rarely, the lobular and ductal component (imitating carcinoma

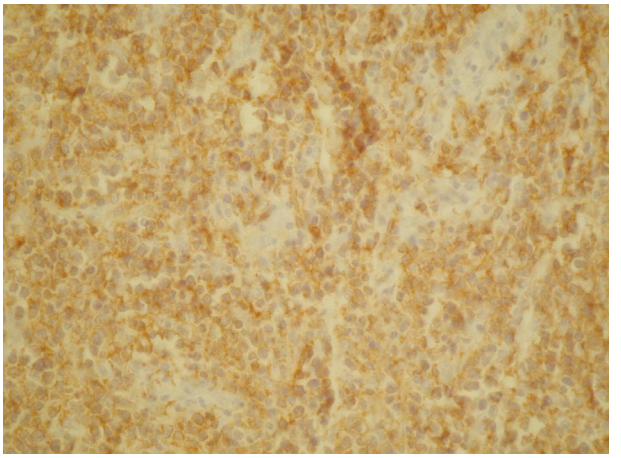

Figure 3. Diffuse large B cell lymphoma, positive reaction to CD20 (immunohistochemical analysis); enlarged 400X

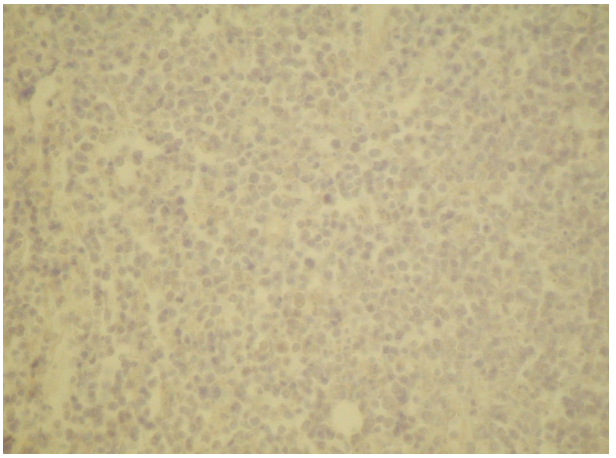

Figure 4. Diffuse large B cell lymphoma, weak positive reaction to CD3 (immunihistochemical analysis); enlarged 400X

in situ). In this population the majority of cells are lymphocytes B, characterised by a positive reaction for the presence of the CD20 receptor. One may also find a small number of lymphocytes T, characterised by a weak positive reaction for the presence of the CD3 receptor.

The lesions may be found both in uni- and bilaterally [7]. The disease is found almost exclusively in women, although literature presents one case report of a 69-year old man with gynecomastia and primary lymphoma of the breast [8]. The authors suggest that the lesion may have been related to the increased estrogen level in the male patient. There also exists a report of a 33-year old female patient diagnosed for bilateral rapid breast enlargement [9]. Over a period of 6 weeks, both her breasts increased three-fold. Fine needle biopsy, followed by surgical biopsy, allowed to establish the diagnosis of lymphoblastic lymphoma. In the course of further investigations, including bone marrow examination, the patient was found to have acute lymphoblastic leukaemia. In this case, the breasts were the primary sites of the disease. The breast is also a very rare site for secondary localisation of lymphoma. Boullanger et al. [10] report a case of a 71-year old woman diagnosed for fatigue and neck and axillary lymphadenopathy. In the course of diagnosis, lesions were also found within the breasts and the eyelids. Mantle cell lymphoma (MCL) was diagnosed and the rare localisations of the disease were pronounced as proof of particular aggressiveness of the disease.

On palpation, one usually finds a sole, movable tumour. However, the lesions may be wider, without evident discernible margins. Diagnosis may be set after histopathological examination, biochemical analyses (immunoglobulins), bone marrow biopsy, spinal puncture and imaging techniques.

The treatment of lymphomas of the breast has not been systematised, due to the few cases observed. However, over the last few years one may observe a tendency to move from surgical treatment combined with adjuvant radiotherapy to chemotherapy combined with loco-regional irradiation. Ribrag et al. [6] have presented a paper, in which the majority of patients were treated according to the CHOP protocol, or similar, i.e. regimes containing cyclophosphamide, hydroxyrubicine, vincristine, prednisone and, additionally – VM26 and bleomycine. The patients had received 3 to 10 courses (6 on average). In 16 out of 20 patients, complete remission was achieved, in two – partial remission. Six patients from this group are dead – four died in the course of progression to the central nervous system. Some 15% of patients underwent recurrence in the contralateral breast. Other common recurrence sites were the bones, the maxillary sinus and the retroperitoneal lymphatic nodes. Mean follow-up was 54 months (10-144 months). Overall survival was 67%, while disease-free survival was 50%.

At the Chair and Clinic of Oncology in Poznań we diagnosed primary lymphoma of the breast in a 52-year old woman who had undergone thymoidectomy for myasthenia gravis 25 years before. She was in very good condition and had not been receiving any medication. She sought surgical consultation because of a 3x4 cm lump in the right breast. We performed a biopsy of the lesion. The diagnosis was "cellulae atypicae". The tumour was excised with wide surgical margins and was, on examination, found to consist of diffuse large B cell lymphoma. In all additional investigations, including bone marrow biopsy, no other sites of disease were found. The patient received six courses of CHOP over the next 16 weeks. Treatment tolerance was very good. Then, over the next 8 weeks the patient underwent photon irradiation (6MeV) – 40 Gy/T to the right breast, 40 Gy/max to both the cervical lymph nodes and the supraclavicular lymph nodes and 8.2 Gy/T to the right axillary nodes. Tolerance of radiation treatment was also very good. During follow-up examination no recurrence was found either within the right breast nor in other sites.

Adenomyoepithelioma malignum

Adenomyoepithelioma malignum is a malignant tumour containing the epithelium and the myoepithelium. Myoepithelial cells are found between the epithelial cells

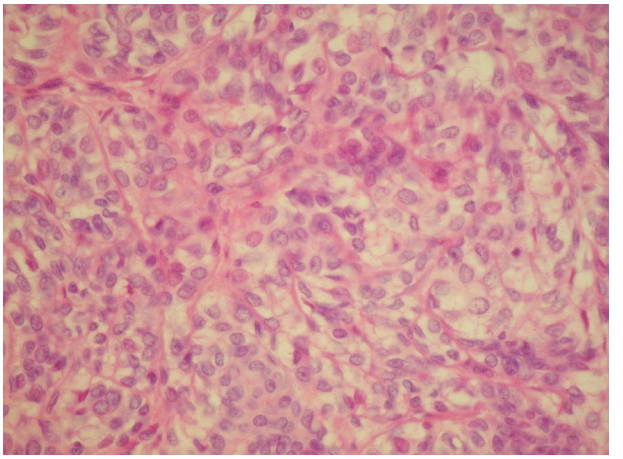

Figure 5. Adenomyoepithelioma malignum H&E staining; enlarged 400X

and the basal membrane. The tumour itself may occur within the ducts or lobuli of the breast, in the sudoriparous glands and, especially, in the salivary glands. It consists of elongated, flat cells, usually discerned using immunohistochemical methods for the presence of myosin, cytokeratin, S-100 protein and and kalponin. Disorders of myoepithelial cell differentiation are being widely investigated, as they are responsible for the development of a number of malignancies – adenoid cystic carcinoma, adenomyoepithelioma (AME), low-grade adenosquamous carcinoma, poorly differentiated myoepithelial-rich carcinoma and pure malignant myoepithelioma. The former of these are characterised by slow growth and seldom metastasise. Pure malignant myoepithelioma metastasises early, while its characteristics render it similar to sarcomatoid carcinoma [11].

Adenomyoepithelioma may be found in salivary glands, and, rarely – in the skin and the breasts. The malignancy is most common in women above 50 years of age. There exists a report of its occurrence in a 47-year old man [12]. Its characteristic feature is its mixed texture – there are case reports of AME co-existent with sarcomatoid carcinoma and low-grade adenosquamous carcinoma [13], as well as osteosarcoma and ductal carcinoma [14]. The least common form (only 12 cases reported to date) is pure malignant myoepithelioma, i.e. a tumour consisting only of malignantly differentiated myoepithelial cells [11].

On palpation, AME is usually found to be a relatively hard, well-delimited tumour, brownish-white on cross-section and often containing foci of necrosis. It seldom exceeds 20-35 mm in diameter, although some lesions have been found to reach as many as 86 mm [15-16].

During histopathological examination, one finds proliferation of glandular elements and of myoepithelial cells. Glandular elements appear as round, oval or cylindrical; glandular cells present a positive reaction for cytokeratins (e.g. the CAN 5.2 type antibody) in immunohistochemical staining. The myoepithelial cells appear as fusiform or polyglonal, with hyperchromatic, atypical nuclei. They infiltrate and overgrow into the interstitium and around the glandular elements. They present a positive reaction for smooth muscle actin and a negative reaction for cytokeratin (e.g. the CAN 5,2 type antibody) in immunohistochemical staining. Atypical mitosis may be observed.

Because of so few recognised cases (approx. 55 [17]) there exists no standard treatment. Van Hoeven et al. have presented an extended case report of 13 patients, who had undergone mastectomy, and 19 patients treated with tumour excision. After 12 to 124 months of follow-up 20% of the patients after tumour excision were found to have local recurrence. Despite this, the authors maintain that only tumours over 30 mm in diameter ought to be treated aggressively, than only with local resection [15]. Takashi et al. [18] report the case of a 60-year old woman who underwent mastectomy for a tumour infiltrating the skin with uninvolved axillary nodes. Despite radical

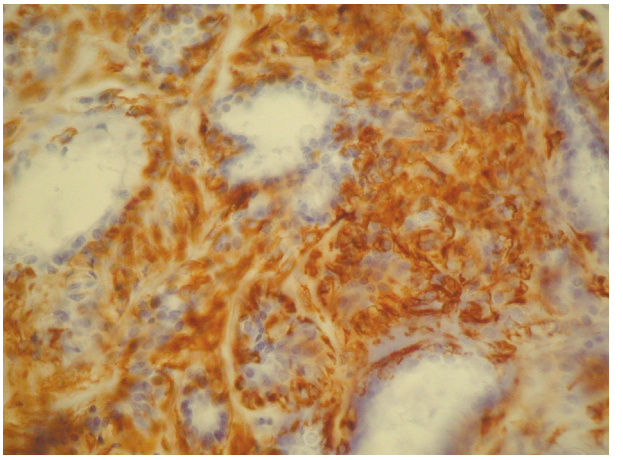

Figure 6. Adenomyoepithelioma malignum, positive reaction of myoepithelial cells to SMA (immunohistochemical analysis); enlarged 400X

treatment, the patient presented with metastases to the bones, the lungs and the mediastinum. The authors stress that AME has a tendency to metastasise along blood vessels, yet up to date there exists no report concerning metastases from tumours initially less than 20 mm in diameter [19].

At present the collected data shows, that AME is a tumour of low malignancy [11]. Local recurrence is relatively common after lumpectomy, but has only been observed once after mastectomy. Metastases are rare, and much more common if the tumour consists of a few different co-malignancies. In such cases the most common site of metastases are the lungs.

At the Chair and Clinic of Oncology of the Poznań University of Medical Sciences we have diagnosed on case of adenomyoepithelioma malignum in a 63-year old woman. The patient sought consultation as she had found a lump in the right breast during self-control. On palpation, we found a solid, movable tumour 20x30 mm located between the external quadrants. In mammography, the lesion was described as an irregular condensation with processes, and in ultrosonography – as a hypoechogenic lesion 18x13x12 mm in size. Both the clinical and the radiological picture suggested the diagnosis of non-metastatic cancer. In order to confirm this diagnosis we performed small-needle biopsy, which confirmed the diagnosis of a malignancy. Radical mastectomy modo Patey was performed. On histopathological examination, the tumour was found to be 20x15 mm in diameter, and appeared as a double-phase papilloma containing adenosis sclerosans. The internal part of the lesion contained foci of necrosis. The interstitium was focally infiltrated by poli-mitotic mesothelial cells, which suggested that the tumour arose from malignant differentiation of the myoepithelial parts of the lesion. *Carcinoma ductale in situ* was also found within the lesion, at one point infiltrating the interstitium. Other histopathological features found within the lesion included: mastopathia fibrosa et cystica, microcalcificationes, fibroadenoma hyalinisans. In the course of surgery 20 axillary nodes were removed which, on

histopathological analysis, were found to contain lymphonodulitis reactiva. In the course of immunohistochemical analysis the following markers were found – CAM 5,2, cytokeratin CK-7, cytokeratin CK-20, actinomyosin ACT, protein S100. In the course of follow-up, over a period of 25 months, the patient is free of recurrence.

Hyperplasia stromalis pseudoangiomatosa

Hyperplasia stromalis pseudoangiomatosa (PASH) often accompanies benign or malignant lesions within the breast. It is usually a chance finding in the course of microscopic examination. In very rare cases, it may form a palpable lesion. The largest group of patients with PASH, consisting of 40 women, has been reported by Powell et al. [20]. The palpable lesions are initially recognised as fibroadenomas. Material obtained from small needle biopsy does not allow to discern the characteristics of PASH which allow to differentiate it from fibroadenoma or tumour phylloides [21, 22]. On histopathological examination the interstitium significantly dominates over the lobular and ductal components. It is necessary to stress that one of the characteristic features of this malignancy is that the interstitium contains mainly collagen, in which the vacant places form irregular patterns adjoining with anastomoses divided by collagen fibres. In some cases, the free spaces contain erythrocytes. Myofibroblasts are found on the outskirts of the tumour. They react positively to cytokeratine and factor VIII on immunohistochemical examination. The ramified system of fissures between the fusiform myofibroblasts may cause the tumour to be erroneously recognised as angiosarcoma. Further diagnostic is predominantly immunohistochemical. The fusiform cells present a positive reaction in tests for vimentin (VIM) and for CD34 receptors, and negative reactions with factor VIII. In a majority of cases, nuclear estrogen receptors cannot be found [20]. Although this particular hyperplasia usually occurs in women, yet there have been reports of its occurrence in men. Damiani and Eusebi [23] describe two cases of PASH in men with gynecomastia and concomitant neurofibromatosis.

On palpation, the lesions are well delimitated, usually a few centimetres in diameter. The largest reported tumour was 15 cm in diameter [24]. On cross-section, the tumour is a greyish-white, fibrous and homogenous.

The aetiology of the disease remains unclear. Abnormal reactions to endo- and exogenous hormones suggest a significant influence of hormonal factors on the development of PASH.

The main treatment method is local excision. Recurrence is rare – over a follow-up period between 6 months and 11 years (4.5 years on average) we recognised five local recurrences in a total of 40 patients [20]. Two patients from this group developed PASH in the contralateral breast. Multifocal lesions are more difficult to treat and in such cases, simple mastectomy should be considered.

At the Chair of Oncology of the Poznań University of Medical Sciences we recognised one case of hyperplasia stromalis pseudoangiomatosa in a 19-year old woman. The patient was referred due to a rapidly growing mass in the right breast. During the two months after the initial diagnosis the (tumour) took up some 75% of the entire volume of the breast. She developed striae on the skin; the asymmetry of the breasts was considered to be 50%. Ultrasonographically, we found a policyclic, hypoechogenic tumour with linear liquid-filled spaces. Cytological analysis was concomitant with benign fibroadenoma iuvenile giganteum or tumour phyllodes benignus (lobules of hyperplastic ductal epitelium with numerous non-atypic myoepithelial cells). We performed local excision of the tumour. It was well delimitated, 16x12x2.5 cm in size. On cross-section, it was the colour of flesh, fibrous, with tiny cysts filled with a greenish, viscous substance. On microscopic examination the picture was dominated by fibrotically modified breast tissue with its lobular and ductal structure maintained. We did not discern any adipose tissue. In order to verify the initial diagnosis we performed immunohistochemical analysis,

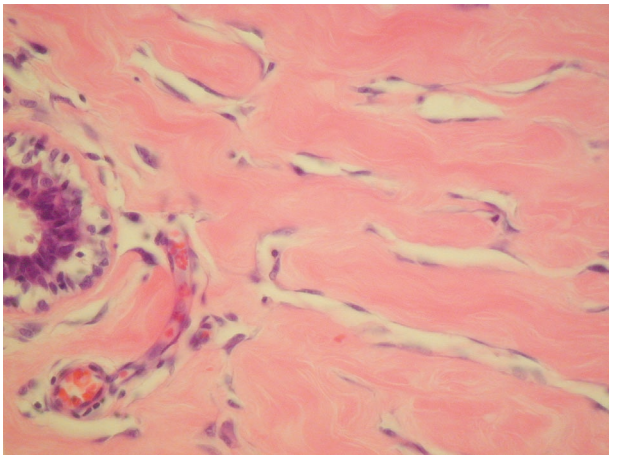

Figure 7. Hyperplasia stromalis pseudoangiomatosa H&E staining; enlarged 400X

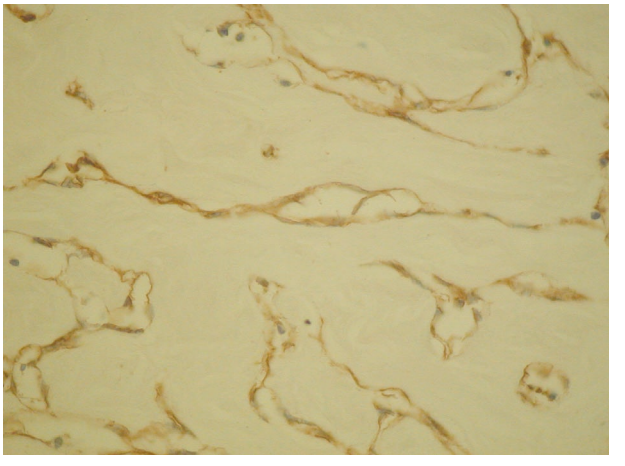

Figure 8. Hyperplasia stromalis pseudoangiomatosa, positive reaction to the CD34 marker (immunohistochemical analysis); enlarged 400X

which allowed to discern the following markers: CD34, VIM, factor VIII, desmin (DES), smooth-mucle actin (SMA). During the most recent follow-up control, two months after the surgical procedure, we did not observe signs of recurrence.

Fibromatosis infiltrativa mammae

The term fibromatosis refers to different forms of connective tissue proliferation. The histopathological picture is similar, regardless of the affected organ. The clinical picture depends upon the anatomical localisation of the lesion. Fibromatosis may be found in muscles, e.g. in the sternocleidomastoid muscle (torticollis), the fasciae e.g. the palmar or plantar fascia (fibromatosis palmaris or plantaris). Fibromatosis located in the penis is referred to as Peyronie's disease, and within the ureter as Ormand's disease. The proliferating and fibrotic connective tissue causes compression of nerves and blood vessels while the aggressive growth resembles the growth of a malignancy. It is often impossible to remove the lesion without any destruction to the neighbouring structures, while recurrences are common. There also exist two reports concerning malignant transformation of fibromatosis into fibrosarcoma [25].

Fibromatosis of the breast is a rare condition. The most numerous groups of patients do not exceed 30 women [26, 27]. All the reported cases refer to women. Pathogenesis remains unknown, however hormonal (pregnancy) genetic (Gardner's syndrome) or traumatic factors are often blamed [25]. Lesions have been described to appear in the direct vicinity a of saline-filled implant or in the site of fibroadenoma removed one year earlier. Bilateral tumours have been reported. The largest tumour was 10 cm in size [27].

On histopathological examination, one finds policellular interstitium, which dominates over the lobular and ductal components. It contains myofibroblasts, which form fasciculi or other forms divided by collagen fibres. These cells appear in different shapes and sizes, but seldom are atypic. On immunohistochemical examination, the reactions to smooth muscle actin and vimentin are positive, and the reaction to desmin negative.

The recommended treatment is local resection with a wide healthy tissue margin. Local recurrence is very common, due to the very long and often indiscernible outgrowing processes, which infiltrate local tissues. Wargotz et al. have described a group of 20 patients treated with local resection of the tumour. Five of them presented with local recurrence, usually within a couple of months after surgery. In one case, recurrence was observed 6 years after surgery. No patient died due to the disease and in no case did metastases occur [26]. In another group of patients [27], recurrence was reported in 27% of the cases, usually over an average period of 30 months after surgery. Usually local recurrence was treated by re-resection with a wide margin, however simple mastectomies were performed in case of excessive recurrence or in case of poor anatomical conditions. In these latter cases reconstruction was suggested approximately 3 years after surgery, in order to minimalise the risk of next recurrence [28]. Neither the tumour size, nor any analysed histological feature did appear to influence the possibility of recurrence [26].

At the Chair and Clinic of Oncology of the Poznań University of Medical Sciences we have diagnosed a case of fibromatosis infiltrativa mammae in a 74-year old patient. The lesion was found in a mammography scan. On palpation, we found a lesion 20 mm in diameter, localised between the two superior quadrants of the right breast. The patient was referred for surgery with the initial diagnosis of breast cancer (basing upon clinical examination and mammography). On excision, we found a poorly delimited, star-shaped lesion, approx. 1 cm in diameter. Peripherally, the lesion infiltrated the local adipose tissues with long ramifications. During intraoperative histopathological examination, the tumour was found to contain proliferating, fusiform mesenchymal cells, while the final histopathological examination provided diagnosis of fibromatosis infiltriva mammae.

The patient remains under close follow-up scrutiny. Clinical examination and mammography performed one

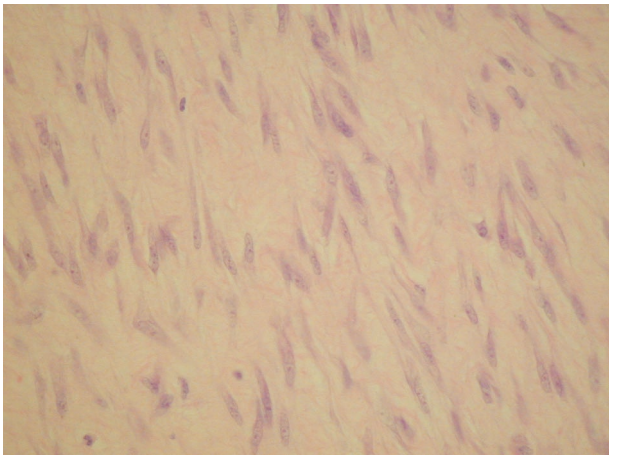

Figure 9. Fibromatosis infiltrativa mammae H&E staining; enlarged 400X

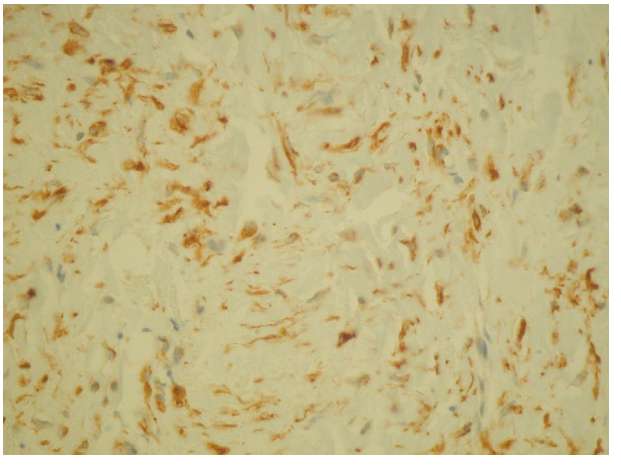

Figure 10. Fibromatosis infiltrativa mammae, positive reaction to vimentin (immunohistochemical analysis); enlarged 400X

year after the surgical procedure did not reveal any signs of recurrence.

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