Case Report

Struck by Lightning and Bitten by Snake: Two Coexisting Neoplasms in Same Anatomical Area

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ABSTRACT

Introduction

The simultaneous occurrence of a benign as well as malignant epithelial lesion, albeit incidental, is a rare occurrence. A multi stage procedure had to be adopted for the appropriate management of this patient.

Case Report

We report one such case of a 65 year old male who presented with a small black lesion on the left side of the nose and complains of left sided nasal obstruction on a much later date.

Discussion

The decision to address the benign pathology initially followed by the malignant pathology owing to its indolent course formed the cornerstone in the management of this patient.

Keywords

Carcinoma, Basal Cell; Papilloma, Inverted; Surgical Flaps, Forehead

Basal cell carcinoma comprises 75% of Non Melanotic Skin Cancers (NMSCs), affecting mainly middle aged or elderly fair skinned people.[1] Inverted papilloma although an uncommon epithelial tumour of the nasal cavity, is the second most common benign tumour of the sinonasal tract and the most common form of sinonasal papilloma, with an incidence of 0.74 to 2.3 new cases per 1 lakh population per year.[2,3]

Case Report

A 65 year old male patient from West Bengal, working in a printing press came with complaints of a small black lesion on the left side of the nose for 7 years, which was insidious in onset and gradually increased in size, followed by ulceration of the involved skin and destruction of the left ala of nose since 2 years. There is no history of prior radiation therapy or any immunosuppression. He also developed left sided nasal obstruction for 8 months, which was unilateral, continuous and associated with recurrent episodes of bleeding from the left nasal cavity.

Local Examination revealed a 1.5 by 1 cm lesion eroding the left ala of the nose with well-defined, irregular and elevated margins and surrounding black ulceration. (Fig.1) On Diagnostic Nasal Endoscopy, a pale papillomatous mass was seen in the left nasal cavity, which appeared to be coming out of the middle meatus. (Fig.2)

Routine hematological and biochemical investigations were within normal limits. Contrast Enhanced CT scan of the nose and paranasal sinuses showed a homogenously hyperdense lesion in left nasal cavity and left maxillary antrum with focal hyperostosis at the infero-medial corner of the orbit. (Fig.3)

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Fig. 1. Eroded left ala of nose

Fig. 2. Diagnostic Nasal Endoscopy: pale papillomatous mass in left nasal cavity coming out from the middle meatus

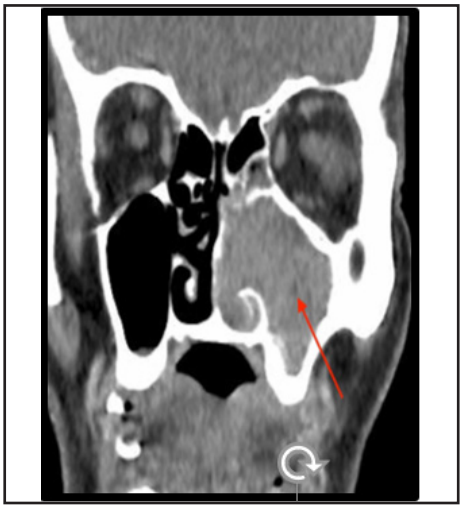

Fig. 3. Pre operative CECT scan of nose and paranasal sinuses coronal view: homogenously hyperdense lesion in left nasal cavity and left maxillary antrum with focal hyperostosis at the infero-medial corner of the orbit

Wedge biopsy from the left ala of the nose revealed features of basal cell carcinoma and punch biopsy of mass from the left nasal cavity revealed inverted papilloma.

Wide local excision of left ala of nose with a margin of 1 cm was done (Fig.4), accompanied by left sided medial maxillectomy via lateral rhinotomy approach with removal of the sinonasal mass in the same sitting. The sites of attachment were denuded of mucosa and drilled upto normal healthy bone.

HPE of the mass from left ala revealed Basal Cell Carcinoma.

HPE of the sinonasal mass revealed Inverted Papilloma.

Subsequent reconstruction of the nasal defect was done using a combination of forehead flap and local advancement flap with conchal cartilage as a base. (Figs. 5,6,7)

This was followed by release of the forehead flap after 3 weeks. (Fig.8)

Patient is on monthly follow up and is doing well, with no recurrence of either lesion till date.

Discussion

According to 2017 WHO Classification of Head and Neck tumours, sinonasal papillomas are divided into inverted, oncocytic and exophytic types.[4] Incidence is 2 to 3 times more common in males than females, with

Fig. 4. Per operative image of excised left ala of nose

Fig. 5. Per operative marking for the advancement flap

Fig. 6. Per operative marking for the forehead flap

majority occurring in the 5th and 6th decades of life. Most common site of occurrence is from the medial wall of maxillary antrum, in the region of the fontanelles. Multiple sites are involved in 30% of the cases. Bilateral papillomas are very rare.[2]

These tumours are locally aggressive and have a tendency to recur.

Possible role of HPV as an etiologic factor in the development of inverted papilloma and its role in malignant transformation is currently unclear.[5,6]

Computed Tomographic (CT) Scan is the primary mode of imaging, and helps in assessing the site of origin of the lesion, which can be reliably identified by the presence of a bony strut,[7] focal hyperostosis[8] and osteitic[9] changes. It helps in evaluating the extent of the lesion and its relationship with the surrounding structures. The cerebriform columnar pattern, which is highly predictive of inverted papilloma, is seen on MRI. This pattern is lost in case of malignant transformation.[10] MRI is complementary to CT and is recommended with enhancement to differentiate between tumour and inflammatory mucosal changes. However, since

Fig. 7. Per operative image of the reconstructed nasal defect

Fig. 8. Per operative image following release of the forehead flap

the diagnosis of inverted papilloma was histologically confirmed and there were no clinical and CT features suggestive of intra orbital or intracranial extension, MRI was not done in this patient.

Histologically, inverted papilloma is characterised by epithelium inverting into the stroma with a distinct and intact basement membrane. It is composed of hyperplastic ribbons of basement membrane enclosed epithelium that grow downward into the underlying stroma.[10] EGFR mutation, which is normally present in inverted papillomas, is preserved even in case of malignant transformation.[11]

Malignant transformation is seen in 5 to 15% of cases, with the most common malignancy being squamous cell carcinoma.[10]

The extent of surgery in this patient was decided based on the site of origin and area of mucosa involved by the lesion, with the aim being to dissect the involved mucosa in the subperiosteal plane and drill the underlying bone till normal bone is visible.[12,13]

Endoscopic endonasal surgery is the gold standard and is preferred over external approach as there is a lower chance of recurrence, absence of a facial incision and reduced post operative pain and paraesthesia.[10] Endoscopic approach should not be used exclusively if a malignancy is present concomitantly with involvement of critical areas, if the inverted papilloma arises from the anterior wall or lateral recess of the frontal sinus or there is massive involvement of the mucosa of the frontal sinus and/or of a supra-orbital cell, or if the orbit is involved.[10]

However, an external approach was advocated in this patient as in our setup, we are more conversant with the open approach.

Follow up by DNE is recommended every 4 months for 2 years, every 6 months for the next 3 years and every year for the next 5 years.[10]

Post operative imaging is required only if the sinus that was originally involved by the tumour is inaccessible for DNE following surgery due to scarring, if the patient is symptomatic or if any residual or recurrent lesion has been histologically documented. At present, MRI is the investigation of choice for suspected recurrence.[10]

Basal cell carcinoma is most common in sun damaged areas of skin. Other risk factors include fair skin (although it affects people with darker skin also), previous cutaneous injury, thermal burn, ionising radiation, smoking, inherited syndromes (such as Gorlin syndrome, Bazex syndrome, Rombo syndrome, xeroderma pigmentosa). Four distinct clinicopathologic types are known-nodular, superficial, morpheaform and fibro-epithelial. Nodular BCC consists of 50% of all BCCs.[4]

Most common site of occurrence of BCC is over the face (especially cheeks, nose, nasolabial fold, forehead and eyelids), but may be seen in any hair bearing area of the skin.

Nodular BCC presents as a shiny, pearly papule or nodule with a smooth surface and arborising telangiectasias. It can gradually enlarge and ulcerate, hence also known as rodent ulcer or phagedenic ulcer. A raised, rolled out border is usually diagnostic. It is locally invasive, but distant metastases are rare.

On HPE, nodular BCC appears as large, round or oval aggregates of basaloid keratinocytes extending from the epidermis into the dermis, with prominent peripheral palisading and peritumoral clefts. The nuclei lack organisation and are more randomly distributed in larger tumour islands.

Imaging techniques like CT or MRI is used to assess invasion of bone, nerves or orbit. Perineural invasion is indicative of a higher chance for recurrence after surgery.[1]

Standard excision with a peripheral margin of 4-5mm and deep margin down to fat have proved to be adequate for removal in most cases of non morpheaform BCC less than 2cm in diameter. Excision is said to be adequate if histologically at least 1 mm of tumour free margin is achieved.[1]

Once the margins for the basal cell carcinoma was proven to be adequate, reconstruction of the defect was planned by using a local advancement flap for reconstruction of the nasal mucosa, supported by conchal cartilage as base, and the cartilage was covered by using the forehead flap based on the supratrochlear artery. There are several challenges in using the forehead flap, such as achieving tension free closure of the donor site as well as the flap site, poor scarring, infection,

dehiscence and flap necrosis. This was minimised by proper patient selection and proper surgical planning, especially by placing parallel vertical galeotomies, 2 to 3 cm apart to the level of the subcutaneous fat for tension free closure of the donor site.

The patient was scheduled for flap detachment approximately 3 weeks from the date of initial surgery.

Literature shows that cure rates for BCC with radiotherapy alone is between 79% to 100%, and thus can be used as a primary option for treatment in patients in whom surgery is contraindicated. Disadvantages include lack of margin control, poor cosmesis, prolonged course of therapy and increased risk of cancers in the future.[1]

The co-existence of a benign as well as malignant epithelial lesion in a single individual is a highly unusual event and thus the management had to be tailored as a multi stage procedure keeping in mind both the pathologies as well as the socio-economic profile of the patient. Although the basal cell carcinoma does not need further follow up due to the completeness of excision, the patient has to be followed up with regular DNE to rule out the recurrence of inverted papilloma in the future.

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