

Frontal sinus involvement of Cutaneous Lymphoid Hyperplasia- A rare case report

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Abstract: Cutaneous lymphoid hyperplasia (CLH) is considered a benign lymphoid reactive process that results from various antigenic stimuli and may have potential for progression to overt lymphoma [1]. It is characterized by the appearance of violaceous, firm nodules most commonly found over the head, neck and extremities [2]. Here we present a rare case of a 47 year old female with frontal sinus involvement of cutaneous lymphoid hyperplasia. This was excised and sent for a histopathological examination. To the best of our knowledge, there is no documented case of cutaneous hyperplasia extending into the paranasal sinuses. The clinical diagnosis, the management, including the surgical intervention are discussed.

Keywords: Frontal Sinus Lesion, Cutaneous Lymphoid Hyperplasia

I. Introduction

Cutaneous Lymphoid Hyperplasia or Spiegler-Fendt sarcoid, is classified as one of the pseudolymphomas, referring to inflammatory disorders, which clinically and histologically closely resemble lymphomas [3]. CLH has a world-wide distribution and affects all races and ethnic groups. It occurs in both adults and children, affecting females more than males [4]. CLH mostly involves the face, neck, chest and upper limbs [5]. Here, we describe a case of CLH in a 47-year old female, with a history of a swelling in the midline of the forehead since 16 years, for which the patient underwent medical management, but was not relieved completely. The patient was referred to our ENT department, where the swelling was excised and the diagnosis of Cutaneous lymphoid hyperplasia was made. Till now, the patient follows up with no complaints.

II. Case report

A 47-year old female patient, was referred to our department with a complaint of a recurrent swelling over the forehead towards the midline, since the past 16 years.

In 1998, the patient developed an asymptomatic midline forehead swelling. A plastic surgery opinion was taken and an excisional biopsy offered a diagnosis of Jessner's lymphocytic infiltration, after which the swelling had regressed. In 2000, a similar skin lesion had developed in the same midline forehead area and spread to the right infraorbital region. The biopsy reported a reactive lymphoid proliferation. A subsequent MRI showed extension into the orbit with no cranial nerve involvement. She was treated with tetracyclines and Potassium Iodide till 2002. During this time, the lesion had not progressed further.

In 2008, the patient developed epiphora from the right eye, revealing an obstructed lacrimal duct, for which an endoscopic dacryocystorhinostomy was done.

In 2015, the patient was referred to us for ENT intervention. The patient presented with a midline forehead swelling which was asymptomatic. There was no past history of insect bites, drug intake and photosensitivity. On inspection, there was a solitary swelling on the glabella, measuring approximately 1x2 cm, oval-shaped with ill-defined margins and an absent impulse on coughing. The swelling extended horizontally between the medial ends of the eyebrows and vertically from the nasal bridge, spanning the glabella area superiorly. The skin over the swelling appeared erythematous with no secondary changes. On palpation, all the inspection findings were confirmed. The swelling had a smooth surface with no local rise in temperature and was non-tender. There were no significant, palpable lymph nodes.

A diagnostic nasal endoscopy showed bilateral normal middle meatus and frontal recess.

A magnetic resonance imaging scan (plain) of brain and paranasal sinuses showed a 1.8 x 0.9 cm, encapsulated, complex nodular, soft tissue lesion at the bridge of the nose with fistulous communication of the left frontal sinus in the midline.

Therefore an excisional biopsy was planned. A horizontal incision was given below the swelling from the medial end of one eyebrow to the other. The swelling was extending into the frontal sinus, eroding its anterior plate and the entire lesion was excised in toto.

Histopathological examination of the resected tissue showed a polymorphous population of lymphoid cells admixed with plasma cells, histiocytes and eosinophils, which is consistent with Cutaneous Lymphoid Hyperplasia.

The patient has improved cosmetically, along with medical management consisting of immunomodulatory drug, Thalidomide. She has no further complaints.

III. Figures



Fig 1 : Midline Forehead Swelling.

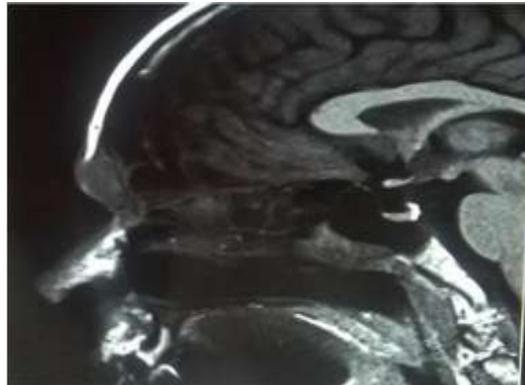


Fig 2 MRI Brain and PNS showing soft tissue lesion with fistulous communication with the frontal sinus.



Fig 3: Postoperative photo of 3 months showing no recurrence of swelling.

IV. Conclusion

Common frontal sinus lesions include osteomyelitis, osteoma and mucocele of the frontal bone, but our patient had no symptoms of headache, nasal discharge, double vision or protrusion of the eyeball. The patient's previous biopsy reports were suggestive of some immunological factor. Therefore a histopathological examination was necessary to appropriately treat the patient.

Our objective, in the patient's best interest, was to remove this cosmetically disturbing lesion, in its entirety and send it for histological testing. Immunohistochemical investigations had confirmed a diagnosis of Cutaneous lymphoid hyperplasia, also referred to as Lymphocytoma cutis.

Cutaneous lymphoid hyperplasia, as the name suggests, is a skin lesion which is benign and consists of lymphocytic aggregates, responding to antigenic stimuli, with the potential to transform into a lymphoma .It is not known to traverse layers of the skin and cause bone destruction, as described in this case. Therefore, it becomes important for us to keep in mind the diagnosis of Cutaneous lymphoid hyperplasia, for a swelling involving the frontal sinus

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