The Professional Medical Journal
www.theprofesional.com

DOI: 10.29309/TPMJ/2020.27.07.4113

CASE REPORT
PROF-0-4113

ADENOID CYSTIC CARCINOMA OF BREAST: A CASE REPORT IN BREAST CLINIC AT A TERTIARY CARE HOSPITAL.

Ramlah Ghazanfor[1], Maham Tariq[2], Mehwish Changeez[3], Sara Malik[4], Ghulam Khadija[5], Hina Hanif[6], Javaria Malik[7]

1. FCPS (Gen. Surgery)
Senior Registrar
Surgical Unit 1, Holy Family Hospital.
2. FCPS (Gen. Surgery)
Postgraduate Trainee
General Surgery,
Holy Family Hospital.
3. FCPS (Gen. Surgery)
Postgraduate Trainee
General Surgery
Holy Family Hospital
4. FCPS (Gen. Surgery)
Postgraduate Trainee
General Surgery
Holy Family Hospital
5. FCPS (Gen. Surgery)
Postgraduate Trainee
General Surgery
Holy Family Hospital
6. FCPS (Radiology)
Assistant Professor Radiology
Holy Family Hospital
7. FCPS (Gen. Medicine)
Senior Registrar Medicine
Holy Family Hospital

Correspondence Address:
Dr. Ramlah Ghazanfor
Senior Registrar Surgical Unit 1,
Holy Family Hospital
ramla.an99@gmail.com

Article received on:
05/09/2019
Accepted for publication:
12/11/2019

ABSTRACT… Incidence of adenoid cystic carcinoma of breast ranges from 0.1% to 1%. Its rare occurrence makes its surgical management options debatable. However rarity of metastasis contributes to an encouraging prognosis inspite of its triple negative receptor status. Herein, we report the case of a 60 year old woman who presented with a breast lump that turned out to be adenoid cystic carcinoma.

Key words: Metastasis, Receptor Status, Surgical Management.

Article Citation: Ghazanfor R, Tariq M, Changez M, Malik S, Khadija G, Hanif H, Malik J. Adenoid cystic carcinoma of breast: A case report in breast clinic at a Tertiary Care Hospital. Professional Med J 2020; 27(7):1533-1536.
DOI: 10.29309/TPMJ/2020.27.07.4113

INTRODUCTION

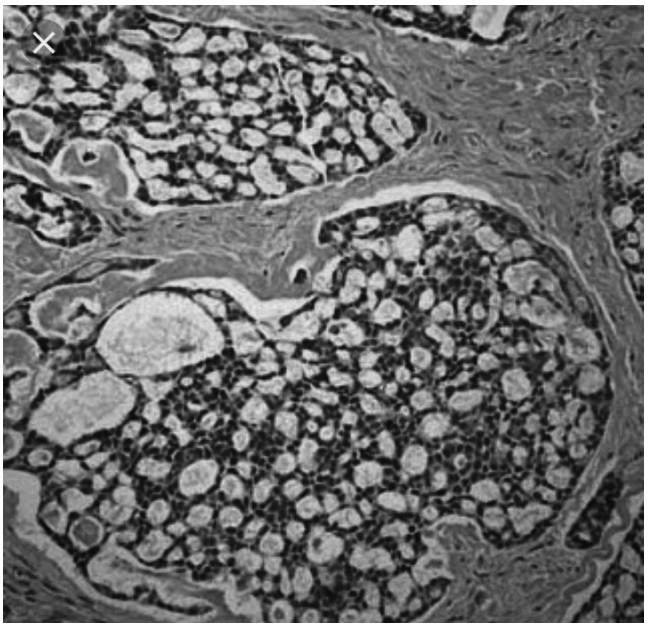
Adenoid cystic carcinoma is a commonly present as a tumor of salivary glands but has been also reported in trachea, nasopharynx, uterus and cervix.[1] In breast it has been reported as a rare occurrence making up to 0.1 to 1% of total reported malignancies. It usually presents as a small slow growing lump during third to fifth decade of life. The incidence after that decreases. Lump is mostly subareolar in position with no clinically obvious axillary or systemic involvement. The Salivary gland tumors are notorious for perineural invasion and local recurrences. However the variant in breast depicts low metastatic potential which explains for the good survival rates of its patients despite its triple negative status.[2]

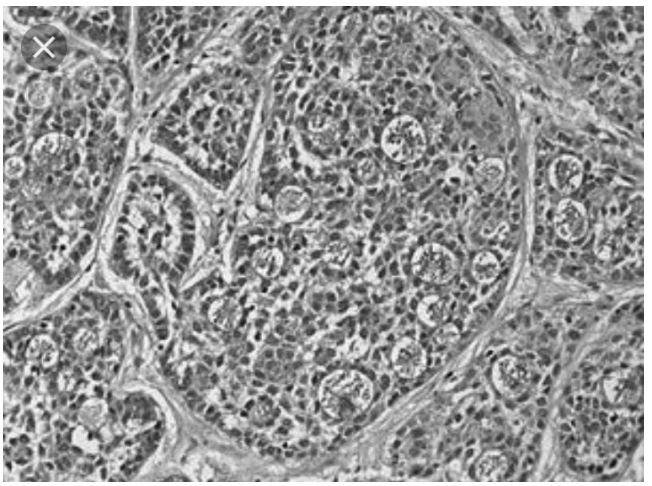
CASE REPORT

A 60-year-old housewife of rural background presented to our breast clinic. She was married and had five kids youngest of whom was 25 years old. She gave history of six-month duration about a small slowly growing lump in left breast just behind nipple. Initially lump was ignored on pretext of being painless. However gradual increase in size along with occasional ache in lump brought her to our Centre. She denied presence of any such swelling in other breast and both armpits. There was no history of nipple discharge or any headache, jaundice, shortness of breath or back ache. She had menarche at 11 years and menopause at 45 years of age. All children were naturally conceived and breast fed. There was no previous history of contraceptive use, hormone replacement therapy, radiotherapy or previous breast ailments. No family history of any breast tumor or other solid organ malignancy was given.

Examination revealed a 2*3 cm firm lump in left breast in retroareloar position extending from 12 to 9 o clock position. It wasn't attached to underlying muscles or skin. Axilla was clinically clear. On X-ray. Mammogram a well-defined soft tissue density lesion with surrounding architectural distortion was noted which was labelled as BIRADS4c. Complementary ultrasound stated it to be a taller than wider lesion. Axilla was clear. Trucut biopsy showed it to be invasive ductal carcinoma and receptor studies showed it to be

triple negative. After normal CT scan she was staged as T2N0MO (stage-1).





Post MDM she was booked for afferent wide local excision and sentinel lymph node biopsy. Post-operative detailed pathology report showed it to be an adenoid cystic carcinoma with clear margins (closest 5 mm away). Sentinel node was found to be uninvolved…MADAM do u intend to write something here? y these dots……. Patient was referred to oncology center for radiotherapy and adjuvant chemotherapy. Hormone therapy was not offered. Follow up at six months and one year showed no local or systemic occurrence of disease.

DISCUSSION

Adenoid cystic carcinoma is a rare variety of breast cancer effecting one out of million women per year (0.1-1% of all breast malignancies).[3] It usually affects menopausal age group with incidence slightly more in black women than in white.[4] Occasional cases in men and children have also been reported(5,6). Risk is highest between third to fifth decade after which it stabilizes.[7] Lump in retro areolar or upper outer quadrant of breast remains most common presenting complaint.[7] Axillary involvement and distant metastasis are rare. However one case of metastatic to Clavicle 13 years after primary surgery has been reported.[8,9] Histologically these tumors resemble their salivary gland counterparts and depict biphasic cellular pattern of myoepithelial as well as epithelial cells (basaloid and ductal components). There can be three varied growth patterns: Glandular, tubular and solid.[10] Three categories are usually defined depending upon the degree of solid element in the tumour. Grade 1 has no solid component while grade three has most. Historically local excision was recommended for grade 1, simple mastectomy for grade 2, and mastectomy and axillary dissection for grade 3. This is now debatable as several studies establish breast conserving surgery comparable to mastectomy in all aspects.[11,12] In the present case, the patient underwent breast conservation surgery and sentinel lymph node biopsy as initial trucut reported lump to be invasive ductal carcinoma of no special type.

Molecular studies show that just like other basal like tumors, ACC of breast is mostly estrogen receptor (ER)-negative and progesterone receptor (PR)-negative and HER-2-neu negative.[13,14] This triple negative biology usually heralds poor prognosis, but several clinical studies confirm ACC to have good survival rates. Absence of perineural advancement, axillary lymh node involvement or distant metastasis are probable reasons behind this good prognosis.[15]

There is a lack of consensus regarding the optimal treatment strategy.[13] Most of the data available is in the form of anecdotal reports or small case series. Historically, mastectomy was preferred due to its almost negligible recurrence rates. Wide local excision on other hand showed

recurrence in 37% cases.[16] However, in later cases, no information about resection margins was provided. Now a day's wide local excision is deemed adequate. However there still remains need for suitable randomized control trials to compare both surgical options.[17]

Axillary lymph node is involved in less than 2% cases. With the mirror trial establishing morbidity of ALND beyond doubt and the Z011 trial questioning its benefits of axillary dissection this matter needs careful deliberation.[18] Current recommendations are to perform axillary dissection only in the presence of nodal metastasis. Sentinel lymph node biopsy can be performed in tumours >3 cm, as it might harbor other invasive types of breast cancer.[19] In our case, a sentinel lymph node dissection was performed, as the tumor was labelled invasive ductal carcinoma on trucut biopsy and diagnosis of a high grade ACC was made in retrospect and a sentinel lymph node was found to be negative.

Radiotherapy following wide local excision is recommended, the Rare Cancer Network (RCN) reported improved local control in ACC patients who received radiotherapy as compared to those who didn't. (5% for the group with post-operative radiotherapy vs. 17% for the group without radiotherapy.[20]

The role of adjuvant chemotherapy therapy and hormonal therapy remains controversial. Absence of metastatic potential and triple negative status apparently preclude the need of both modalities respectively. Several studies assert that both chemo and hormonal therapy donot improve survival rates.[21]

The 5-year survival is reported to be up to 90%, with usually a 100% disease-free survival rate.[22] Late local recurrences or distant metastasis are rare but not unheard off. To date metastasis have been reported in lung, liver, kidneys and brain.[23] These mandate close follow up for long periods of time.

CONCLUSION

Adenoid cystic carcinoma, owing to rarity of metastasis has a good prognosis despite being triple negative. Exact management i.e breast conservation or mastectomy with or without axillary clearance is still under debate and further studies are needed in order to prove the superiority of one technique above the other.

REFERENCES

1. Liu C, Zhu W-D, Chen C. **Adenoid cystic carcinoma of salivary gland with mandible erosion and distant metastases to lung.** QJM Mon J Assoc Physicians. 2019 Aug 8;

2. Wang S, Li W, Wang F, Niu Y, Hao C, Wang X, et al. **36 cases adenoid cystic carcinoma of the breast in China: Comparison with matched grade one invasive ductal carcinoma-not otherwise specified.** Pathol Res Pract. 2017 Apr; 213(4):310–5.

3. Thompson K, Grabowski J, Saltzstein SL, Sadler GR, Blair SL. **Adenoid cystic breast carcinoma: Is axillary staging necessary in all cases? Results from the California Cancer Registry.** Breast J. 2011 Oct; 17(5):485–9.

4. 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 BCR. 2010; 12(4):R54.

5. Hjorth S, Magnusson PH, Blomquist P. **Adenoid cystic carcinoma of the breast. Report of a case in a male and review of the literature.** Acta Chir Scand. 1977; 143(3):155–8.

6. 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.

7. Boujelbene N, Khabir A, Boujelbene N, Jeanneret Sozzi W, Mirimanoff RO, Khanfir K. **Clinical review--breast adenoid cystic carcinoma.** Breast Edinb Scotl. 2012 Apr; 21(2):124–7.

8. Glover TE, Butel R, Bhuller CM, Senior EL. **An unusual presentation of adenoid cystic carcinoma of the breast with metastatic disease in the clavicle. BJR Case Rep [Internet]. 2017 Jan 28 [cited 2019 Aug 11]; 3(2).** Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6159263/

9. 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.

10. Ro JY, Silva EG, Gallager HS. **Adenoid cystic carcinoma of the breast. Hum Pathol.** 1987 Dec; 18(12):1276–81.

11. Cao JQ, Olson RA, Tyldesley SK. **Comparison of recurrence and survival rates after breast-conserving therapy and mastectomy in young women with breast cancer.** Curr Oncol Tor Ont. 2013 Dec; 20(6):e593-601.

12. Kim K, Park HJ, Shin KH, Kim JH, Choi DH, Park W, et al. **Breast Conservation Therapy Versus Mastectomy in Patients with T1-2N1 Triple-Negative Breast Cancer: Pooled Analysis of KROG 14-18 and 14-23.** Cancer Res Treat Off J Korean Cancer Assoc. 2018 Oct; 50(4):1316–23.

13. Thomas DN, Asarian A, Xiao P. **Adenoid cystic carcinoma of the breast.** J Surg Case Rep. 2019 Jan; 2019(1):rjy355.

14. Arpino G, Clark GM, Mohsin S, Bardou VJ, Elledge RM. **Adenoid cystic carcinoma of the breast: Molecular markers, treatment, and clinical outcome.** Cancer. 2002 Apr 15; 94(8):2119–27.

15. Bhutani N, Kajal P, Singla S. **Adenoid cystic carcinoma of the breast: Experience at a tertiary care centre of Northern India.** Int J Surg Case Rep. 2018; 51:204–9.

16. Sumpio BE, Jennings TA, Merino MJ, Sullivan PD. **Adenoid cystic carcinoma of the breast. Data from the Connecticut Tumor Registry and a review of the literature.** Ann Surg. 1987 Mar; 205(3):295–301.

17. Defaud-Hénon F, Tunon-de-Lara C, Fournier M, Marty M, Velasco V, de Mascarel I, et al. **[Adenoid cystic carcinoma of the breast: clinical, histological and immunohistochemical characterization].** Ann Pathol. 2010 Feb; 30(1):7–16.

18. Giuliano AE, Ballman KV, McCall L, Beitsch PD, Brennan MB, Kelemen PR, et al. **Effect of axillary dissection vs no axillary dissection on 10-year overall survival among women with invasive breast cancer and sentinel node metastasis: The ACOSOG Z0011 (Alliance) Randomized Clinical Trial.** JAMA. 2017 12; 318(10):918–26.

19. Canyilmaz E, Uslu GH, Memiş Y, Bahat Z, Yildiz K, Yoney A. **Adenoid cystic carcinoma of the breast: A case report and literature review.** Oncol Lett. 2014 May; 7(5):1599–601.

20. Khanfir K, Kallel A, Villette S, Belkacémi Y, Vautravers C, Nguyen T, et al. **Management of adenoid cystic carcinoma of the breast: A Rare Cancer Network study.** Int J Radiat Oncol Biol Phys. 2012 Apr 1; 82(5):2118–24.

21. El Amarti L, Raiss H, Layachi M, Baldé S, Ettahri H, Elghissassi I, et al. **Breast adenoid cystic carcinoma: A rare case report and review of the literature.** Gulf J Oncolog. 2017 May; 1(24):66–9.

22. Romeira D, Cardoso D, Miranda H, Martins A. **Adenoid cystic carcinoma: Triple negative breast cancer with good prognosis.** BMJ Case Rep. 2016 Feb 3; 2016.

23. Wei L, Liang X, Li S, Liu J. **Adenoid cystic carcinoma of the breast: Report of 25 cases.** Zhonghua Zhong Liu Za Zhi. 2014 Feb; 36(2):147–50.

AUTHORSHIP AND CONTRIBUTION DECLARATION

Sr. #	Author(s) Full Name	Contribution to the paper	Author(s) Signature
1	Ramlah Ghazanfor	1st Author	
2	Maham Tariq	2nd Author	
3	Mehwish Changeez	3rd Author	
4	Sara Malik	4th Author	
5	Ghulam Khadija	5th Author	
6	Hina Hanif	6th Author	
7	Javaria Malik	7th Author	