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RESEARCH ARTICLE

PLANTAR CARCINOMA CUNICULATUM: A RARE PRESENTATION IN THE FOOT REGION

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Abstract

Carcinoma cuniculatum is a rare variant of squamous cell carcinoma. The clinical presentation is usually a non-verrucous exophytic plaque or tumor of the plantar region with a penetration in the deep tissues. Histological examination shows a proliferation of well-differentiated keratinocytes. We describe a patient affected by a slowly enlarging tumoral lesion overlying the fifth metatarsum of the left foot. Clinical examination and radiological investigations suggested a chronic osteomyelitis and a first histological examination of a punch biopsy was suggestive of a pseudo-epitheliomatous hyperplasia. The patient underwent several cycles with systemic antibiotics without improvement. Finally, the fifth metatarso was amputated and the skin lesion was completely removed. The histological examination of the whole operatory mass allowed a diagnosis of carcinoma cuniculatum invading the bone.

Introduction:-

Carcinomacuniculatum (verrucouscarcinoma) is an exoendophytic type of low-grade squamouscellcarcinomathatoftenpresents as a slowlyenlargingpapillated tumour.[1] First described in the English-languagemedicalliterature by Aird et al. in 1954, the prevalence of CC remains unknown.[2] In addition to the foot, it may also occur in the oral and anogenital regions.[3]

Males in their fifties are known to be the mostusuallyaffected (79–89% of individuals.[4]Recalcitrantplantarwarts or chroniculcersmaygiverise to carcinomacuniculatum. The occurrence of CC in keratodermatous skin is an uncommon occurrence that has been documented in severe and widespreadkeratoderma, but itisextremely rare in focusedplantarkeratoderma. Clinically, itischaracterized by an exophytic mass thatisfungating and has severalsinusesfilledwithkeratin.

Wepresent the findingsfromtwo patients whohadplantarcarcinomacuniculatum.

Observation 1:-

A 55-year-old man ,with a 4-month history of a painful, nonhealinglesion on the ball of his right foot.Hehadattempted to removea long-standing callus at this site withoutsuccess and the area hadbecomeulcerated and malodorous. . He had no family history of keratoderma or cutaneous squamous cell carcinoma (SCC). There was no pasthistory of medicinal or occupational arsenic exposur.

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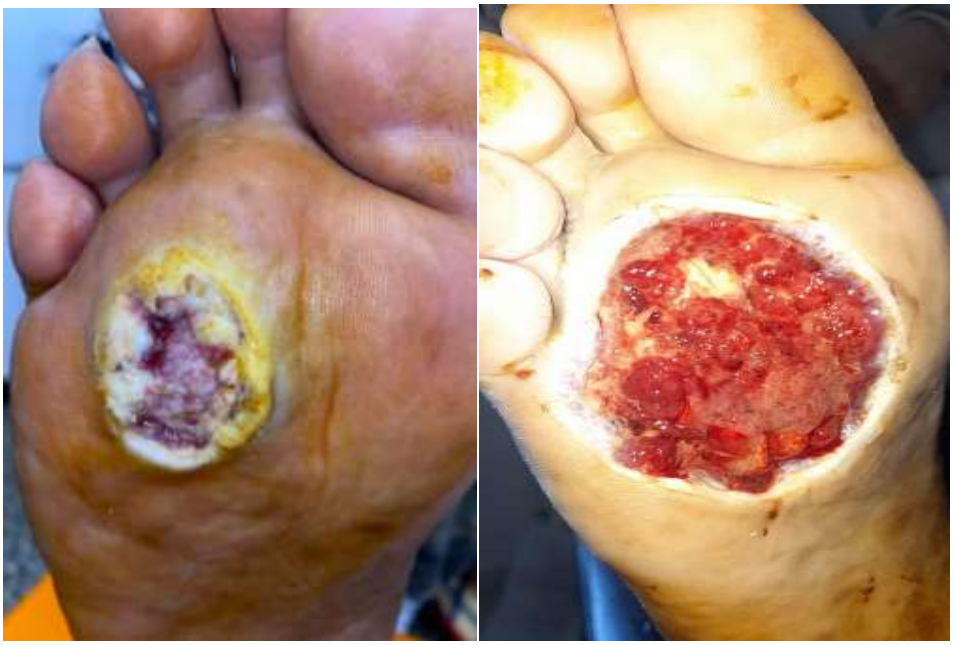
General examinationwasunremarkablewith no palmarkeratodermapresent. Examination of the feetrevealed an ulcerating-bourgeoningtumor on the plantar surface of the right foot, about 2.5cm long, welllimited, with a verrucous hyper-bourgeoning background, neitheroozingnorhemorrhagic, and with a keratotic border with a peripheralhorn.

Twocallositieswerealsonoted; on the plantar surface and on the second toe of the left foot, measuringapproximately 1 cm and 0.5 cm respectively.

Then a skin biopsy of the tumorwasperformedwithanatomical-pathologicalexaminationwhichshowed an exophyticepithelialtumorwithepithelialoutgrowthssurmounted by a thick ortho and parakeratotic stratum corneumharboringnumerousvacuolatedkoilocyticcells on the surface, sometimesbinucleated, thisis in relation with a cuniculatumcarcinoma. The standard biological exams were normal and an X-ray of the right foot wasdonewhichshowed no signs of osteitis or bone infiltration. A thoracic-abdominal-pelvic CT scan wasperformed and was not showinganyparticularities. The patient wasreferred to theirdepartment for surgicalexcision . The patient was re-admitted to our service at day 20 postoperatively to follow the process of adequatedirectedhealing . The after-effectswermarked by a very good clinicalimprovement and a completehealing of the lesion.



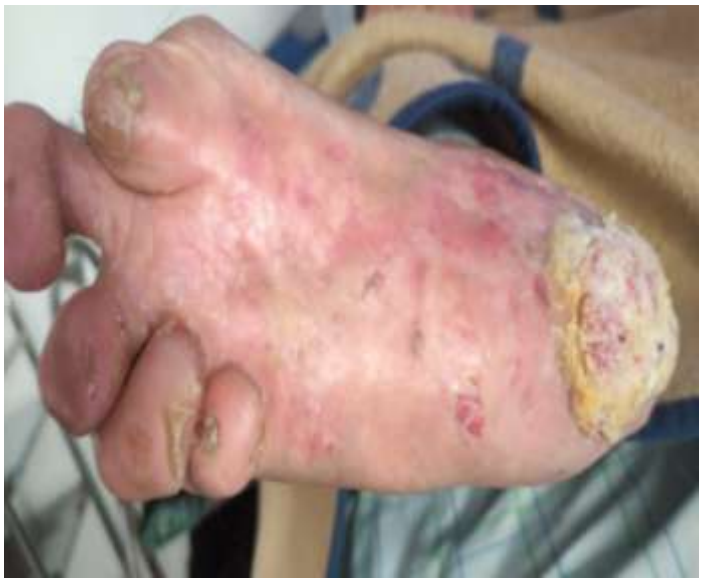
Observation 2:-

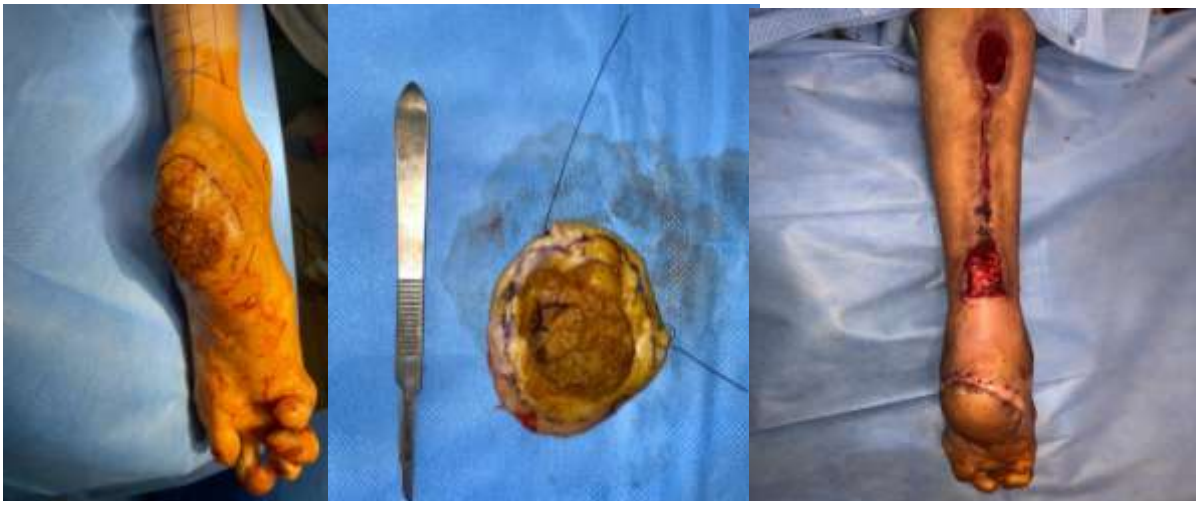
A 48 yearold male patient who came to the dermatological consultation with an ulceratingtumoralreadydiagnosed at an earlyagewithvowhinkel syndrome consisting of diffuse palmoplantarkeratoderma, pseudoainhum of the toes, and sensorineuralhearingloss

Examination of the feetsrevealed a diffuseplantarkeratoderma and an ulcerating-bourgeoningtumor on the left heel , about 04 cm long, welllimited, with a verrucous hyper-bourgeoning background, neitheroozingnorhemorrhagicnormalodorous, and with a keratotic border.

The diagnosis of carcinomacuniculatumwasthenheld on the histologicalexamination of a deepbiopsyspecimen.

The standard biological exams were normal and an X-ray of the right foot wasdonewhichshowed no signs of osteitis or bone infiltration. A thoracic-abdominal-pelvic CT scan wasperformed and was not showinganyparticularities. He wasWesoughtadvice the department of plastic surgery for exeresis and reconstruction by Neuro Sural Flap.





Discussion:-

Carcinomacuniculatumis a rare subtype of verrucouscarcinomausuallyaffecting the sole of the foot.[3] It destroysnearby structures, includingbone, whenitinvades a localized area. Althoughextremely rare, metastases have been documented.[3] The clinicaldifferentialdiagnosisincludesplantarwart, amelanoticmelanoma and sarcoma.Due to the earlycommonclinicalcharacteristics and slow progression thatfrequentlyresemble a viral plantarwart, the diagnosismay be delayed. Eventually, though, a fungating plaque progresses. Deep incisional biopsies are required for a diagnosissincesuperficial biopsies mayrevealnonspecificcharacteristicsthatprovide false reassurance. Additionally, histology can bemodest, showinglittle to no cellular pleiomorphism and a "pushing" edge as opposed to an infiltrative one. The termcuniculatum, whichis Latin for "rabbitwarren," comesfrom sinus apertures on the skin's surface thatconnect to interconnecting tunnels and cleftsthatinvadedeeply in multiple directions. In the original description, this structure wassupposed to resemble a rabbit warren.[2] The histopathologicdifferentialdiagnosisincludeskeratoacanthoma and pseudoepitheliomatous hyperplasia.[1-5]

To estimate the amount of excision required for a cure, twoMRIs can bedone to assess the tumor's size and depth. Since MRI often has a high sensitivity but a lowspecificity, itistypically not useful for diagnostic purposes.[6]

In cases of severe, widespreadkeratoderma, such as Huriez syndrome, Vowhinkel syndrome,[7]mutilatingkeratoderma,[8] Clouston's syndrome,[9] porokeratosis, and Unna-Thost hyperkeratosis.[10]The development of an SCC in keratodermatous skin is an uncommon occurrence.

The standard treatments for carcinomacuniculatum are surgical excision and Mohs' technique (serial excision for microscopicanalysis), both of which are associatedwith a high cure rate and a lowrecurrence rate. Alternative treatmentsinclude curettage and electrodesiccation, cryosurgery, carbon-dioxide laser therapy and radiotherapy.[3]Typically, the tumourremains indolent for years, althoughitmayextendinto the subcutaneous tissue or metastasize.[5]

In conclusion, when soft tissue lesions are not clearlydifferentiatedclinically, like those of a cuniculatumcarcinoma, a simple 3- or 4- millimeter punch biopsyshouldbeperformed to prevent the undesirable and destructive consequences of thislesion. It istherefore important to differentiatebetweencunuculatumcarcinoma andotherdifferential diagnoses such as plantarwarts, callosities, or others in order to achieveappropriate and prompt surgical management and better follow-up of the tumor.

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