THE JOURNAL OF
MEDICINE, LAW & PUBLIC HEALTH

Vol 6, No 2. 2026
Reviews

Male Breast Adenoid Cystic Carcinoma – A Narrative Review of a Rare Disorder

Sajad Ahmad Salati[1*]

[1]*College of Medicine, Qassim University, Saudia Arabia*

Summary

Background: Adenoid cystic carcinoma (ACC) is primarily a disease of the salivary glands and is exceptionally rare in the male breast, representing less than 0.1% of all male breast cancers. Due to its rarity, there is no established consensus or recognized guidelines for management. This review aims to synthesize existing literature to improve clinical awareness and knowledge of male breast ACC (MB-ACC).

Methods: A comprehensive literature search was conducted across PubMed/MEDLINE, Scopus, and Google Scholar for cases of MB-ACC published between 1969 and 2025. Inclusion criteria focused on peer-reviewed case reports providing clinical, histopathological, and immunohistochemical data. Approximately 20 cases were identified and analyzed.

Results: MB-ACC often presents as a slow-growing, subareolar mass, with a median age of onset (41.5 years) younger than typical male breast cancer. Diagnosis is confirmed via histopathology showing cribriform, tubular, or solid patterns with a double-layered cell structure. Most tumors are triple-negative (ER-/PR-/HER2-), though they paradoxically exhibit an indolent clinical course and favorable long-term prognosis compared with other triple-negative cancers. Management is primarily surgical, with modified radical mastectomy being the most frequently reported procedure.

Conclusion: Although MB-ACC shares histological features with aggressive basal-like carcinomas, it carries a favorable prognosis. Early diagnosis through awareness is essential to avoid advanced presentation. Given the potential for late recurrence, long-term follow-up is recommended.

Keywords: Adenoid Cystic Carcinoma, Immunohistochemistry, Male Breast Neoplasms, Modified Radical Mastectomy, Rare Diseases, Triple Negative Breast Neoplasms.

Received: 2025-10-18 | Accepted: 2026-01-02 | Published: 2026-03-17

DOI: 10.52609/jmlph.v6i2.274

*Corresponding author: docsajad@gmail.com

INTRODUCTION

Male breast cancer (MBC) accounts for less than 1% of all breast cancer diagnoses worldwide, making it a rare disease in comparison with female breast cancer [1, 2]. According to histopathology, invasive ductal carcinoma of no special type (NST) accounts for most of MBC (75%–90%), with tumours frequently being luminal A or B molecular subtypes [3]. Papillary carcinoma and, rarely, invasive lobular carcinoma are other, less prevalent variants.

Male breast adenoid cystic carcinoma (MB-ACC) is an exceptionally rare type of MBC [4]. It is a distinct cancer with clinicopathological characteristics that set it apart from other types of MBC, but due to its extreme rarity, the literature is meagre. Adenoid cystic carcinoma (ACC) was first described by Robin and Laboulbene in 1853 as "*tumeur heteradenique*", and Geschikter in 1945 was the first to use the term "ACC" in breast oncology [4]. Ferlito [5] was the first to report MB-ACC in 1969, and about 20 cases have been reported in peer-reviewed literature since then.

Due to the scarcity of literature, the diagnostic and therapeutic recommendations for MB-ACC have yet to be determined; currently, both diagnostic and therapeutic modalities rely on guidelines developed for female patients [4]. The purpose of this narrative review is to highlight the pathophysiology, clinical characteristics, and existing treatment options to increase healthcare providers' awareness and add to the existing body of information.

METHODOLOGY

Search Strategy and Data Sources

To identify all documented cases of primary male breast adenoid cystic carcinoma (MB-ACC), a systematic literature search was conducted between August and December 2025. The search encompassed the PubMed/MEDLINE, Scopus, and Google Scholar databases, covering the period from 1969 (the date of the first reported case) through December 2025. The search strategy utilized a combination of Medical Subject Headings (MeSH) and free-text keywords, including: "male breast cancer," "adenoid cystic carcinoma," "MB-ACC," and "primary breast adenoid cystic carcinoma."

Study Selection and Systematic Filtering

Following the initial identification of records, titles and abstracts were screened for relevance. Full-text articles were then retrieved and evaluated against strict inclusion and exclusion criteria to ensure diagnostic and clinical accuracy.

Studies were included if they were peer-reviewed case reports, case series, or observational studies involving male patients with a confirmed diagnosis of primary adenoid cystic carcinoma (ACC) of the breast, and if they provided sufficiently granular clinical, histopathological, or immunohistochemical (IHC) data.

Studies were excluded where anatomical specificity was lacking, such as cases in which ACC originated from extra-mammary sites (for example, salivary glands, lung, or skin) with secondary metastasis to the breast. We also excluded abstracts, letters, and conference proceedings that did not provide definitive pathological confirmation—such as clear descriptions of cribriform, tubular, or solid growth patterns—or relevant IHC markers (for example, CD117/c-KIT and p63). Studies focused exclusively on female cohorts, or those in which male-specific data could not be separated, were likewise excluded. To avoid double-counting rare cases, potentially overlapping reports from the same institution or author were cross-referenced using patient age and clinical history, and only the most comprehensive or most up-to-date report was retained.

Data Extraction and Synthesis

Data were systematically extracted regarding patient demographics, clinical presentation (e.g., mass location, duration of symptoms), imaging characteristics, surgical interventions, adjuvant treatments (radiotherapy/chemotherapy), and long-term oncological outcomes. Given the rarity

of MB-ACC, the data were qualitatively synthesized to provide a comprehensive overview of the current diagnostic and therapeutic landscape.

AETIOLOGY

The exact aetiology of male breast adenoid cystic carcinoma (MB-ACC) remains largely elusive. In general, male breast cancer risk factors such a BRCA2 mutations, Klinefelter syndrome, and hyperestrogenism are well-recognized [6]. However, these factors have not been consistently linked to the ACC subtype in men, which often lacks the strong hormonal association seen in ductal MBC.

Unlike the more common hormone-receptor-positive MBCs, MB-ACC typically presents a triple-negative phenotype. At the molecular level, the MYB-NFIB gene fusion, resulting from the t(6;9)(q22-23;p23-24) translocation, is considered a hallmark of salivary gland ACC and has been confirmed in cases of female breast ACC [7, 8]. The genomic data for male patients is still very limited; however, this genetic rearrangement is hypothesised to be a key driver of MB-ACC. It contributes to the distinct "triple-negative paradox," where aggressive markers are present alongside an indolent clinical course [9].

In the end, a significant information gap in the literature is the absence of extensive genetic research in MB-ACC. To ascertain whether the aetiological causes of MB-ACC are similar to those of its salivary gland counterparts or if distinct male-specific variables exist, more research employing next-generation sequencing (NGS) is required.

EPIDEMIOLOGY

The incidence of MB-ACC is very low, with fewer than 20 cases reported since its first description by Ferlito in 1969 [5]. Of these, about half of the cases were reported in the past decade, which may indicate a trend toward increased incidence [4]. The susceptibility of various racial subpopulations to MB-ACC is definitely not known, although 75% of the cases reported in the past three decades were reported in Asia [4]. Regarding the female variant of breast ACC, a population-based cohort study conducted in the USA by Ghabach et al. [9] indicated that black females have a lower rate of incidence than their white counterparts.

The age of onset for MB-ACC is lower (median 41.5 years) than that seen in other male breast cancer patients, and many cases have been reported in patients in their teens and twenties. This contrasts with female breast ACC patients, where the median age of onset is 58-66 years old [4].

CLINICAL RESULTS

Common Clinical Patterns

The most consistent presentation of MB-ACC is a slow-growing, hard, palpable subareolar lump that may be accompanied by pain and/or tenderness [10-12]. The tumour mass is solitary in most cases, and the mean tumour size in the reported cases ranges between 1.2 and 5 cm (the median is 2.6 cm). There is no significant difference in incidence between the left and right breasts [4]. Less common symptoms include skin ulceration, discharge, nipple retraction, and distant metastasis. Delays in seeking medical treatment may result from the lump's extremely slow growth [11]. Li et al. [4] reported a case of MB-ACC in a 24-year-old male who presented with a 1 cm left subareolar lump that had been growing slowly for 5 years. Similarly, Wan et al. [12] described a 38-year-old male who experienced nipple retraction with minimal pain and redness in 2013, but chose not to seek medical help. The condition worsened over time until he presented to the hospital in 2015.

Clinical Contraindications and Divergence

While the literature generally characterises MB-ACC as an "indolent" disease, individual case reports present sharp contradictions regarding its biological behaviour.

Most patients demonstrate an indolent course and can remain asymptomatic for years [4,12].

However, Yoo et al. reported a strikingly aggressive outlier: a 41-year-old patient who presented with advanced distant metastases to the lungs, bone marrow, and axial skeleton [13].

Although axillary lymphadenopathy is generally rare in male breast ACC (MB-ACC), contradictory findings have been described. For example, Li et al. reported postoperative PET/CT evidence of metabolically active lymph nodes in the neck and bilateral axillae [4].

Clinically, most cases present as a simple breast mass; nevertheless, unusual presentations have been reported, particularly among elderly patients. These include aggressive ulcero-proliferative lesions and persistent mucinous discharge, which can mimic more typical invasive breast carcinomas [14].

INVESTIGATIONS (IMAGING)

Mammography: MB-ACC may appear as a lobulated or circumscribed mass in the subareolar area, sometimes with unclear margins or spiculations, thereby resembling other benign or malignant tumours [10].

Ultrasonography: Ultrasonography (USG) is typically used to evaluate the mass, but findings are often non-specific, including clear or indistinct boundaries, irregular shapes, and hypoechoic and heterogeneous masses [4,10].

In the case reported by Tang et al. [10], USG revealed an irregular, mixed echoic, partially compressible mass (21 mm × 20 mm × 9 mm) in the subareolar region of the right breast. Ultrasonography in a case presented by Pang et al. [15] revealed an irregular hypoechoic lesion (12.8 mm × 9.9 mm ×6.7 mm) with an unclear boundary and minimal vascularity. The lesion had an attachment to the nipple and did not display any posterior acoustic shadowing or echogenic halo. In the case presented by Li et al. [4], serial ultrasonography revealed a well-defined oval hypoechoic mass in the retroareolar region of the left breast that progressively enlarged over the course of 2 years (Figure 1).

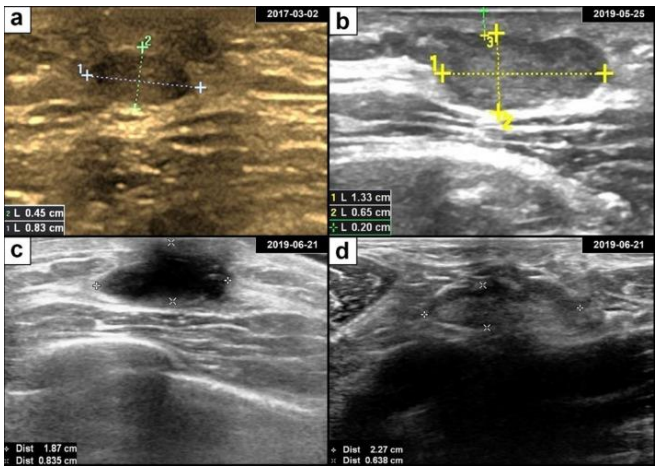

Figure 1. Breast adenoid cystic carcinoma ultrasound and postoperative findings. (a) Ultrasonography revealed a well-defined oval hypoechoic mass in the retroareolar region of the left breast. (b) The subareolar mass progressively enlarged over the course of 2 years. (c) Ultrasonography showed a hypoechoic area after lumpectomy. (d) Ultrasonography of an enlarged axillary lymph node with a clear boundary. Image source: Li et al. (4) doi: 10.14740/jmc3790; reused under the terms of the Creative Commons Attribution Non-Commercial 4.0 International License.

***Computed tomography (CT) scan*:** By displaying the size, location, and possible dissemination to bone or lymph nodes, a CT scan can aid in the diagnosis of MB-ACC. Features such as osteolytic (bone-destroying) lesions, an uncommon but typical pattern of bone metastases for this cancer type, can be seen on the scan [10].

***Magnetic resonance imaging (MRI)*:** MRI findings for MB-ACC are not specific enough for a definitive diagnosis but can provide valuable information that can aid in management, including a characteristic enhancement pattern with gadolinium, T2 signal, and potential internal structures.

Pang et al. [15], in 2019, were the first to use this modality to evaluate MB-ACC when they demonstrated its appearance as a solitary oval subareolar nodule with a spiculated margin and invasion of the nipple. The lesion displayed low signal intensity on T1WI and high signal intensity on T2WI. The time-signal intensity curve demonstrated plateau enhancement kinetics, and

tumour enhancement was rapid and homogeneous on dynamic contrast-enhanced scanning MRI.

***Positron emission tomography (PET)*:** The local extent of MB-ACC and its distant spread can be found using PET. Li et al. [4] re-evaluated their operated case when a biopsy revealed the diagnosis of MB-ACC and, as depicted in Figure 2, the ^{18}F-FDG PET-CT (Fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography) revealed a disordered subcutaneous structure in the left breast region accompanied by increased metabolism, indicating postoperative changes. No significant metabolic abnormalities were observed anywhere else, except in a few small lymph nodes in the neck and both the axillae.

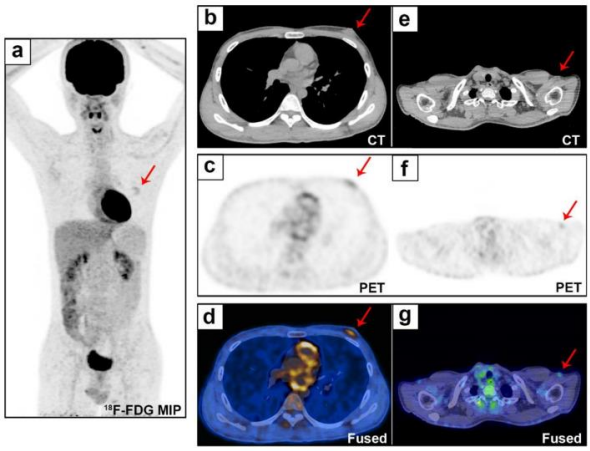

Figure 2. Whole-body evaluation by ^{18}F-FDG PET/CT. The ^{18}F-FDG PET/CT MIP (a) shows uptake (SUVmax 2.5) in the left breast lesion (b and d, red arrows), with a disordered tissue structure (c, red arrow). Small lymph nodes were found in the bilateral neck and bilateral axillary regions (e-g, red arrows, SUVmax 1.65). ^{18}F-FDG PET/CT: ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography; MIP: maximum intensity projection; SUVmax: maximum standardized uptake value. Image source: Li et al. (4) doi: 10.14740/jmc3790; reused under the terms of the Creative Commons Attribution Non-Commercial 4.0 International License.

HISTOLOGICAL CHARACTERISTICS

Histopathological examinations are essential to confirm the diagnosis of MB-ACC (Figure 3). Morphologically, MB-ACC and salivary gland ACC are similar. Microscopic examination of the excised tissue usually reveals diffuse tumour invasion of the subcutaneous tissue, with cells arranged in three microscopic growth patterns, either alone or in combination—cribriform, tubular, and solid [16]. Occasionally, microcystic patterns may be present.

Within each tumour pattern, double-layered structures comprising inner glandular epithelial and outer myoepithelial/basal cells can be found. The glandular epithelial cells exhibit a cuboidal shape, acidophilic cytoplasm, round nucleus, and occasional nucleoli, whereas the myoepithelial/basal cells exhibit a spindle shape, low cytoplasmic volume, intense nuclear staining, and sporadic mitosis [12]. However, unlike salivary gland ACC, MB-ACC uncommonly features nerve infiltration [4].

Ro et al. [17] have proposed a classification of breast ACC (either gender) based on the proportion of solid components within the lesion. Accordingly, there are three grades: grade I with no solid growth, grade II with solid growth less than 30%, and grade III with solid growth more than 30%. They have further observed that grade II and III tumours tend to be larger in size and more prone to relapse.

IMMUNOHISTOCHEMICAL CHARACTERISTICS

The immunohistochemical characteristics of MB-ACC resemble those of basal-like carcinoma (Figure 3). The outer myoepithelial cells are usually positive for 34βE12, smooth muscle actin (SMA), calponin, vimentin, S-100, and P63 markers, whereas the inner glandular epithelial cells often express CD117 (C-kit), epithelial membrane antigen (EMA), CK5/6, CK8/18, and CK14, but mostly do not express oestrogen receptors (ER), progesterone receptors (PR), and human epidermal growth factor receptor 2 (HER-2), though some cases with weak ER positivity have been reported [8]. The basement membrane is highlighted by collagen type IV staining.

MOLECULAR GENETIC CHARACTERISTICS

MYB is a proto-oncogene located in 6q22-23 and has been documented in a variety of malignant lesions. A defining molecular event in ACC is the recurrent reciprocal translocation of t(6;9)(q22-23;p23-24), which results in the MYB-NFIB gene fusion [7-8,18].

Frequency in MB-ACC: The molecular genetic data of MB-ACC are limited. Among the cases reported in the literature, only three were reported to have undergone MYB gene testing, and MYB-NFIB gene fusion was detected in only one case [12].

Diagnostic significance: The MYB-NFIB fusion has high diagnostic significance. It serves as a specific biomarker that distinguishes ACC from its mimics, such as solid papillary carcinoma or cribriform ductal carcinoma in situ (DCIS). While MYB protein overexpression (detected via IHC) is a sensitive surrogate, the presence of the actual gene fusion via FISH is considered the gold standard for confirming the diagnosis in challenging or high-grade cases [19].

Prognostic significance: To date, the presence of the MYB-NFIB fusion does not have clear prognostic significance in MB-ACC. Studies in both breast and salivary ACC have failed to show a definitive correlation between fusion status and overall survival or recurrence risk [18].

Recommendations for routine testing: Routine molecular testing for the MYB-NFIB fusion is not currently mandatory for all cases, as the diagnosis can often be established through classic histomorphology and a standard IHC panel (p63, CD117, and triple-negative status). However, it can strengthen the diagnosis in cases with an aggressive clinical course, high-grade histological features, or those with a "solid" variant morphology where the differential diagnosis is broader [19].

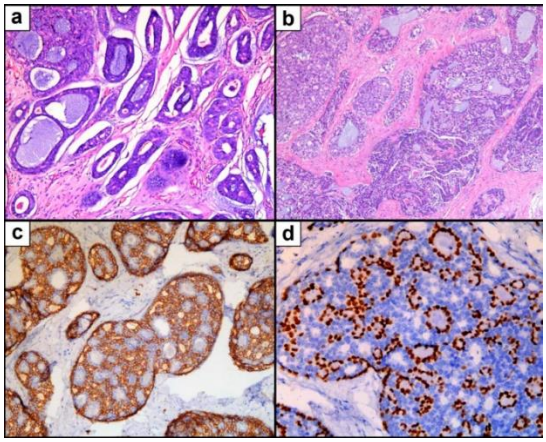

Figure 3. Microscopic pathology of breast adenoid cystic carcinoma. (a) H&E staining showing a cribriform growth pattern. (b) H&E staining showing tubular and solid growth patterns. (c) IHC staining for CD117. (d) IHC staining for P63. H&E: hematoxylin and eosin stain; IHC: immunohistochemistry. Image source: Li et al. (4) doi: 10.14740/jmc3790; reused under the terms of the Creative Commons Attribution Non-Commercial 4.0 International License.

The clinical and pathological characteristics of the identified cases of MB-ACC are summarized in Table 1.

Table 1. Summary of Clinical and Pathological Characteristics of Reported MB-ACC Cases

Characteristic	Summary of Findings in Male Patients
Total Cases	Fewer than 20 cases reported since 1969.
Median Age	41.5 years (Range: 13 to 82)
Common Symptoms	Slow-growing subareolar lump, pain, skin ulceration, nipple retraction.
Median Tumour Size	2.6 cm (Range: 1.2 cm to 5 cm)
Imaging Features	Non-specific; hypoechoic masses on USG; plateau enhancement on MRI.
Histology Patterns	Cribriform (most common), tubular, and solid.
Molecular Subtype	Usually triple-negative (ER-, PR-, HER2-), but CD117+ and p63+.

DIFFERENTIAL DIAGNOSIS

Cribriform ductal carcinoma: Cribriform ductal carcinoma (CDC) has only one type of epithelial cell, and the basement membrane materials or basophilic secretions are absent. Furthermore, myoepithelial/basal cells are arranged successively in a linear pattern around the duct of CDC [4,12].

Invasive cribriform carcinoma: Invasive cribriform carcinoma (ICC) resembles breast ACC morphologically. However, cribriform cell nests of ICC are highly irregular, contain mucoprotein secretions or necrotic tissue, and display an absence of basement membrane material and myoepithelial/basal cells. In addition, immunohistochemical analysis often yields positive results for ER and PR receptors and negative results for p63 tumour markers [12].

Microglandular adenosis: The glands of microglandular adenosis (MA) are covered with only a single layer of glandular epithelium without any myoepithelial/basal cells, and the lumens contain acidophilic secretions. In atypical cases with carcinogenesis, there is a fusion of glandular structures, thereby exhibiting solid growth with high nuclear grades and significant mitosis, which is not found in MB-ACC. The immunohistochemical p63 negativity of MA also aids differentiation from MB-ACC [12].

Acinic cell carcinoma: Cells in acinic cell carcinoma (ACC) contain an abundance of coarse, red-stained granules in the cytoplasm, irregularly shaped nuclei, and distinct cell boundaries, with an absence of myoepithelial/basal differentiation [4,12].

MANAGEMENT

Currently, there are no management guidelines for MB-ACC due to its rarity, and treatment modalities are based largely on those for female patients. These modalities (Table 2) include the following:

Surgery: This is the mainstay of treatment for MB-ACC. Modified radical mastectomy (MRM) is the most common surgical approach [10,12]. While breast-conserving surgery (BCS) is a viable and frequent option for female patients with ACC, it is associated with an unacceptably high local recurrence rate in males [12].

The evidence-based rationale for this is two-fold. First, there are important anatomical constraints. Unlike the female breast, the male breast contains only a rudimentary amount of glandular tissue and lacks Cooper's ligaments. This limited volume means that a tumour of even moderate size often lies in close proximity to the nipple–areolar complex and the pectoralis fascia. As a result, achieving a wide (1–2 cm) tumour-free margin—critical in adenoid cystic carcinoma (ACC) to reduce the risk of local relapse—is technically difficult without performing a total or modified radical mastectomy [20].

Second, the biology of ACC further supports a more extensive surgical approach. ACC is recognised for its infiltrative growth pattern and its potential for perineural invasion, which may extend beyond the palpable mass [12]. In the thin male chest wall, these microscopic extensions are more likely to reach the skin or underlying muscle. Accordingly, studies have reported that when lumpectomy is attempted in men, positive or close margins are frequently encountered [20]. Consequently, MRM remains the gold standard for MB-ACC to ensure oncological safety by providing a definitive clear margin in a restricted anatomical space.

***Axillary lymph node management*:** Routine axillary lymph node dissection is generally not necessary due to the low rate of lymph node metastasis. In the case presented by Tang et al. [10], 41 axillary lymph nodes were dissected, and all were negative for malignancy. The value of sentinel lymph node excision (SLNE) is not clearly known; researchers including Pang et al. [15], have proposed routine SLNE rather than axillary node dissection.

Adjuvant therapies: The role of other therapies is not well-defined [10]. Hormonal treatment and

anti-HER2 treatment are rarely used in MB-ACC, because hormone receptors and HER2/neu expression are usually absent [10]. Systemic chemotherapy is also typically reserved for high-grade tumours or those with lymph node or distant metastases. Radiotherapy may be recommended for positive surgical margins after breast-conserving surgery [4,13].

Table 2. Comparison of Outcomes by Treatment Approach in MB-ACC

Treatment Approach	Common Indications	Observed Clinical Outcomes
Modified Radical Mastectomy (MRM)	Standard primary treatment for most cases.	Associated with the lowest reported local recurrence rates due to achieving wider negative margins in male breast tissue.
Breast-Conserving Surgery (BCS)	Occasionally attempted for very small, peripheral tumours.	Linked to an unacceptably high rate of local recurrence in men, often requiring subsequent salvage mastectomy.
Sentinel Lymph Node Excision (SLNE)	Clinically N0 (node-negative) axilla.	Provides accurate staging with minimal morbidity; outcomes are excellent given the naturally low rate of nodal metastasis in ACC.
Axillary Lymph Node Dissection (ALND)	Clinically positive nodes or high-grade (Grade III) solid tumours.	No documented survival advantage over SLNE for node-negative disease; typically yields negative results even in large tumours.
Adjuvant Radiotherapy	Positive or close surgical margins or following BCS.	Improves local control in the setting of conservative surgery but is generally unnecessary following a successful MRM with clear margins.
Systemic Therapy (Chemo/Hormonal)	High-grade tumours or documented distant metastasis.	Limited impact on survival or recurrence due to the typical triple-negative status and indolent nature of ACC.

PROGNOSIS

The specific prognosis for male breast adenoid cystic carcinoma (MB-ACC) is distinct from other forms of male breast cancer (MBC). Despite often presenting with a triple-negative phenotype (ER-/PR-/HER2-), MB-ACC is characterised by a paradoxically indolent clinical course and high long-term survival rates. While common triple-negative MBCs (invasive ductal carcinoma, NST) are typically aggressive, MB-ACC demonstrates a much more favourable outlook.

Several clinical and histological factors serve as predictors of recurrence or metastasis. The histological grade, determined by the proportion of solid growth as described by Ro et al. [17], is a primary determinant of outcome.

-Grade I tumours (no solid growth) follow a highly indolent path with minimal risk of distant spread.

-Grade II and III tumours (containing solid components) are associated with larger tumour sizes and a significantly higher propensity for local relapse and distant metastasis, particularly to the lungs and bone.

Furthermore, specific oncological markers such as positive surgical margins, Nottingham grade, and neovascularisation are closely associated with the recurrence and distant metastasis of these tumours [12]. In the male patient, achieving wide negative margins is particularly challenging due to the lack of substantial breast tissue, making margin status a vital prognostic indicator. Additional predictors of poor outcomes include a larger tumour size at presentation (median 2.6 cm) and the presence of axillary lymphadenopathy, although lymph node involvement remains rare in this disease [10,14].

In the review of 15 cases where details of follow-up were mentioned, Pang et al. [15] found that 10

cases (66.7%) were asymptomatic at 4-82 months (mean 35.7 months), whereas 3 cases (20%) suffered from lung and/or bone metastases and 2 patients (13.3%) had local recurrences after 12-84 months.

Because of the documented risk of late recurrence, which can occur many years after the initial diagnosis, early detection and lifelong surveillance are critical. Lack of awareness can lead to delayed diagnosis and more advanced disease at presentation, potentially compromising the otherwise excellent survival profile of this rare malignancy.

PSYCHOSOCIAL CONSIDERATIONS

The psychosocial impact on patients, particularly younger men, can be substantial even if MB-ACC has a generally good clinical prognosis. However, because there are so few cases, this aspect has not been thoroughly investigated. However, it may be inferred from data from other research pertaining to various breast ailments that a diagnosis of a "female-gendered" disease frequently results in significant psychological suffering, including shock, social isolation, and stigma connected to gender identity [21].

For young men, undergoing a modified radical mastectomy can result in substantial body image disturbances and a sense of "physical incompleteness." Unlike female patients, for whom breast reconstruction is a standard part of the care pathway, male patients are less frequently offered or aware of chest wall contouring options, which can impact long-term quality of life (QoL). Healthcare providers must recognise that the indolent nature of the tumour does not diminish the patient's fear of recurrence, which remains a persistent psychological burden. To lessen these consequences, a multidisciplinary approach is suggested to MB-ACC that includes early referral to psycho-oncology treatments and male-specific support groups [22].

HEALTHCARE RESOURCE UTILIZATION

The management of MB-ACC entails significant healthcare resource utilisation. Because imaging is non-specific, patients often require advanced diagnostic pathways, including ^{18}F-FDG PET/CT to evaluate local and distant spread. Furthermore, the requirement for an extensive immunohistochemical panel—including CD117, p63, and CK markers —to differentiate ACC from mimics such as invasive cribriform carcinoma adds to the economic burden, particularly in resource-limited settings.

FUTURE DIRECTIONS AND RESEARCH PRIORITIES

To move beyond the current reliance on case reports, the following research priorities are proposed:

Molecular profiling: Future research should focus on the prevalence of the MYB-NFIB gene fusion specifically in male cohorts to determine whether it can serve as a definitive diagnostic biomarker or a target for emerging MYB-degraders currently in Phase I/II trials.

***International registries*:** Given the rarity of MB-ACC, the establishment of a global "Rare Breast Tumor Registry" is essential to aggregate data on long-term survival and the efficacy of breast-conserving surgery versus mastectomy.

Clinical trial integration: While primary MB-ACC trials are unfeasible, patients with recurrent or metastatic disease should be prioritized for trials investigating Notch pathway inhibitors or targeted tyrosine kinase inhibitors, which have shown promise in salivary gland ACC.

***Patient-reported outcome measures (PROMs)*:** Future studies must formally incorporate validated QoL instruments to assess the long-term functional and aesthetic outcomes of surgical interventions in men.

CONCLUSIONS

Male breast adenoid cystic carcinoma (MB-ACC) represents a rare clinical paradox, characterised by a triple-negative phenotype that behaves with a remarkably indolent clinical course. While the long-term prognosis remains favourable compared with other male breast malignancies, management is complicated by a lack of

standardised protocols and the absence of large-scale genomic data.

Histological grade is the main factor influencing clinical outcomes, and solid growth patterns are important indicators of recurrence. A comprehensive, long-term surveillance program is essential due to the ongoing risk of late recurrence. Furthermore, a holistic approach to care is necessary to address the significant psychosocial impact of mastectomy and the high healthcare resource utilisation required for a definitive diagnosis.

Ultimately, the rarity of MB-ACC remains a significant barrier to evidence-based practice. Future efforts must prioritise the establishment of international registries and the exploration of molecular drivers to move toward more personalised and targeted therapeutic strategies for this unique patient population.

ABBREVIATIONS

MB-ACC: Male breast adenoid cystic carcinoma
MBC: Male breast cancer
ACC: Adenoid cystic carcinoma
SLNE: Sentinel lymph node excision
CDC: Cribriform ductal carcinoma
ICC: Invasive cribriform carcinoma
ER: Estrogen receptors
PR: Progesterone receptors
HER-2: Human epidermal growth factor receptor-2
USG: Ultrasonography
CT: Computed tomography
PROMs: Patient-reported outcome measures

REFERENCES

1. Varzaru VB, Anastasiu-Popov DM, Eftenoiu AE, Popescu R, Vlad DC, Vlad CS, et al. Observational study of men and women with breast cancer in terms of overall survival. *Cancers* (Basel). 2024 Sep 1;16(17):3049. doi: 10.3390/cancers16173049.
2. Zhao L, Cheng H, He D, Zhang Y, Chai Y, Song A, et al. Decoding male breast cancer: epidemiological insights, cutting-edge treatments, and future perspectives. *Discov Oncol.* 2025 Mar 19;16(1):360. doi: 10.1007/s12672-025-02140-y.
3. Sang G, Pan H, Lu C, Sun R, Zha X, Wang S, et al. Clinical features and prognostic factors of male breast cancer vs. female breast cancer. *Transl Cancer Res.* 2021 May;10(5):2199-2209. doi: 10.21037/tcr-21-1.
4. Li JX, Zhang XM, Xiao YX, Tang ZM, Huang T, Ming J. Male adenoid cystic carcinoma of the breast. *J Med Cases*. 2021 Dec;12(12):503-510. doi: 10.14740/jmc3790
5. Ferlito A. Adenoid cystic carcinoma of the male breast. *Riv Anat Patol Oncol.* 1969; 35:441-455.
6. Brinton LA, Carreon JD, Gierach GL, McGlynn KA, Gridley G. Etiologic factors for male breast cancer in the U.S. Veterans Affairs medical care system database. *Breast Cancer Res Treat.* 2010 Jan;119(1):185-192. doi: 10.1007/s10549-009-0379-0.
7. Persson M, Andrén Y, Mark J, Horlings HM, Persson F, Stenman G. Recurrent fusion of MYB and NFIB transcription factor genes in carcinomas of the breast and head and neck. *Proc Natl Acad Sci U S A.* 2009 Nov 3;106(44):18740-18744. doi: 10.1073/pnas.0909114106.
8. Wetterskog D, Lopez-Garcia MA, Lambros MB, A'Hern R, Geyer FC, Milanezi F, et al. Adenoid cystic carcinomas constitute a genomically distinct subgroup of triple-negative and basal-like breast cancers. *J Pathol*. 2012 Jan;226(1):84-96. doi: 10.1002/path.2974.
9. Ghabach B, Anderson WF, Curtis RE, Huycke MM, Lavigne JA, Dores GM. Adenoid cystic carcinoma of the breast in the United States (1977 to 2006): a population-based cohort study. *Breast Cancer Res*. 2010;12(4): R54. doi: 10.1186/bcr2613
10. Tang P, Yang S, Zhong X, Yao J, Zhang Y, Dong H, et al. Breast adenoid cystic carcinoma in a 19-year-old man: a case report and review of the literature. *World J Surg Oncol*. 2015 Feb 6; 13:19. doi: 10.1186/s12957-015-0442-8.
11. Liu J, Jia W, Zeng Y, Deng H, Rao N, Su F. Adolescent male adenoid cystic breast

carcinoma. *Am Surg*. 2012 May;78(5): E288-289.
12. Wan D, Zhou H, Zhang Y. Adenoid cystic carcinoma of the breast in a male patient: a case report and literature review. *Front Oncol*. 2022 Jul 7; 12:905997. doi: 10.3389/fonc.2022.905997.
13. Yoo SJ, Lee DS, Oh HS, Kim HJ, Kim MH, Ahn YC, et al. Male breast adenoid cystic carcinoma. *Case Rep Oncol.* 2013 Oct 12;6(3):514-519. doi: 10.1159/000356062.
14. Kshirsagar AY, Wader JV, Langade YB, Jadhav KP, Zaware SU, Shekhar N. Adenoid cystic carcinoma of the male breast. *Int Surg.* 2006 Jul-Aug;91(4):234-236.
15. Pang W, Wang Z, Jin X, Zhang Q. Adenoid cystic carcinoma of the breast in a male: A case report. *Medicine* (Baltimore). 2019 Aug;98(32): e16760. doi: 10.1097/MD.0000000000016760.
16. Miyai K, Schwartz MR, Divatia MK, Anton RC, Park YW, Ayala AG, et al. Adenoid cystic carcinoma of breast: Recent advances. *World J Clin Cases*. 2014;2(12):732–741. doi: 10.12998/wjcc.v2.i12.732.
17. Ro JY, Silva EG, Gallager HS. Adenoid cystic carcinoma of the breast. *Hum Pathol*. 1987;18(12):1276–1281. doi: 10.1016/S0046-8177(87)80413-6.
18. D'Alfonso TM, Mosquera JM, MacDonald TY, Padilla J, Liu YF, Rubin MA, et al. MYB-NFIB gene fusion in adenoid cystic carcinoma of the breast with special focus paid to the solid variant with basaloid features. *Hum Pathol*. 2014 Nov;45(11):2270-2280. doi: 10.1016/j.humpath.2014.07.013.
19. Lataifeh H, Abuhijla F, Wahbeh L, Abuhijlih R, Asha W, Alzibdeh A, et al. Adenoid cystic carcinoma of the breast with aggressive behavior: A case report and review of the literature. *Oncol Lett.* 2025 Sep 4;30(5):510. doi: 10.3892/ol.2025.15256.
20. Maciag H, Ziolkowski P, Wrzecion S, Kolodziej P. Adenoid cystic carcinoma of the breast in an 80-year-old male - a case report. *Wspolczesna Onkologia*. 2006;10(7):340
21. Ruddy KJ, Giobbie-Hurder A, Giordano SH, Goldfarb S, Kereakoglow S, Winer EP, et al. Quality of life and symptoms in male breast cancer survivors. *Breast.* 2013 Apr;22(2):197-199. doi: 10.1016/j.breast.2012.12.014.
22. Raeisi Dehkordi S, Kennedy SKF, Peera M, Wong HCY, Lee SF, Alkhaifi M, et al. Examining stigma in experiences of male breast cancer patients and its impact as a barrier to care: a narrative review. *Ann Palliat Med*. 2024 Sep;13(5):1291-1300. doi: 10.21037/apm-24-67.