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Solitary Eosinophilic Granuloma of Rib: A Case Report and Literature Review

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Abstract: ***A 10 year old child presented with pain and swelling in the right chest wall since last 2 months. Having no local sign of inflammation x-ray showed a osteolytic lesion in the shaft of 9th rib with thinning of cortex but no sign of pathological fracture or periosteal reaction.MRI of chest show T2 hyper intense and intermediate intense T1 lesion in the 9th rib. Blood investigations were all normal. Clinical and radiological features are non-confirmatory and produce dilemma by giving a basket full of different provisional diagnosis. We have no option other than to go for a biopsy. Biopsy of lesion was consistent with the features of Eosinophilic granuloma. After confirming the diagnosis from histopathology study possible involvement of other body part was ruled out by radiology. On 2 month follow-up, chest x-ray showed the lesion was found to be healing spontaneously with signs of reossificatrion. At 5 months follow X-ray lesion was completely healed radio logically and no symptoms. Patient was follow up for 9 months regularly and there is no recurrence of lesion or any new other site involvement an the child was asymptomatic. A review of available literature was done regarding the various modalities of treatment available for solitary eosinophilic granuloma and their results.***

Keywords: solitray eosinophilic granula,rib, spontaneous healing, dillema of diagnosis, how to treat.

1. Introduction

Eosinophilic granuloma (EOG) represents the localized form of Histiocytosis x,where as disseminated form of Histiocytosis x being known as Litterer - siwe (acute) and Hand-schuller Christian disease (subacute) .[1]

Eosinophilic granuloma is first described as a distinct entity by Lichtenstein and Jaffee in 1940. [2]

Though represent different manifestation of similar pathological process , no clear cut separation line exist between three forms of above said disease causing confusion of classification of a particular case.

Eosinophilic granuloma usually a disease of children and young adults .it is rare in persons over 30yrs age and 75 percents are solitary bone lesion and more than 50% cases involve the skull, ribs and femur.[3]

Biopsy is a must for making diagnosis confirmatory. It shows a fibrous stroma with histiocytes,giant cells admixed with eosinophils and occasional foam cells.[4,5]

Wide varieties of treatments described in literature including supervised neglect, local intralesional steroid injection, curettage with radiataion,curettage bone grafting, radiation therapy alone, chemotherapy and cryosurgery. [6-13 ,32] We report a case of solitary eosinophilic granuloma of rib in 10 year child that healed rapidly after biopsy without any intervention. The consent of parents obtained after due explanation that their child's case may be used as a learning resourse.

2. Case Report

A 10 year old child presented with pain in the right chest wall since last 2 months. Pain was insidious onset, dull aching and gradually increasing in intensity and no relation to activity and subsides on taking analgesics. On examination a swelling of size 3 × 1.5 cm found on lateral aspect of his chest wall, On palpation swelling arising from underlying rib and was mild tender. There is no local sign of inflammation like redness or increase in local temperature. On x-ray there is a osteolytic lesion in the shaft of 9th rib with thinning of cortex but no sign of pathological fracture or periosteal reaction.MRI of chest show T2 hyper intense and intermediate intense T1 lesion in the 9th rib. Bilateral lung and rest of the chest wall is normal on MRI. Blood investigations show no abnormality Biopsy of lesion done. Biopsy show features like plenty of giant cell histiocytic type which are multinucleated, histiocytes with bland looking grooved and clefted nuclei in a back ground flooded with eosinophils and lymphocyte. After confirming the diagnosis from histopathology study x-ray of skull ,pelvis and long bones of child was done and no lesion at found at any other site. child was kept on follow up and advised to come for follow-up after 1 month but patient was lost to follow-up and came to us after 2 month. On repeating chest x-ray the lesion was found to be healing spontaneously with signs of reossificatrion. At 5 months follow X-ray lesion was completely healed radio logically and no symptoms. Patient was follow up for 9 months regularly and there is no recurrence of lesion or any new other site involvement.

3. Discussion

Solitary eosinophilic granuloma is a challenge, both in aspect of diagnosis and treatment. Though solitary eosinophilic granuloma of rib is not much rare but there is only handful of case reports in literature. Patients usually children and young adults ,presented to physician with a history of pain and swelling over bone with localized tenderness. Presentation some time may be complicated with mild fever, incrased TLC, eosinophilia, and laboratory investigation may show increased ESR.[14] In plain radio - graph it usually appear as round/oval osteolytic lesion with sharp border and punched out appearance's /MRI needed for better delineation of lesion and to know it's extra skeletal extension.[15]

A wide variety of lesion may come into consideration as differential diagnosis of eosinophilic granuloma of rib are lymphoma, leukemia, Ewing sarcoma, metastasis, myeloma, fibrous dysplasia , GCT, osteomyelitis and heamangioma.[14-19]

The case of ours created a dilemma for diagnosis as clinical, radiologic and MRI features are inconclusive. To reach a final diagnosis we have no other option left than to go for a biopsy. Further confirmation can be done by IHC staining by cd1 or s100 [29] or by using electron microscopy[5, 28.]

Reaching at a final diagnosis is no way decrease our curiosity, because the treatment part is more confusing. Searching the literature we found a wide variety of treatment modalities, all claming its own success rate. In the table below we have summarized the results of different studies available in literature.

	No of cases	age	Treatment method	Results
Kelley et al	9	All cases age < 14 yrs	Curettage only - 6 Curettage + RT - 2 Curettage + BG - 1	All healed
Hunter T	8	Skeletally mature 3	Excision - 3 Biopsy - 3 Biopsy + RT- 3	All healed, no recurrence at 1 yr
F. Plasschaert et al	32	Skeletally mature15 Skeletally immature17	Skeletally immature – Biopsy – 6 Biopsy + Curettage +/- BG – 11 Skeletally Mature Biopsy + Curettage +/- BG – 15	No recurrence in skeletally immature group. 4 recurrence in skeletally mature group
ALAN W. YASKO et al	35		Intralesional steroid	All healed except one (needed BG)
Capanna R et al	11	All < 16 yrs	Intralesional steroid	All healed
SALVATORE SESA et al	19 solitary	All ,16yrs	Observation - 6 Curettage- 4 Curettage+ BG - 4 Cast- 3 Excision +RT+CT -1 Curettage+IF+RT -1	Complete response- 14 Partial response- 2 No response-3 Recureence- 3
Cohen M et al	*9*		*Intralesional steroid*	*8 healed* *No complication*

BG :BONE GRAFT RT: RADIO THERAPY CT : CHEMOTHERAPY IF: INTERNAL FIXATION

How does intralsional steroid act in healing of eosinophilic granuloma is not yet fully understood. Apart from above studies, some of other interesting studies on treatment of eosinophilic granuloma also described in literature. Han et al in their study on 33 patients of localized LCH of bone, compared the efficacy of anticancer therapy and versus indomethacin treatment. They found there is no significant difference between time for radiological healing and for functional recovery between two treatment group though more complication noticed in patients treated by chemo and surgery.[30] Indomethacin act by inhibiting PGE2 synthesis by LCH cell.[31]

4. Conclusion

To conclude from above discussion that it is very difficult to find a straight approach to reach the final diagnosis and to treat successfully a case of eosinophilic granulomaof bone in children. Here we are proposing a step wise approach that may be considered for diagnosis and treatment of eosinophilic granuloma of bone.

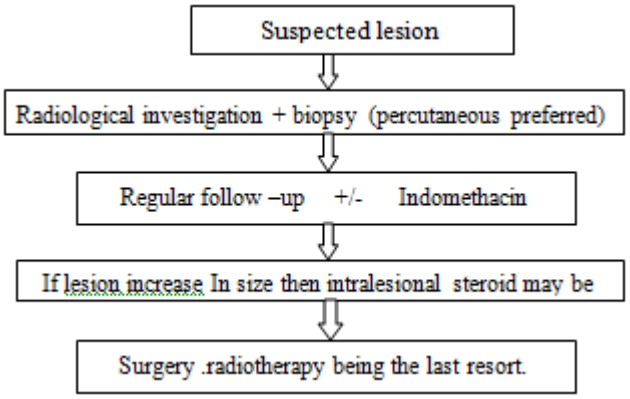

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Illustration Details

1) Posterio- Anterior view of chest X-Ray showing a osteolytic lesion in the shaft of 9th rib with thinning of cortex but no sign of pathological fracture or periosteal reaction.
2) MRI of chest showing Intermediate intense T1 lesion in the 9th rib.Bilateral lung and rest of the chest wall is normal on mri.
3) Histopathology microscopic photograph showing features like plenty of giant cell histiocytic type which are multinucleated, histiocytes with bland looking grooved and clefted nuclei in a back ground flooded with eosinophils and lymphocyte.
4) Posterio- Anterior view of chest X-Ray at 2 month follow-up showing signs of healing and re-ossification.