MedCrave
Step into the World of Research

Journal of Cancer Prevention & Current Research

Case Report

Open Access

CrossMark

Miraculous response to radiotherapy in a rare case of malignant nodular hidradenoma nose

Abstract

Malignant nodular hidradenoma is very rare adnexal tumor with exceedingly low incidence of 0.001%. The biological behavior of the tumor is aggressive, with local recurrences reported in more than 50% of the surgically-treated cases. Treatment options are surgery and/or Radiotherapy. Till date there is no clear consensus on its treatment. We report a rare case of malignant nodular hidradenoma nose in an 80-years old male who responded miraculously to Radiotherapy.

Keywords: malignant hidradenoma, radiotherapy, sweat gland tumor

Volume 9 Issue 3 - 2018

Anthialisha Nongkynrih, Karun Kamboj, Anil Kumar Dhull, Vivek Kaushal

Department of Department of Radiation Oncology, Post Graduate Institute of Medical Sciences, India

Correspondence: Anil Kumar Dhull, Radiation Oncologist, Department of Radiation Oncology, Post Graduate Institute of Medical Sciences, PO Box 100, GPO Rohtak-124001, Haryana, India, Fax +91-126-22-99999; Email anilkdhull@gmail.com

Received: February 01, 2018 | **Published:** May 08, 2018

Abbreviations: EBRT; external beam radiotherapy

Introduction

Hidradenoma was first recognized as a distinct entity by Mayer in 1941.[1] Its malignant form, the malignant nodular hidradenoma is a very rare adnexal tumor. Its incidence is as low as <0.001%.[2] It has been reported most frequently on the head and neck and rarely on the extremities. The natural history of the disease is varied. However, owing to the propensity to metastasize to regional lymph nodes and distant viscera the tumor is said to be aggressive.[3] There is no clear consensus on its treatment till date. Surgical excision is the preferred choice of treatment wherever possible. The role of adjuvant therapy is controversial.[4] We report the rare case of Malignant Nodular Hidradenoma Nose in an 80 year old male who responded miraculously to Radiotherapy.

Case Presentation

An 80-years old male non-smoker presented to radiotherapy department with swelling on dorsum of nose for the past 1 year which was insidious in onset, gradually progressive, associated with foul smelling pus discharge and mild pain. There is no history of loss of weight and loss of appetite. Patient had no history of comorbidities. No significant family history was reported. General physical and systemic examination was normal.

Local examination revealed an ulceroproliferative growth of size 5×5cm over dorsum of nose extending to involve the left eye with foul smelling pus discharge from it (Figure 1). Histopathology and immunohistochemical profile confirmed the diagnosis as malignant nodular hidradenoma of nose. Magnetic resonant imaging of face and Neck revealed a soft tissue lesion involving skin and subcutaneous tissue of nose, extending to left periorbital region with infiltration into left preseptal space, with loss of fat planes with left medial rectus muscle and anterior sclera and reduced space of anterior chamber along with infiltration of left nasal wall with extension into nasal cavity. There were no metastases as evident from chest X-ray and ultrasonography abdomen.

Oncosurgery opinion was sought for consideration of radical surgery. However due to the massive extent of the disease, it was deemed inoperable. Thus, patient was initially treated with palliative intent using a hypofractionated External beam Radiotherapy (EBRT) regimen of 20 Gy/5 fractions/ 1week (4 Gy per fraction). Patient tolerated the treatment well (Figure 2) (Figure 3). Upon evaluation of response to treatment one month after completion of EBRT patient was found to have very good subjective as well as objective response as evident in Figure 4. Patient is asymptomatic and was planned for supplementary Radiotherapy; however he lost to follow-up subsequently.

Figure 1 Clinical photograph of patient at presentation showing an ulceroproliferative growth of size 5×5 cm over dorsum of nose extending to involve the left eye.

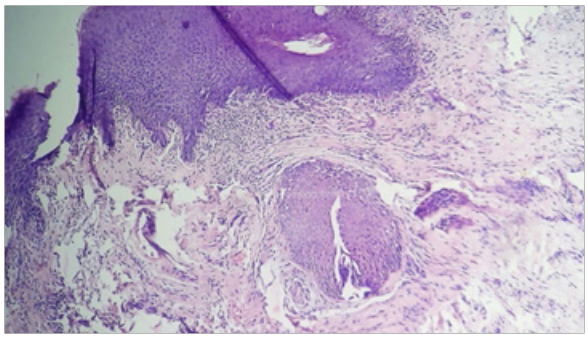

Figure 2 Haematoxylin and eosin stained slide under 10× magnification showing intradermal development of malignancy and separation from epidermis with a fibrous septa.

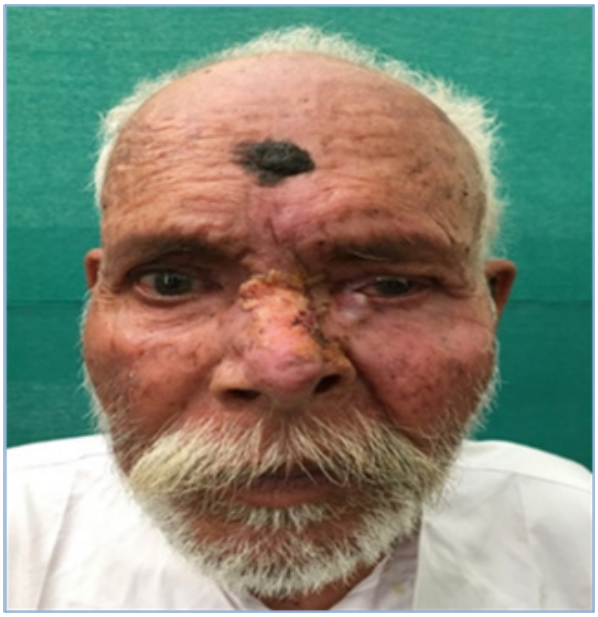

Figure 4 Clinical photograph of patient one month after treatment showing significant regression of disease.

Discussion

Sweat gland tumor was first reported in 1865 and its classification however developed in the late 1950s.[5] They comprise <1% of all skin neoplasms with an overall incidence of 6%.[5] The Malignant nodular Hidradenoma is an uncommon malignant cutaneous adnexal tumor that can show differentiation toward various components of eccrine sweat glands.[6] Malignant nodular Hidradenoma usually occur in the sixth decade of life.[7] Its incidence is reported to be <0.001% and is equal in both genders.[2,7]

The tumor presents clinically as an asymptomatic solitary skin lesion.[8] They may also appear as a nodule with or without ulceration.[9] These tumors usually occur in the scalp, face or anterior surface of the trunk.[1] It is usually a slow growing tumor however its aggressive behavior is seen in recurrent cases.[1] Its aggressive form can metastasize to regional lymph nodes or even to other distant sites.[2]

The differential diagnosis includes primary skin tumors with follicular, sebaceous, or sweat gland differentiation.[10] They can also

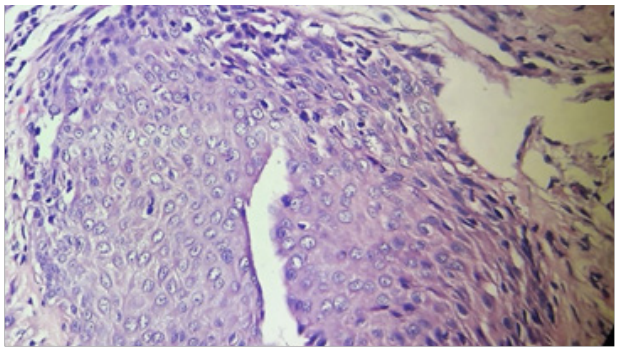

Figure 3 Haematoxylin and eosin stained slide under 40× magnification showing malignant cells with eosinophilic cytoplasm and nuclear atypia.

be mistaken with infundibular and pilar cysts, cutaneous tuberculosis or dermatofibrosarcoma protuberans.[3] Hidradenomas can mimic cutaneous metastatic disease from clear cell tumors such as renal cell carcinoma.[10]

Histopathologically, hidradenomas are said to arise from the ductal epithelium of eccrine sweat glands.[7] They are comprised of two type of cells: first, the round or polygonic cells with round nuclei and clear cytoplasm and second, the multifaceted cells comprised of oval nuclei and basophilic cytoplasm evenly arranged at the periphery of the first cellular line.[11] Characteristics of malignancy are poor circumscription, presence of nuclear atypia, mitotic activity, presence of predominantly solid cell islands, infiltrative growth pattern, necrosis, and angio-lymphatic permeation.[12]

Being a very rare disease, it is impossible to have a clear consensus on its treatment. Till date surgical excision seems to be the standard of treatment wherever it is possible. However it has been reported that recurrence rate after excision can range from 10% to 50%.[13] Wong et al recommended wide surgical resection with a least 2-cm of clear margins for both primary disease and local recurrences.[14] A special technique, Mohs micrographic surgery has been reported to have lower recurrence rate.[5] The need for lymph node dissection remains unclear. It is indicated when there are signs of lymph node local invasion; prophylactic dissection otherwise has not yet proved to increase the disease free interval. Sentinel lymph node biopsy can be done to identify a high risk of recurrence to assess the indication for a radical lymphadenectomy.[13]

The role of adjuvant therapy is controversial. Adjuvant radiotherapy should be considered if there is dermal lymphatic invasion, nerve-sheath involvement, deep structure infiltration, positive resection margins, highly anaplastic morphology, and extracapsular lymph node extension. Harari et al. reported complete remissions after external beam radiotherapy for sweat gland tumors with positive margins after surgery. In their study primary surgical beds were treated with 70Gy, using a combination of photons and electrons, and regional lymphatic chains with 50Gy.[15] The use of upfront radiation radiotherapy without surgical excision is rare. It is recommended to individualize the treatment strategy.[16]

In our patient palliative external beam radiation therapy was given as the disease was too extensive for surgical excision. To our surprise, patient responded miraculously with significant regression of disease after radiotherapy. We, thus conclude that Radiotherapy is a good

Citation: Nongkynrih A, Kamboj K, Dhull AK, et al. Miraculous response to radiotherapy in a rare case of malignant nodular hidradenoma nose. *J Cancer Prev Curr Res.* 2018;9(3):97–99. DOI: 10.15406/jcpcr.2018.09.00328

palliating tool for malignant nodular hidradenoma. This paves the way for new research to establish the role of Radical Radiotherapy in such cases.

Acknowledgements

None.

Conflict of interest

There is no conflict of interest in publishing the article.

References

1. Lliapakis IE, Korkolis DP, Koutsoumbi A, et al. Malignant hidradenoma: a report of two cases and review of the literature. *Anticancer Research*. 2006;26:2217–2220.
2. Sinhasan SP, Harthimath BC, Sylvia MT, et al. Metastatic malignant nodular hidradenoma: A rare case report with review of literature. *Clin Cancer Investig J*. 2015;4(5):651–654.
3. Nair PS. A clinicopathologic study of skin appendageal tumors. *Indian J Dermatol Venereol Leprol.* 2008:74(5):550.
4. Souvatzidis P, Sbano P, Mandato F, et al. Malignant nodular hidradenoma of the skin: Report of seven cases. *J Eur Acad Dermatol Venereol*. 2008;22(5):549–554.
5. Giorgini E, Tugnoli G, Aprile S, et al. Malignant nodular hidradenocarcinoma arising on the areola of a male patient: Case report of an "orphan disease" and review of the literature. *J Carcinog Mutagen*. 2012;3:129–133.
6. Maiti T, Somanna S, Devi BI, et al. Malignant nodular hidradenoma of scalp. *Journal of Neurosciences in Rural Practice.* 20144;5(4):423–425.
7. Singh G, Kumar H, Sridevi NS, et al. Malignant clear cell hidradenoma of the upper eyelid. *Electronic Physician*. 2012;4(2):490–492.
8. Ohta M, Hiramoto M, Fujii M, et al. Nodular hidradenocarcinoma on the scalp of a young woman: Case report and review of literature. *Dermatol Surg*. 2004;30(9):1265–1268.
9. Crowson AN, Magro CM, Mihm MC. Malignant adnexal neoplasms. *Modern Pathology*. 2006;19:93–126.
10. Volmar KE, Cummings TJ, Wang WH, et al. Clear cell hidradenoma: A mimic of metastatic clear cell tumors. *Arch Pathol Lab Med.* 2005;129:113–116.
11. Hashimofo K, Di Bella RJ, Lever WF. Clear cell hidradenoma. *Arch Dermatol*. 1967;96:18–38.
12. Baghli AA, Reddy SS, Reddy MA. Malignant Nodular Hidradenoma of the Eyelid: A Rare Sweat Gland Tumor. *Middle East Afr J Ophthalmol.* 2010;17(4):374–376.
13. Amel T, Olfa G, Faten H, et al. Metastatic hidradenocarcinoma: Surgery and chemotherapy. *N Am J Med Sci.* 2009;1:372–374.
14. Wong A, Suster S, Nogita, et al. Clear cell eccrine carcinomas of the skin. A clinicopathologic study of nine patients. *Cancer.* 1994;73(6):1631–1643.
15. Harari PM, Shimm DS, Bangert J. The role of radiotherapy in the treatment of malignant sweat gland neoplasms. *Cancer.* 1990;65(8):1737–1740.
16. Bansal N, Khurana A, Kaur P, et al. Malignant Nodular Hidradenoma of Face. *Online J Health Allied Scs*. 2014;13(3):7.