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Case report

Inflammatory Pseudotumor Presenting as a Facial Swelling

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A B S T R A C T

We present a case of inflammatory pseudotumor (IPT) presenting as a facial swelling after an accidental hit on a right side of a face. As swelling did not resolve, dental examination and teeth extraction were done by a dentist presuming the swelling was misdiagnosed with infection of dental origin. Swelling grew even bigger and patient was referred to Department of Maxillofacial Surgery. CT scan of the face and FNA of the lesion was ordered. A homogenous tumor mass in the right infraorbital region in front of anterior wall of the maxillary sinus was seen on CT. The result of the FNA was reactive hyperplasia of the lymph node. Since the lesion was easily accessible surgical exploration and complete extirpation was done. Pathohistological analysis indicated a low grade B-cell Non Hodgkin lymphoma. PCR showed policlonality of B cells discarding the diagnosis of lymphoma. Pathohistological review showed diffuse intramuscular, perineural and perivascular infiltration with small lymphocytes without formation of germinative centers. Imunohistochemistry was positive for CD20 and CD3. Taking into account all features the diagnosis of IPT was established. Diagnosis of IPT is a diagnosis by exclusion, combining clinical, radiological and pathohistological characteristics. Lack of clear histologic criteria makes differential diagnosis extremely difficult. Our case is unique regarding localisation of head & neck IPT, no case presenting on the face in infraorbital region has been described in the literature. Although IPT is very rare in general and especially on the face, one should be aware of it when considering differential diagnosis of facial swelling.

***Key words**: inflammatory pseudotumor, plasma cell granuloma, inflammatory miofibroblastic pseudotumor, face, head & neck*

Introduction

Inflammatory pseudotumor (IPT) is a rare benign condition that can sometimes be difficult to distinguish from malignant processes. There are many synonims for inflammatory pseudotumor: plasma cell granuloma, inflammatory myofibroblastic pseudotumor, xanthogranuloma, histiocytoma, and myofibrohistiocytic proliferation[1]. Most often it affects lungs, gastrointestinal tract and orbits. It is rare in the head and neck. Various extraorbital sites have been described in the literature: larynx[2,3], hypopharynx[4], scull base[5,6], parapharyngeal space[7], trachea[8,9], paranasal sinuses[10,11], tonsills[12], inner and middle ear and temporal bone[13], parotid[14]. We report of a case of inflammatory pseudotumor occuring on the face in the infraorbital region, a site that has never been described as a site of origin of IPT.

Case Report

Forty four year old male sustained an accidental hit in a right side of the face after which the swelling never completely resolved but infact got bigger. In the meantime he was treated by his dentist who extracted first upper right molar and first upper right premolar presuming the swelling in the fossa canina was caused by dental infection. Even the intraoral incision was performed in order to drain the abscess (swelling) but there was no pus. Three months later he was reffered to our outpatient department because of infraorbital swelling. On clinical examination right-sided infraorbital, hard, painless swelling was noted (Figure 1). Intraoraly just a moderate swelling in the right upper vestibulum was present in the region of upper right canine. Otherwise head and neck examination was normal. Fine needle aspiration

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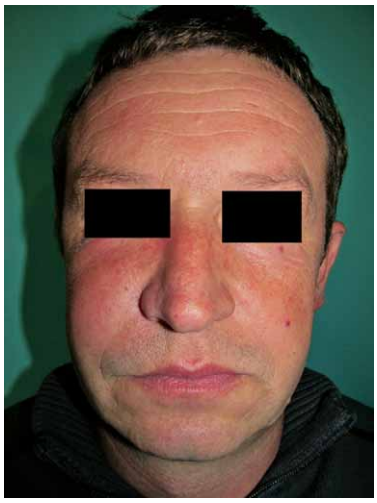

Fig. 1. Patient preoperatively with right-sided infraorbital swelling.

(FNA) suggested that it was a reactive hyperplasia of a lymph node. CT scan of the face was performed as well, there was a 35x15x25mm homogenous mass in the right infraorbital region in front of anterior wall of the maxillary sinus without bone destruction (Figure 2). Since all studies showed inconclusive results and the lesion was easily accessible surgical exploration was done. The lesion was approached through mucosal incision in the

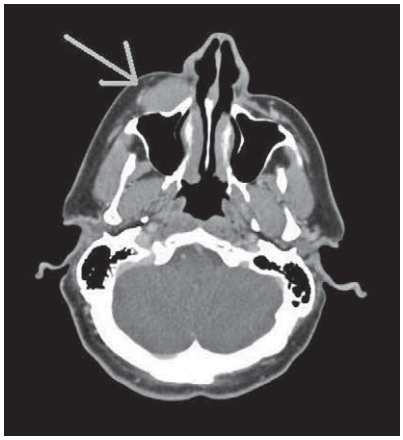

Fig. 2. CT scan, axial view, arrow pointing at infiltrative homogenous mass infraorbitally lying on the anterior wall of the maxilla.

right upper vestibulum. The mass was easilly identified and extirpated. It was infiltrative to soft tissues of the cheek while it was easy to dissect it from the bone (anterior wall of the maxillary sinus). Postoperative course was uneventfull and the patient was discharged the next day. Because of massive infiltration of the soft tissues with lymphocytes pathohistological analysis indicated a low grade B-cell Non Hodgkin lymphoma so the material was sent further for PCR. PCR showed policlonality of B cells. According to PCR results hematologist discarded the diagnosis of lymphoma or any other hematologic disease. Pathohistological revision of the material was done. Again diffuse intramuscular, perineural and perivascular infiltration with small lymphocytes was seen without formation of germinative centers (Figure 3). Immunohistochemistry was positive for CD20 and CD3 (Figure

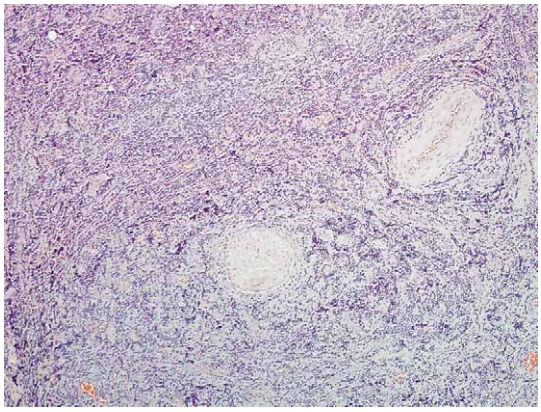

Fig. 3. Diffuse infiltration of perivascular and perineural spaces with lymphocytes and plasma cells (HE, magnification x100).

4). Given in consideration clinical presentation, pathohistological feature and immunohistochemical findings, the diagnosis of inflammatory pseudotumor was estab-

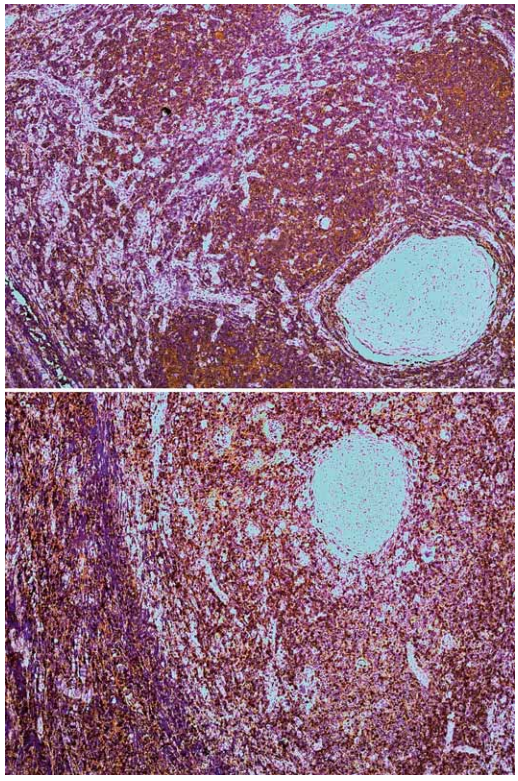

Fig. 4. Positive immunohistochemistry reaction to CD20 (above) and CD3 (below).

lished. The patient has been followed-up for six months so far, and no signs of reccurent disease appeared (Figure 5).

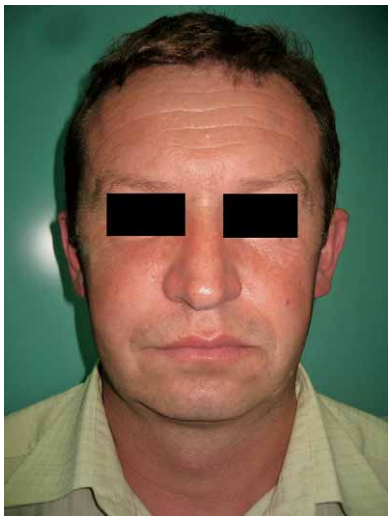

Fig. 5. Patient six months postoperatively with no relapse or recurrent disease.

Discussion and Conclusion

Clnical presentation of IPT is usually non-specific mimicking malignant tumors. Lesions are present for months or years, painless and locally destructive. Radiologic presentation is also not specific, typically an infiltrative homogenous mass is seen on CT.

There are various histologic presentations of IPT and that is the explanation for the different synonyms that were used. It is not a single entity but a group of lesions presenting histologically with inflammatory changes. Usually there is strong infiltration of tissue with myofibroblastic spindle cells, lymphocytes and plasma cells. Coffin et al.[15] described three basic histologic patterns: (1) myxoid, vascular, and inflammatory areas resembling nodular fascitis; (2) compact spindle cells with intermingled inflammatory cells resembling fibrous histiocytoma; and (3) dense platelike collagen resembling a desmoid or fibrous scar. In our case there was a predominant infiltration of small lymphocytes suggesting possible lymphoma, requiring further PCR analysis of B cell clonality.

The etiology and pathogenesis of IPT is still unknown. Some consider that it is an immunologic host response to many different stimuli, including infectious agents, microorganisms, adjacent necrotic tissue, neoplasms, foreign bodies, and some kinds of tissue injury[16]. Our patient did sustain an injury in a face after which the swelling appeared and never resolved supporting the theory of tissue trauma in etiology of IPT.

Treatment modalities of IPT consist of surgery, corticosteroid therapy and radiotherapy. Response rates to corticosteroid therapy are around 80% and usually rapid and satisfactory, although recurrences up to 20% after cessation of corticosteroid therapy have been reported[17].There is no standard dose and duration of corticosteroid therapy although initial dose of 60mg of prednisolone has been recommended[7]. Some advocate prolonged therapy to low dose prednisolone, up to six months to prevent recurrences or relapse[7].Surgery is in our opinion first choice for lesions which are easily accessible and resection will not cause significant morbidity (as was in our case). If this is not the case, first line treatment should be corticosteroid therapy. Lesions resistant to corticosteroid therapy or recurrences are amenable to surgical resection or even local radiotherapy[18] although it should be used cautiously because of radiation induced malignancies.

Diagnosis of IPT is diagnosis by exclusion, combining clinical, radiological and pathohistological characteristics. Lack of clear and certain histologic criteria makes differential diagnosis between IPT and spindle cell malignancies, extramedulary lymphoma, Wegeners granulomatosis and collagen vascular disease difficult[11].

Our case is unique regarding localization of head & neck IPT, no case presenting on the face in infraorbital region has been described in the literature. Although IPT is very rare in general and especially on the face, one should be aware of it when considering differential diagnosis of facial swelling.

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UPALNI PSEUDOTUMOR KOJI SE MANIFESTIRA KAO OTEKLINA LICA

S A Ž E T A K

Prikaz slučaja upalnog pseudotumora na licu kod pacijenta koji je primio slučajni udarac u desnu stranu lica. Nakon što oteklina od udarca nije splasnula pacijenta je pregledao stomatolog koji je izvadio zube sumnjajući na oteklinu odontogene etiologije. Oteklina je bila i dalje sve veća te je upućen na Kliniku za maksilofacijalnu kirurgiju. Napravljeni su CT lica i citološka punkcija lezije. CT je pokazao homogenu tumorsku masu infraorbitalno desno s prednje strane maksile. Po citološkoj punkciji radilo se o reaktivnoj hiperplaziji limfnog čvora. S obzirom da je lezija bila lako dostupna učinjena je kirurška eksploracija i ekstirpacija. Patološka analiza je govorila u prilog Non Hodgkin lifoma B stanica niskog stupnja. PCR-om je dokazana pak poliklonalnost B stanica odbacujući dijagnozu limfoma. Patohistološka revizija je pokazala difuznu intramuksularnu, perineuralnu i perivaskularnu infiltraciju malim limfocitima bez formiranja germinativnih centara. Imunohistokemija je bila pozitivna na CD20 i CD3. Uzimajući u obzir sve značajke postavljena je dijagnoza upalnog pseudotumora. Dijagnoza upalnog pseudotumora se postavlja na temelju isključenja, kombinirajući kliničke, radiološke i patohistološke karakteristike. Nedostatak jasnih histoloških kriterija čini diferencijalnu dijagnozu izuzetno teškom. Naš slučaj je jedinstven po lokalizaciji pseudotumora na glavi i vratu, nije još objavljen slučaj pseudotumora na licu u infraorbitalnoj regiji. Iako je općenito upalni pseudotumor rijedak a pogotovo na licu, treba ga imati na umu kada mislimo o diferencijalnoj dijagnozi otekline na licu.