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Case Report

Inflammatory pseudotumour of nasal cavity and sinuses: a case report with review of literature

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ABSTRACT

Inflammatory pseudotumor (IPT) is a benign non-neoplastic rare mass lesion clinically and radiologically simulating neoplasm thus generally requiring a biopsy to exclude malignancy. It occurs almost anywhere all over the body with most common locations being lungs and orbit with only 15% of it occurs in head and neck non-orbital region. Here we are describing a case of 52-year-old female patient having bleeding from left nostril with swelling, redness of left side of nose and face, initially painless. Radiological imaging was highly suspicious of malignancy. HPE showed inflammatory lesions. The fungal culture report of nasal specimen was showing 'no growth'. Blood examination was within normal limits; blood sugar levels was within normal limits. Systemic steroids were started which showed drastic improvements. Most of the IPTs in head and neck region shows a benign course, but cases related to paranasal sinuses seems to be of highly aggressive behaviour, with poor response to surgery, chemotherapy, radiotherapy; showing recurrences and fatal outcome. In such cases corticosteroid therapy generally shows good outcome. Tumors for which complete resection would result in morbidity may be treated with corticosteroids conservatively and close follow up. Understanding of natural history and pathophysiologic features of IPTs is helpful in differential diagnosis and may prevent unnecessary surgeries.

Keywords: Inflammatory pseudotumor, Inflammatory lesion, Steroid, Non-neoplastic, IPT

INTRODUCTION

Inflammatory pseudotumors (IPTs) are unencapsulated growth. They are well-circumscribed, quasi-neoplastic tumors of inflammatory cells with unregulated growth. In 1939 it was first described by Bunn. Umiker and Iverson coined the term 'inflammatory pseudotumor' in 1954. IPT might be secondary to autoimmune dysfunction, smoking, trauma, vasculitis, foreign bodies, fibroblastic proliferation, acute infection response and reparative process of post-inflammation.[1] Mycoplasmata, mycobacteria, *Rhodococcus equi*, *Klebsiella rhinoscleromatis*, nocardiae, actinomycetes, *Epstein-Barr virus*, Herpes simplex 8 have been implicated in pathologic specimens.[2,3] Interleukin 1 contributes to the systemic and local effects of IPT. On pathologic examination there is presence of spindle cells, plasma cells, lymphocytes. Fibroblasts, myofibroblasts, histiocytes, inflammatory infiltrate may also be seen. Lack of atypia/mitoses can rule out neoplasia.[4]

Lymphoid tissue in IPT consists of both B-cells and T-cells with plasma cells exhibiting polyclonal proliferation of kappa and lambda light chains, thus differentiating it from lymphoma or plasmacytoma.[4] Necrosis and mitotic figures are usually absent. In a literature review of 84 cases of IPTs, three basic patterns were recognised: (a) myxoid, vascular, inflammatory; (b) spindle cells with lymphocytes and plasma cells; (c) dense collagenous tissue resembling scar tissue.[2] Immunohistochemical exam of IP reveals positive stains for cytokeratin, CD20, smooth muscle actin, ALK, and increased IgG4-positive plasma

cells.[2] IP is usually positive for vimentin, EMA, CD138, CD20, CD68, and negative for S100 in immunohistochemical staining.[1]

Patients present with fever, weight loss, hypergammaglobulinemia, growth retardation, thrombocytosis, iron deficiency anemia, symptoms related to mass effect, or a combination of these. Differential diagnosis includes carcinoma, fibrodysplastic lesions, lymphoma, chronic fungal infection, Wegener's granulomatosis, extramedullary plasmacytoma.[5] Radiologic features of IPT are nonspecific and variable. They are not characteristics. It might because of the amount of cellular infiltration and fibrosis. Radiographically, extra-orbital IPT of head and neck region tends to be showing a poorly demarcated lesion with aggressive features that might include bone erosion, sclerosis, remodeling on CT scan.[3]

On MRI, the lesions are isointense on T1-weighted image and Hypointense on T2-weighted image.[3] T1-weighted images following gadolinium contrast typically reveals marked enhancement.[2] On ultrasound images, lesions can be hyperechoic or hypoechoic. It can be well-circumscribed or ill-defined borders. These lesions often have increased vascularity during color or power Doppler examinations. Contrast-enhanced MRI and CT may show a heterogeneous or homogenous lesion. Delayed imaging showing increased enhancement may be due to the presence of fibrosis.

CASE REPORT

A 52-year-old female patient, previously healthy, presented with complaints of initially non tender swelling and redness of nose and left side of face, difficulties in breathing and occasional bleeding from left nostril for last 3 months with associated upper lip swelling. Discharge present from the medial canthus of left eye of 3 months in duration. No associated history of photosensitivity and any drug hypersensitivity. There was also an associated healed ulcer over the left submandibular region. There was no history of rheumatological disease or any kind of prior radiation exposure. She had visited other hospital, where initial treatment with antibiotics did not relieve her symptoms.

Examination

The skin over the nose and adjacent nearby area extending towards left side of face including left cheek looked erythematous, swollen, non-tender to touch. Nasal endoscopy done on the day of admission revealed an ulcer proliferative reddish irregular mass at the level of inferior turbinate occupying the nasal cavity in the left side. It was a soft mass insensitive to touch, did not bleed on touch, but while taking biopsy from the tissue, there was bleeding. The mass was mostly attached to the lateral nasal wall. There were signs of left nasolacrimal duct obstruction with normal extraocular movement. No palpable cervical lymphadenopathy. Oral cavity examination was normal regarding teeth, gum, both soft and hard palate.

Investigations

Blood examination was within normal limits, blood sugar levels was within normal limits. The fungal culture report of nasal specimen was showing 'no growth' after 21 days of aerobic incubation at 25 and 37°C. CECT imaging of nasal cavity and paranasal sinuses showed mucosal thickening in bilateral frontal, bilateral ethmoidal, bilateral maxillary and bilateral sphenoidal sinuses; hypodense collection in left maxillary sinus and few ethmoid air cells, widening of left maxillary ostium. Erosion was noted in nasal septum, nasal bone and walls of ethmoid air cells. This picture was highly suspicious of malignancy. Repeat HPE after a course of antibiotics also showed only inflammatory cells. Histopathological examination of the biopsy taken from the sinonasal mass was showing to be that of 'inflammatory lesion'. PAS done for fungal elements came out to be negative.

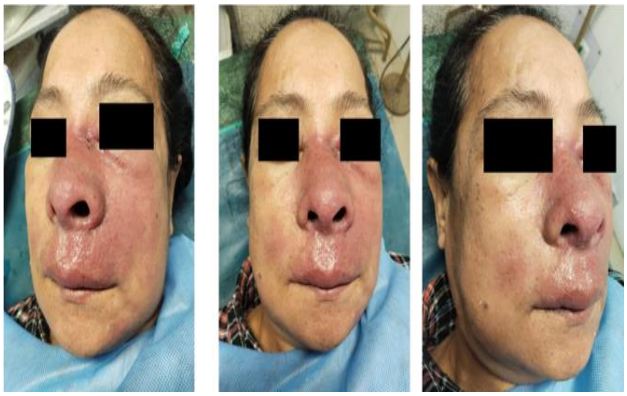

Figure 1: During treatment.

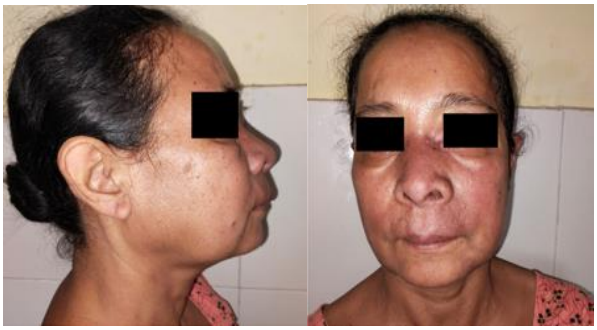

Figure 2: After treatment.

Treatment

IV antibiotics were tried of injection ceftriaxone+sulbactam initially followed by IV antibiotics of infusion metrogyl. Later on oral prednisolone (wysolone) was tried which showed drastic improvement. Prednisolone dispersible tablet 20 mg was given thrice daily in tapered doses and continued. Patient was followed

up and since last 3 three months, there was no signs of recurrence.

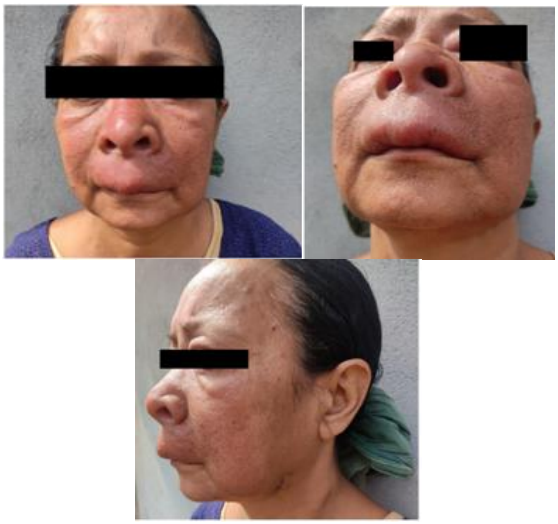

Figure 3: Review phase.

DISCUSSION

Study showed a bimodal peak distribution of IPT in the third and fifth decades of life having higher occurrence with female preponderance with male to female ration of 1: 2.6.[6] Treatments for IP in the nasal cavity and sinuses depend on the general condition of the patient, tumor location and biological behavior. Treatment includes corticosteroids, surgical resection or both. At least six months of maintenance- dose corticosteroids are recommended to prevent recurrence of disease.[2] Patients unresponsive to systemic corticosteroids may benefit from intralesional steroids.[2] 'A favourable response with corticosteroids is seen especially in IPs that that are predominated by plasma cells and lymohocytes.'[2] Corticosteroid therapy usually yields a favorable response in early lesions that include lymphoid follicles; while mature lesions responds less to it with more fibrous content.[7] Alternative treatments include Igs, radiotherapy, small molecule inhibitors.[2]

Complete resection was found to be the most effective surgical therapeutic option.[5] Recurrences after surgical excision of head and neck IPT approaches 10 to 20%.[2] When such resection is not feasible, local radiotherapy may be tried. Radiotherapy may be used in patients who respond poorly to steroid therapy, have medical contraindications to prolonged use of high-dose corticosteroids or relapse when steroid tapered. While corticosteroid therapy can significantly shrink the tumor size, but there are not enough follow-up data yet so as to confirm its efficacy.[1]

CONCLUSION

IPT is a diagnosis of exclusion, after eliminating the possibilities of benign and malignant neoplasia, collagen disease, vasculitis, infection and other inflammatory diseases. IPTs almost always erodes the surrounding bone and requires active treatment. Based on the site of lesion and potential for functional impairment, treatment of IPT can be divided into medical and surgical modalities. Surgery could be advocated only if it can be performed with acceptable cosmetic and functional morbidity. Resection should be considered when alternative noninvasive treatments have failed; and if the mass can be resected with minimal morbidity; if the diagnosis cannot be established by FNA and core biopsy alone. Multiple biopsies are often required to establish a diagnosis of IPT. In this case, IPT showed good response to systemic steroid after controlling associated secondary infection and inflammation.

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Ethical approval: Not required

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