Case Report:
Malignant Nodular Hidradenoma of Face.

Authors
Nupur Bansal, Senior Resident,
Anil Khurana, Medical Officer,
Paramjeet Kaur, Associate Professor,
Ashok KR Chauhan, Senior Professor,
Department of Radiotherapy, Regional Cancer Centre, Pt B.D.S. Sharma PGIMS, Rohtak,

Ajit Singh Rathee, Professor,
Ajay Kapoor, Senior Resident,
Department of Burns and Plastic Surgery, Pt B.D.S. Sharma PGIMS, Rohtak

Rajeev Sen, Senior Professor,
Department of Pathology, Pt B.D.S. Sharma PGIMS, Rohtak

Address for Correspondence
Dr. Nupur Bansal,
Department of Radiotherapy,
Regional Cancer Centre,
Pt B.D.S. Sharma PGIMS,
Rohtak, India.
E-mail: drmupurbansal@gmail.com

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Abstract: We report a case of malignant nodular hidradenoma in an old woman, who presented with a nodular swelling in the right side of nose near the medial canthus of the right eye. Wide excision of the nodular mass with a clear margin of healthy surrounding tissue was performed along with primary closure. Post operatively, adjuvant radiation therapy was given on a telecobalt machine due to the presence of high risk features. In general, malignant forms of hidradenomas are not usual and treatment strategies should be individualized.

Key Words: Malignant hidradenoma, Eccrine sweat gland tumor, Radiotherapy.

Introduction:
The recognition of hidradenoma as a distinct entity was first reported in 1941 by Mayer. The malignant form of hidradenoma is extremely rare, with less than 50 cases ever reported in the literature. All these cases were characterized by a significant rate of locoregional recurrence. Some patients developed distant metastatic spread as well.[1] A variety of names are applied to dominantly dermal-based malignant eccrine tumors, including hidradenocarcinoma, malignant acrospiroma and clear cell eccrine carcinoma.[2] The overall incidence of all eccrine carcinomas is 6% which represent <1% (0.1-1) of all skin neoplasms.[3] Body involvement is 65% on the soles, 10% on the palms and 25% in other regions (extremities, face, neck and trunk).[4]
Figure 1: Skin lesion on the right eyelid

Figure 2: The site of lesion after surgery

Figure 3: Photomicrograph shows skin lined with stratified squamous epithelium. Dermis shows lobulated mass extending into subcutaneous tissue revealing follicular and clear cells.

Figure 4: Photomicrograph shows two cell types - 1. Polygonal cells with rounded nucleus and slightly basophilic cytoplasm 2. Clear cells with rounded nucleus and clear cytoplasm.

Figure 5: CECT scan of head with normal findings.
Discussion

Hidradenoma usually affects middle-aged women, although its malignant form shows no age or gender predilection. It is predominant in females. Though, in our case, the disease was noticed as a nodule in an old female without any lymph node involvement.

As described in the literature, these neoplasms present as nodules, frequently with superimposed ulceration and rapid growth, affecting the head and neck and the distal extremities of the elderly. Regional lymphadenopathy, with or without serous discharge, may develop years after initial treatment. These tumours are postulated to arise from the intradermal duct of eccrine sweat glands. Histologically, sweat glands may be either eccrine or apocrine in nature. Eccrine glands are present throughout the skin but are most abundant in the palms, soles, and axillae. Apocrine glands are found in relatively few regions of the body, mainly the axillae, around the nipples, the anogenital region, and occasionally a small number on the abdomen and chest. Eccrine hidradenomas are also referred as nodular hidradenoma or clear cell hidradenomas arising from the eccrine sweat glands. This nomenclature is based on electron microscopy and histochemical studies. The differential diagnosis includes primary skin tumors with follicular, sebaceous, or sweat gland differentiation. Hidradenomas can mimic cutaneous metastatic disease from clear cell tumors such as renal cell carcinoma. The possibility of a primary basal cell carcinoma with eccentrical differentiation and a lobular, hyalinised syringoma should also be included in the histological differential diagnosis. The criteria for malignancy include poor circumscription, presence of nuclear atypia, mitotic activity, presence of predominantly solid cell islands, infiltrative growth pattern, necrosis, and angio-lymphatic permeation. The 5-year postsurgical survival rate for malignant nodular hidradenoma is reported to be less than 30%. Surgical excision remains the therapeutic modality of choice. At the time of excision of eccrine tumors, some researchers have utilized the Sentinel Lymph Node Excision and prevent the occurrence of regional lymph node metastasis. These authors suggest that spotting a sentinel lymph node could be the right diagnostic approach for staging the disease as it happens for breast cancer. Positivity of sentinel lymph node also permits to assess the indication of excision of the primary and local recurrences. Mohs micrographic surgery may prove superior to the conventional excision and manifest a lower recurrence rate. Elective regional lymphadenectomy after lymphoscintigraphy should also be performed. At the time of excision of eccrine tumors, some researchers have utilized the Sentinel Lymph Node Excision and some reported complete resections after external beam radiotherapy for sweat gland tumors with positive margins after surgery, the dose and technique of radiotherapy are not consensual. In the work of Harari et al., primary surgical beds were treated with 70 Gy, using a combination of photons and electrons, and regional lymphatic chains with 50 Gy. Hyperfractionation schemes were used in two patients to minimize late normal tissue effects.

As mentioned in various studies described above, the treatment strategy is individualized. In our patient, wide local excision of the mass was done. Adjuvant radiotherapy was given due to the presence of high risk features. The main post radiotherapy side effect seen in our patient was epiphora. Though, side effects and complications may vary according to primary site and are dose dependent. There are both acute and long-term sequelae of radiation therapy for head and neck cancer that occur because of effects on normal tissues. Some common adverse effects include mucositis, xerostomia, trismus, hearing loss, and facial nerve dysfunction. Severe late complications include the risk of soft tissue necrosis, osteoradionecrosis, oro cutaneous fistula, blindness, and second malignancies. With the advent of new techniques of radiotherapy; such side effects are less frequent and better tolerated.

Conclusion

Malignant nodular hidradenoma is a rare oncological entity, with no particular clinical or histopathological features. It should be included in the differential diagnosis of skin lesions.

References