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RESEARCH ARTICLE

INFLAMMATORY MYOFIBROBLASTIC TUMOUR: CASE SERIES OF 12 PATIENTS IN RURAL HOSPITAL OF CENTRAL INDIA

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ABSTRACT

Background: Inflammatory myofibroblastic tumour is rare and benign type of soft tissue neoplasm. It is proliferative type of lesion which mimics like a malignancy due to its unknown etiology clinically, radiographically and histopathologically. It consists of myofibroblastic proliferation along with various types of inflammatory infiltrates. The diagnosis is made based on clinical, radiological, histopathological and Immunohistochemistry (IHC) findings which differentiate it from other soft tissue tumours. Excision method is used in all patient as treatment part.***Objective****:* To characterize the clinical presentation, radiological presentation, diagnosis by histopathology and IHC for the treatment of inflammatory myofibroblastic tumour. ***Materials and methods****:* This is an observational retrospective study based on a case series. Data was collected after reviewing the medical records of patients from HIS diagnosed with inflammatory myofibroblastic tumour at our rural hospital in Sewagram between January 2010 and July 2023. The data included age, sex, symptoms, location, size, USG/CT, Histopathology /IHC, surgical approach and follow up. ***Results****:* There were 12 patients diagnosed with inflammatory myofibroblastic tumour. Their mean age was 47.5 years, out of 12 patients, 09 were women and 03 were men. ***Conclusions****:* Inflammatory myofibroblastic tumour is very challenging for establishing the diagnosis due to its varied clinical, radiological and histopathological presentation. IMT can confuse with malignant tumours. Diagnosis depends on their histopathological and IHC findings of patient while treatment part on the size and location of tumour. Treatment options vary patient to patient, ranging from surgical resection from minimum endoscopy procedure to open surgery.

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INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is rare, benign soft tissue lesion which has characteristic aggressive proliferation.[1,2,3]. It has other various names like inflammatory myofibroblastic proliferation, plasma cell granuloma and inflammatory pseudotumor, fibrous xanthoma, pseudosarcoma, lymphoid hamartoma, myxoid hamartoma.[2,5] Most common site of involvement includes lung followed by liver, orbit, retroperitoneum, extremities.[1,2,3]In Head and neck region involvement is rare.[3,5] In this region it represents 14-18% of all extrapulmonary IMTs including epiglottis, maxillary sinus, submandibular region, oral cavity. [1,3] In oral cavity various location like gingiva, tongue, hard palate, mandible involved.[1]They usually affect children and young adults. [1] Clinically presented as firm, painless mass, swelling of short duration or following specific symptoms of that site.[1] The etiology most of the cases remain unknown but various initiating factors such as reactive, infection, autoimmune and neoplastic.[1] Here we present case series of 12 patients of IMT and brief discussion about their clinical, radiographic and histopathological features in our rural hospital.

MATERIAL AND METHOD

It is a retrospective study for which information was collected from medical record of our HIS system with diagnosed cases of inflammatory myofibroblastic tumor in our institute from January 2010 to July 2023.In which clinical, histopathological, demographic and radiological characteristic were recorded. Informed consent from the patient is not taken as we are collecting only data from our archives. Treatments included open surgery, laparoscopic management, and perioperative chemotherapy in some cases. Data also include symptoms, size, location, surgical management, type of management.

No ethical issue is there because data is collected only from our HIS system.

Data availability-The patients of this study did not give written consent for their data to be shared publicly hence research supporting data is not available.

RESULTS

In this case series we find 12 cases of inflammatory myofibroblastic tumour from our HIS system.From January 2010 to July 2023, 12 cases of patient with diagnosis of inflammatory myofibroblastic tumors were found. Median age of presentation is 32 years.

Out of 12 patients 9 are female and 3 are male patient. Most common location is abdomen consist of 5 cases. Thumb, cheek, back, lid, knee, neck, parotid each show one case of myofibroblastic tumour. Presentation of tumor was swelling in 6 patient and 6 patients presented with pain in region of swelling. Total 8 patient undergoes radiological examination either USG/CT scan show mass which is either well circumscribed /irregular with neoplastic etiology was given[figure2]. Concerning the tumor size, the largest measuring is 14.5x 12.5x 9.5 cm and smallest measuring 1.2 x 0.8 x 0.5 cm[Gross image at figure 1].Histological and immunohistochemical characteristic.With regards to histopathological distribution one patient show mammary type of myofibroblastic tumor[Figure 4 and 5].

1. Gross image

Figure 1. Gross image of Inflammatory Myofibroblastic Tumor of case no.4 which is well circumscribed grey white solid tumor mass with size of about 12×8 ×5cm. Cut surface was firm grey white without any areas of hemorrhage and necrosis

2. Radiological image

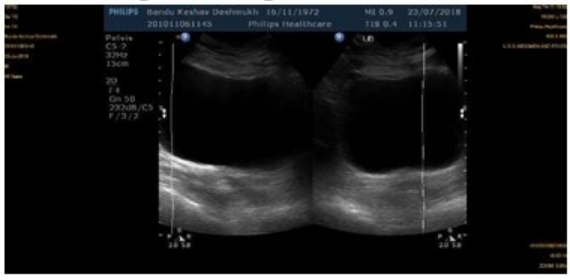

Figure 2. USG abdomen and pelvis show image of patient no.4 which show a large hypoechoic mass lesion with well-defined borders approximately size 13 cm×8.4×7.9cm

3. Radiological image

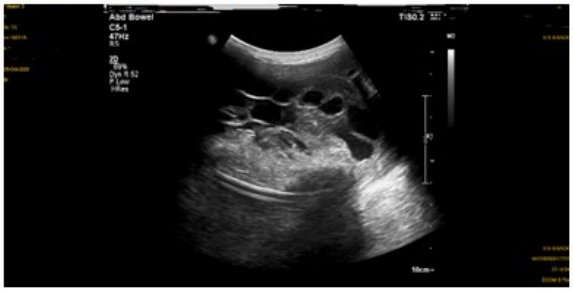

Figure 3. USG back show image of case no.3 patient which well circumscribed hypoechoic mass lesion which shows multiple linear septations? neoplastic etiology

4. Histopathology image

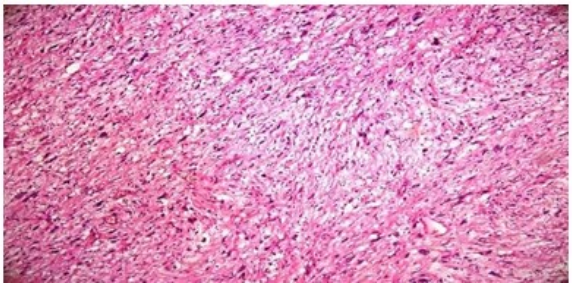

Figure 4.Histopathology image of case no.4 which show low power view of H and E slides in which it shows spindle cells in loose fascicles

5. Histopathology image

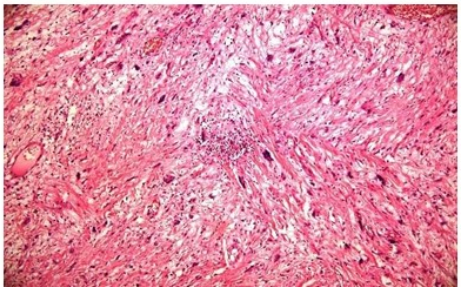

Figure no. 5. H and E slide of patient no.4 which shows spindle cells with chronic inflammatory infiltrate all over the slides and bizarre type of cells.

6. Histopathology image

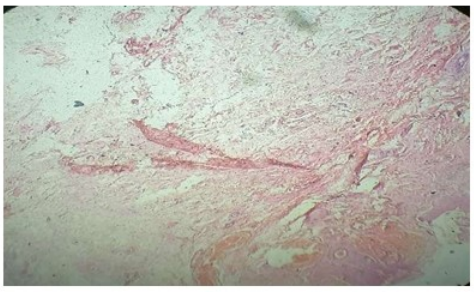

Fig.6. H and E slide of case no.3 which show spindle cells in loose fascicles with inflammatory cell infiltrate.

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SN	Age	Symptoms/complaint	Size	Location	Surgery	Histopathology report	IHC	USG/CT	Follow up
1	28 /M	Swelling over right thumb since2 month	2.5x2x1cm.	Thumb	Excision	Inflammatory myofibroblastic tumor of soft tissue.	Not done	Not done	NA
2	66/M	Swelling over left side of cheek since 1 year.	size 3.8x2.5x2 cm.	Cheek	Wide excision.	Section from tumour show squamous epithelium underlying tissue show proliferation of spindle cell. Involving soft tissue with underlying muscle, intense infiltration of lymphoplasmacytic infiltration with myofibroblasticproliferation.Inflammatorymyofibroblastic tumor of soft tissue.	SMA and Vimentin positive. Desmin is negative consistent with IMT	CECT Neck- Neoplastic lesion in left lower gingivobuccal sulcus and left buccal mucosa	NA
3	73/M	Swelling over back since 2 month.	1.5x1x 0.5cm.	back	Wide local excision	Inflammatory Pseudo tumor with intense inflammatory infiltrate of plasma cells, eosinophils and lymphocytes interspersed with marked fibroblastic proliferation.	Not done	USG back shows wellcircumscribed hypoechoic mass lesion which shows multiple linear septation? neoplastic etiology	NA
4	50/M	Pain in left lumber region of abdomen since 4 month	12×8 ×5cm	abdomen	Exploratory Laparotomy	Section shows spindle cell tumour. At places arranged in loose fascicles. There is infiltration of chronic inflammatory cells at few places. There are many pleomorphic cells. Some of them are bizarre. Necrosis and haemorrhage are not seen. Histopathological features are consistent with inflammatory myofibroblastic tumour. Immunohistochemistry. The tumour cells are positive for Desmin, SMA and CD34. Mammary type of myofibroblastoma in soft tissue.	The tumour cells are positive for Desmin, SMA and CD34. Mammary type of myofibroblastoma.	USG Abdomen and Pelvis- Heterogeneously echotextured lesion in the pelvis suggestive of? neoplastic etiology. CT Abdomen and Pelvis reveals - Hypodense mass lesion in the region of rectovesical pouch? neoplastic etiology	NA
5	70/F	Painless swelling over upper lids since 1 year.	3.5x2x1 cm.	lid	Excision	Inflammatory pseudotumor.	Not done	CECT orbit- Multiple glandular and lymphoid enlargement suggestive of inflammatory/neoplastic etiology.Histopathological correlation.	NA
6	67/M	Pain in abdomen since 2 month and pain in left lumber region.	1.2 x0.8x0.5 cm.	Lumber region	Exploratory laparotomy	Inflammatory myofibroblastic tumor	Not done	CECT Abdomen and Pelvis Heterogeneously enhancing irregular hypodense mesenteric masses with desmoplastic reaction and retraction of adjacent bowel loops, possibility of carcinoid tumour should be considered.	NA
7	32/M	Pain and mass in left hypochondriac region since 2 month	14.5 x 12.5 x 9.5 cm.	abdomen	Exploratory laparotomy	Inflammatory myofibroblastic tumor	Not done	USG Abdomen and Pelvis Hypoechoic mass in left lumbar region likely to be malignant left renal mass.	NA
8	54/M	Left parotid swelling since 2 month.	1.5 cm	parotid	Excision	Inflammatory pseudotumor	Not done	Not done	NA
9	36/M	Painin central abdomen since 1-2 month.	5.5 x 5 x 4 cm.	abdomen	Exploratory laparotomy.	Inflammatory myofibroblastic tumour of stomach	Not done	USG Abdomen and Pelvis- Hypoechoic round mass in epigastric region? neoplastic etiology.	NA
10	32/F	Pain in abdomen since 5 month and mass on left hypochondriac region extending to iliac region.	14.5 x 12.5 x 9.5 cm.	abdomen	Exploratorylaparotomy.	Inflammatory myofibroblastic tumour	Not done	CT abdomen- Dysplastic left kidney with compensatory hypertrophy of right kidney	NA
11	30/M	Swelling over right side of neck since 5 months.	2cm diameter	Neck	excision	Pseudoinflammatory tumour	Not done	Not done	NA
12	20/F	Pain andswelling over bilateral knee joint since last 5 month.	1.5 x0.3x0.2cm	knee	Excision	Inflammatory myofibroblastic tumour	Not done	Not done	NA

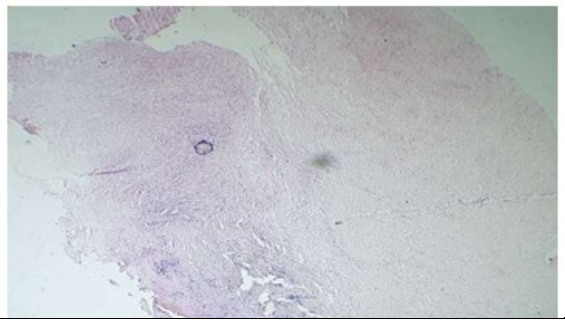

Figure 7-H and E slide of case no.2 patient low power view of spindle cell arranges in loose fascicles with chronic inflammatory infiltrate

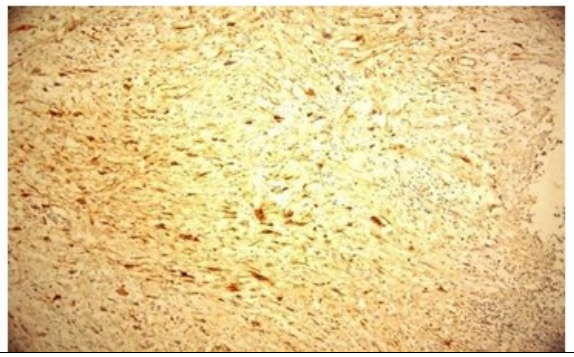

Figure 8 –IHC slide of case no.4 patient which show Desmin marker which shows positivity for spindle cells

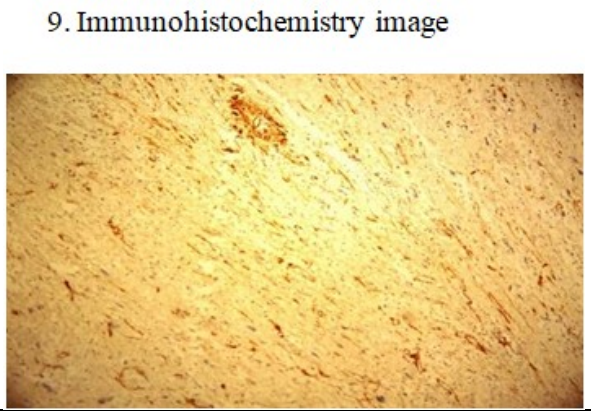

Figure 9 - IHC slide of case no.4 which shows SMA marker which shows positivity for spindle cell

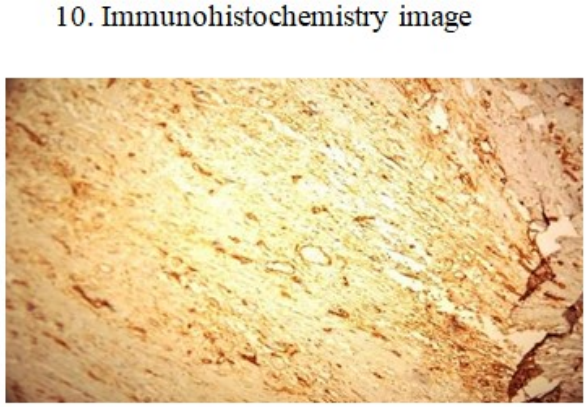

Figure 10-IHC slide of case no.4 shows CD34 marker which shows positivity

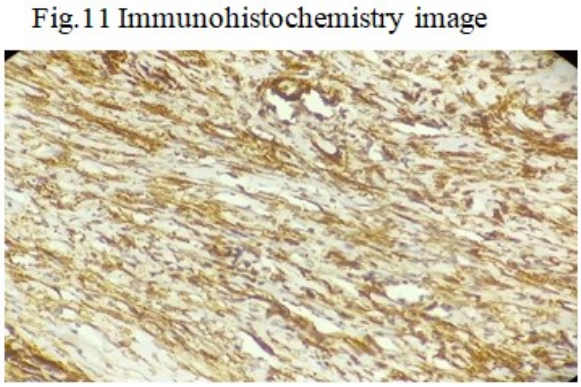

Figure 11-IHC slide of case no.2 shows SMA marker which shows positivity for spindle cell

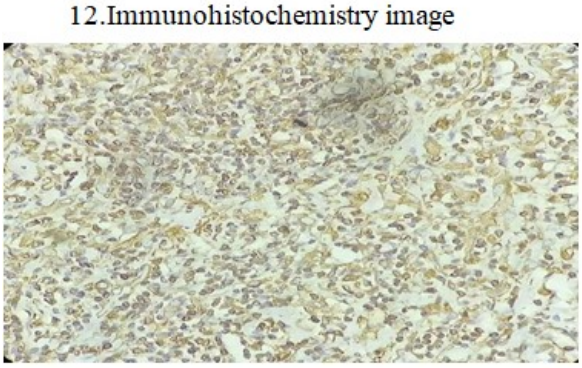

Figure 12-IHC of case no.2 in which Vimentin marker shows positivity

Out of twelve cases Immunohistochemistry was done for two patients in which SMA, vimentin, CD34, Desmin was done for the patient. One patient show positivity for SMA, Vimentin and Non positivity for Desmin[figure11 and 12].One patient show positivity for SMA, Desmin and CD34[figure 8, 9 and 10]. All the twelve patients received excision method for the removal of tumour. In follow up there is no patient came with recurrence of tumour.

DISCUSSION

Inflammatory myofibroblastic tumor (IMT) is benign and rare soft tissue lesion.[3]It has various names like inflammatory pseudotumour, plasma cell granuloma, inflammatory myofibrohistiocytic proliferation.[2]

In history it was 1st described in 1905 by Birch-Hirschfield and later by Bunn in 1939 when he observed in lung. [1]Umiker*et al* named as IMT as it was similar to malignant neoplasm clinically, radiologically and histopathologically.[1] Pathogenesis of inflammatory myofibroblastic tumor is remains unknown but various initiating factors have been proposed such as ALK gene rearrangement, reactive, infectious viruses like Epstein-Barr virus and HHV-8, Trauma, chronic inflammation, autoimmune and neoplastic etiology.[1,2] IMT of head and neck region are generally benign and treated by radical excision, steroids, irradiation and chemotherapy. [1,3,5] Latest treatment modality is CO_2 laser. [1]This tumour is classified as tumour of intermediate biological behaviour so continuous follow up is needed in this patient.[5] Due to varied clinical and radiological manifestation it often mimics malignancy so establishing the diagnosis of inflammatory

myofibroblastic tumor is very challenging.[1] IMT grabbed attention of many pathologists, surgeon and oncologist due to its peculiar clinical behavior, histopathological response and therapeutic response.[2]Yuksel C *et al* shows a case series study of 17 patient of IMT in which 70% cases are female patient which is similar to our study in which 9 patients are female and also female predominance is noted by MA Z,TIAN X *et al*. [7,9,10]Cantera J *et al* and MA Z, TIAN X *et al*shows in his study that maximum cases are in head and neck region mostly in orbit while we have only one case around orbit area.[8,9] MA Z, TIAN X *et al* studies shows most of the cases in lung region while in our study it is in abdomen region.[9]Yuksel C *et al* and Mahipathy S, Durairaj A *et al* shows that patient is presented with painful swelling while in our study most 6 patients are presented with pain at the lesion. [7,1]Ultrasound, MRI and CT scans are helpful up to certain extent for the diagnosis of IMT which is similar to the MA Z, TIAN X *et al* study.[9]Alhumaid H, Bukhari M *et al* studies case series of myofibroblastic tumour patient which on microscopy shows spindle cell with lymphoplasmacytic infiltrate similar to our study.[10]Also same study shows IHC positivity for SMA and Vimentin which is consistent with our study while it also shows positivity in Desmin which is not consistent with our study.[10]

IMT is manifested by nonspecific and variable symptomology depending upon the localization of the tumour.[6] Inflammatory myofibroblastic tumor usually show indolent course but in some casesit shows extensive invasion, recurrence and metastasis.[2] IMT shows 25% cases recurrence and distant metastasis in less than 2% of cases.[6] IMT differential diagnosis includes benign fibrous histiocytoma, fibrosarcoma, leiomyosarcoma, nodular fasciitis, solitary fibrous tumour, benign fibrous histiocytoma.[5]On immunohistochemistry spindle cell show reactivity to vimentin, Desmin, Smooth muscle actin and muscle specific actin.[4,5] IMT are classically negative for myoglobin and S100 protein.[1]According to WHO there are three variant of IMT 1)Loosely organized myofibroblast in an edematous myxoid background with plasma cell, lymphocytes, eosinophil and blood vessels resembling nodular fasciitis. 2) Collagen sheets with scattered plasma cell and eosinophils resembling a scar or desmoid tumour.3)Dense aggregates of spindle cell arrayed in a variable myxoid and collagenized background and admixed with a distinctive inflammatory infiltrate, diffuse clusters of plasma cell and lymphoid nodules resembling fibrous histiocytoma or fibromatosis.[1]

CONCLUSION

IMTs are low-grade rare benign tumours which show variable clinical and radiological presentation. Only histological and IHC features can confirm the diagnosis which reveals characteristic features such as fascicles of spindle cells in an inflammatory cell background and IHC.

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Conflicts of interest: There are no conflicts of interest.

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