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Combined management of Psammomatoid Juvenile Ossifying Fibroma and Dentigerous Cyst in a 7 year male: A case report

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Abstract:

Psammomatoid juvenile ossifying fibroma (PJOF) is a rare uncommon, benign fibroosseous variant of ossifying fibroma. It usually affects paranasal sinuses, orbit & skull. Although a benign entity juvenile ossifying fibroma is known to be locally aggressive & has a high tendency to recur. To aim of this paper to report a case of extensive PJOF with Dentigerous cyst in which complete excision of tumour and cyst enucleation was done by intraoral surgical approach. A 7-year-old male patient presented with mass on left side of middle third of face since last 2 years. In Left side cheek he noticed fullness & experienced pressure with no other dental or ophthalmic symptoms. After clinical, radiological & histopathological examination the diagnosis was confirmed as PJOF with dentigerous cyst. Intraoral surgical approach was performed and complete excision of tumour and cyst enucleation was done under GA. Good cicatrisation soft he intraoral wound were evident. He was on follow-up since last 1year and no sign of recurrence or any complication.

Key words: Jaw neoplasm, oral and maxillofacial surgery, juvenile ossifying fibroma, psammomas.

Introduction:

Juvenile ossifying fibroma (JOF) is an uncommon, benign fibro-osseous anomaly that impacts the craniofacial structure of young individuals. It constitutes a distinct clinical and histopathological condition, frequently mistaken for malignant growths [1]. JOF typically manifests in early life, with the majority of cases diagnosed during the first or second decade of a patient's life. It originates from periodontal ligament. It constitutes 2% of all oral tumours in children. It has an equal predilection for males and females. JOF commonly occurs in the facial bones (85%), calvarium (12%) and mandibular region (10%). Very rarely it has been reported extracranially (3%)[2]. The psammomatoid juvenile ossifying fibroma (PJOF) primarily affects the orbital bones and paranasal sinuses, whereas the trabecular variant often involves the jawbones [3].

The PJOF are unique lesions that occur commonly in children. Psammoma-like bodies are the hallmark of this neoplasm. Mandibular lesions are infrequent and may be confused with odontogenic cysts. It is essential to correlate clinical, radiographic, and histopathological

findings to ensure an accurate diagnosis [4].

CASE PRESENTATION

A 7-year-old male patient reported to Dept of oral and maxillofacial surgery outpatient department with the chief complaint of painless swelling over the front left side of upper jaw and asymmetry of face since last two year. Patient was apparently alright before 2 years, then he noticed swelling over left side of anterior maxilla which gradually increased to the present size, and the patient did not have any functional problems. In Left side cheek he noticed fullness & experienced pressure with no other dental or ophthalmic symptoms. There was no history of trauma, pain, any discharge and extraction of any teeth. Extra-oral examination revealed 3x3cm swelling on left anterior maxilla, extending antero-posteriorly left side ala of nose to zygomatic arch. (Fig-1) Intra-oral examination revealed Swelling was single, diffuse, round to oval in shape and approximately 5x4 cm in size. (Fig-2)Palpation revealed a firm, diffuse, non-tender, non-pulsatile swelling which was fixated to the underlying structures, no paresthesia of upper lip or restricted mouth opening. There was palpable buccal expansion. Intraorally, there was the obliteration of the vestibule from 62 to 26 region. No pus discharge or any inflammatory fluid discharge was present.

Patient was undergoing dental treatment outside pedodontics since last 1 year and there was biopsy and radiographic investigation (CBCT and OPG) was done which suggestive of dentigerous cyst of left anterior maxilla with impacted permanent canine tooth.

Routine blood investigation revealed normal study. Aspiration biopsy was performed to rule out arterio-venous malformation, cystic lesion, and fibro-osseous lesion. On aspiration nothing was revealed. Patient was advice orthopantomogram and 3D CT FACE. CT FACE suggestive of 30+25+26mm sized well defined circumscribed lesion with expansion of ground glass attenuation of maxilla on left side and 10+5mm well defined circumscribed cystic lesion present in left maxillary sinus with impacted canine tooth in cystic lesion. (Fig-3) Based on clinical and radiological findings differential diagnosis of Ameloblastoma, odontogenic keratocyst and adenomatoid odontogenic tumor were considered. Under local anesthesia incisional biopsy was performed intraorally. Histopathology analysis confirmed diagnosis of psammomatoid juvenile ossifying fibroma. After clinical,radiographic and histopathologic examination confirmatory psammomatoid ossifying fibroma with Dentigerous cyst was diagnosed. In view of the diagnosis and well-defined and non-invasive aspect, the intraoral surgical approach for complete excision of tumor and cyst enucleatio nunder GA chosen.

Patient was operated under general anesthesia with nasotracheal intubation. Intraoral incision was given from extending from mesial to deciduous central incisor to permanent 1stmolar region. (Fig-4A) Intra-operatively a well differentiated, bony, protruding, capsule fibroma was detected attach to anterior maxillary wall.(Fig-4B) Complete excision was performed using a drill and micromotor with 1cm of surrounding normal bone. (Fig-5A)Impacted canine tooth with cystic lining is removed from maxillary sinus. (FIG-5B) Excised specimen of size 4+3cm sent for histopathology and confirmation of safe margin (FIG-6A &6B). Sinus curettage was done. Peripheral ostectomy was done (FIG-7A). A defect was closed in layers with 3-0and 4-0vicryl sutures (FIG-7B).

Histopathological examination showed predominantly fibro-osseous proliferation composed of ossicles admixed with fibrous stroma and show presence of calcified psammomatoid bodies and osteoblastic rimming, the intervening fibrous stroma showed bland round to polyhedral to spindle cell arranged in fascicular and stortiform pattern, confirming the diagnosis of a Psammomatoid Ossifying Fibroma.(Fig-8)

Patient currently with 1-year postoperative follow-up and has good cicatrization of intraoral

surgical wounds. Extraoral examination showed harmonic projection of the facial middle third. There were no signs of recurrence during follow-up and the patient chose not to perform surgery for the reconstruction of residual defects after tumour excision.

FIG-1: front and lateral view of extra images shows a left swelling over anterior maxilla and facial asymmetry.

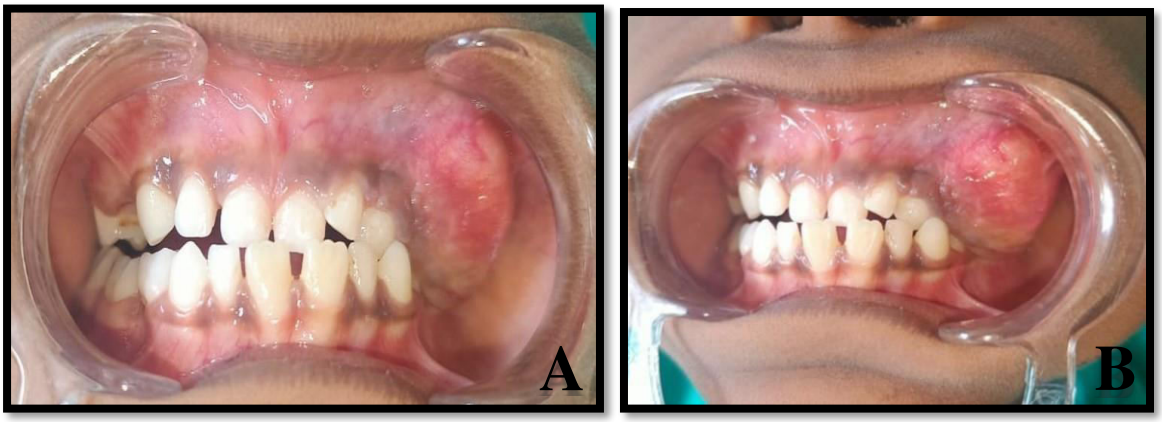

FIG 2: Intraoral images show a diffuse swelling in left maxillary anterior vestibule

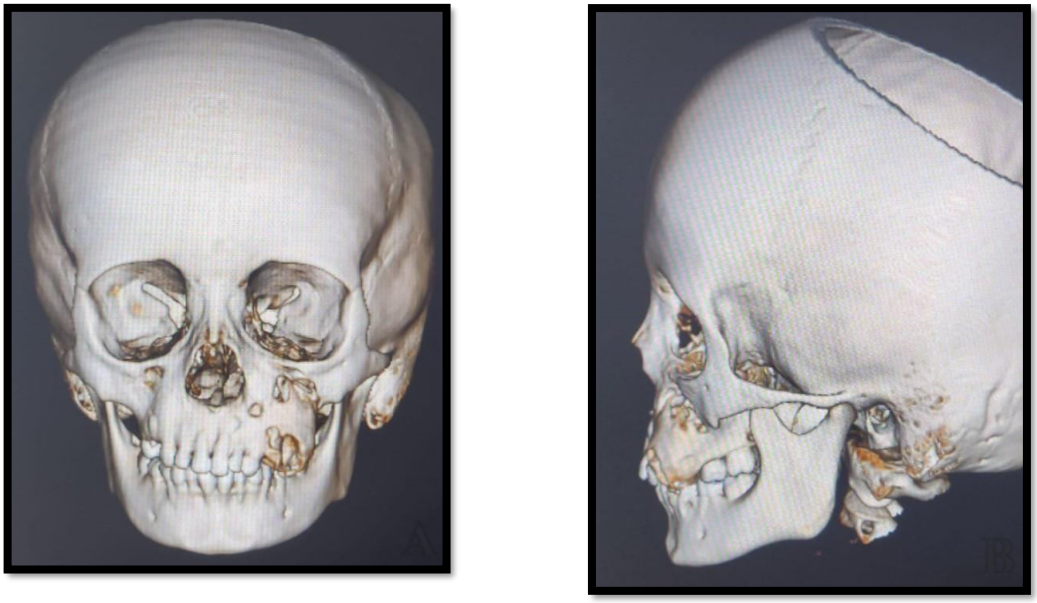

FIG-3: Computed images shows a bony swelling over left anterior maxilla.

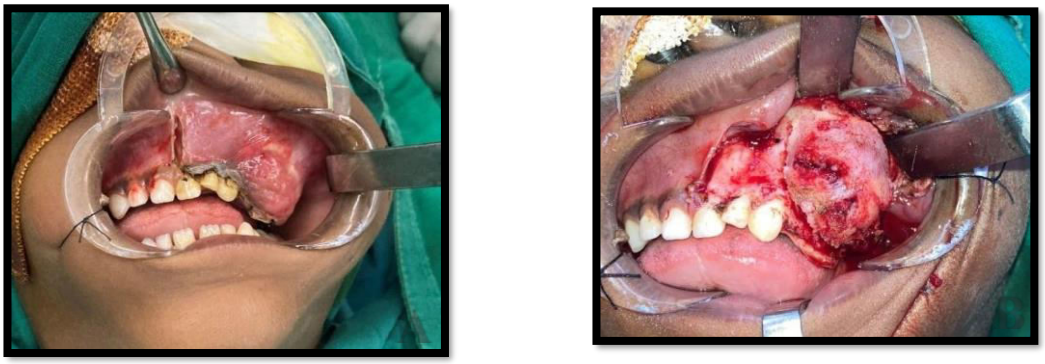

FIG-4: (A) suggest an incision marking extending from mesial of left deciduous central incisor to permanent first molar region (B) suggest an exposed tumour mass with marking of osteomy

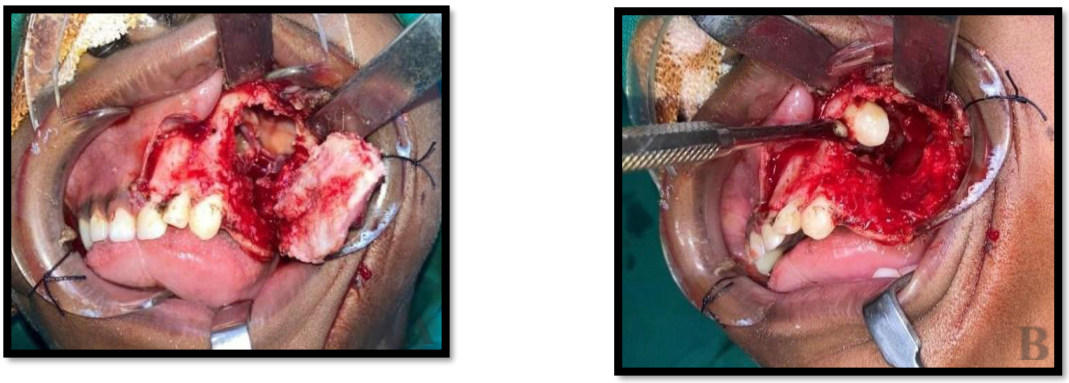

Fig-5: (A)excised tumour mass and cyst with impacted canine seen in maxillary sinus (B) Image showing an impacted maxillary canine in maxillary sinus

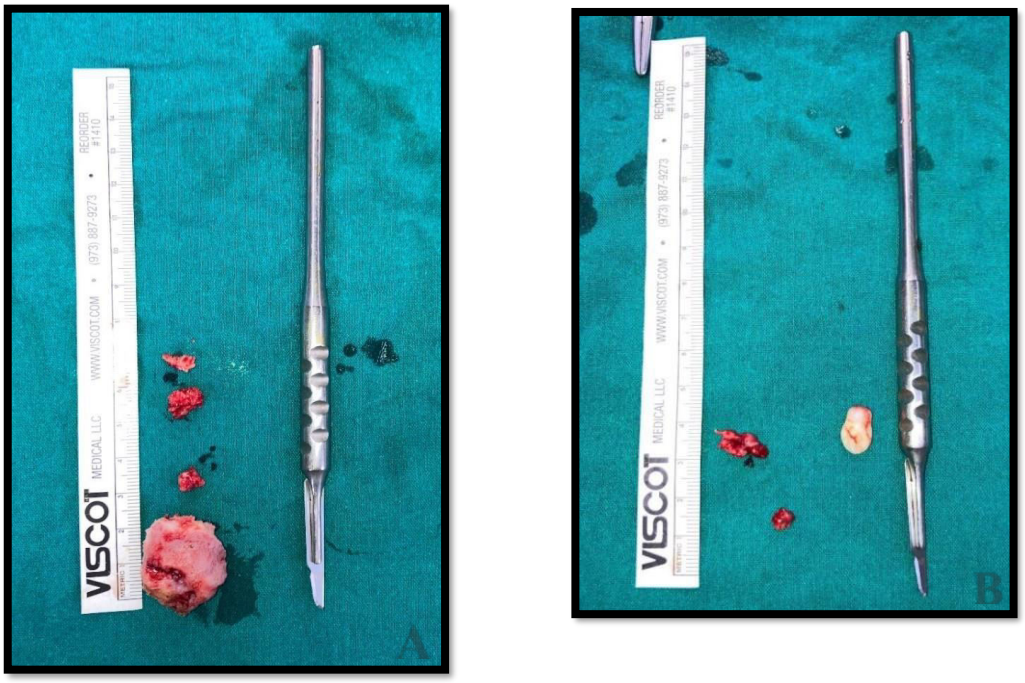

Fig-6: (A) image showing excised tumor mass (B) excised cystic lining with impacted canine tooth

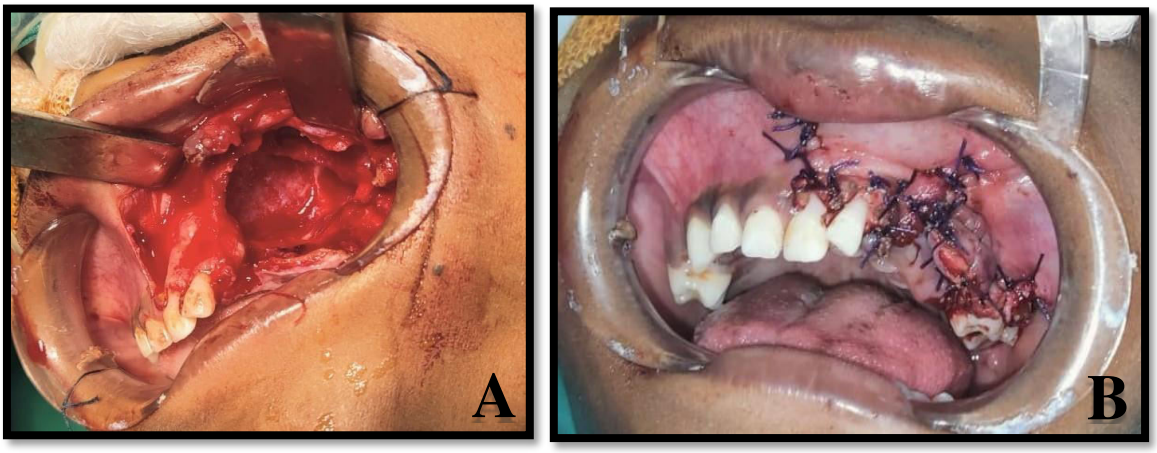

Fig-7: (A)image showing an excised complete tumour mass and dentigerous cyst (B)Closure of surgical site

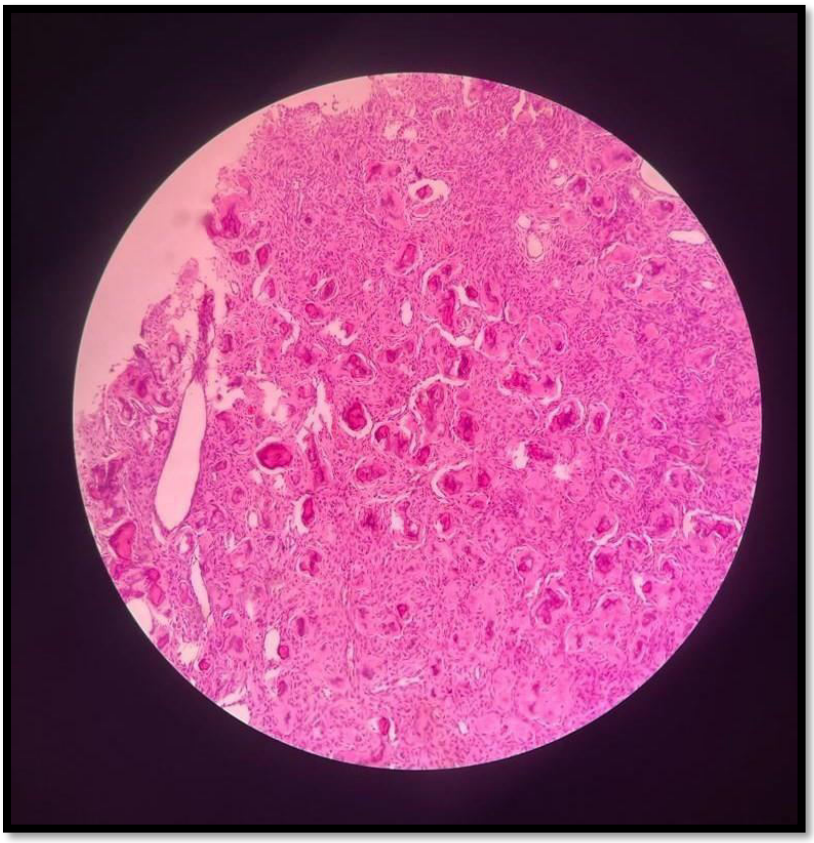

FIG-8: Histopathological examination showed predominantly fibro-osseous proliferation composed of ossicles admixed with fibrous stroma and show presence of calcified psammomatoid bodies.

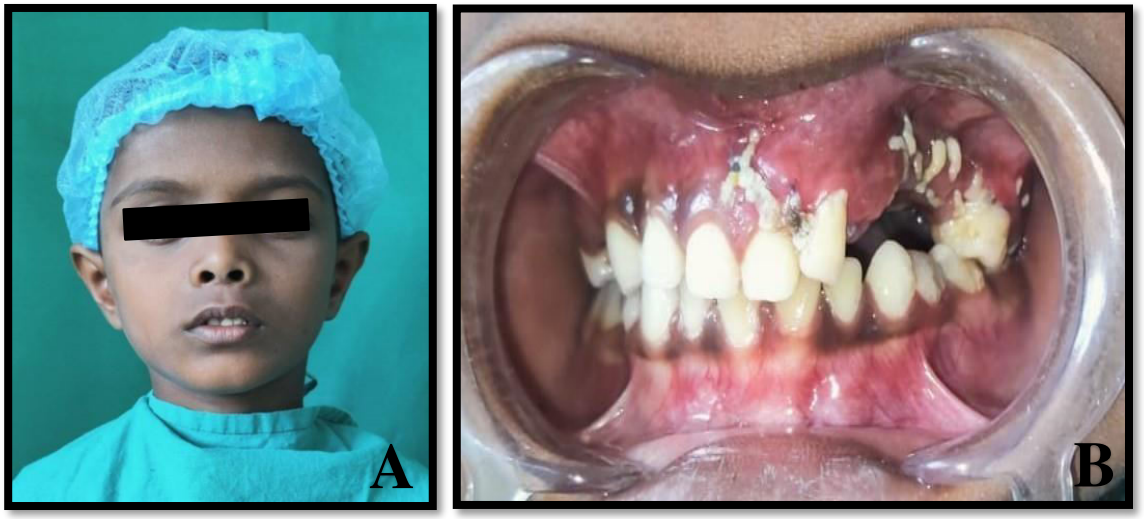

FIG-9:1Month post-operative images (A) Extra-oral image (B) intra-oral image

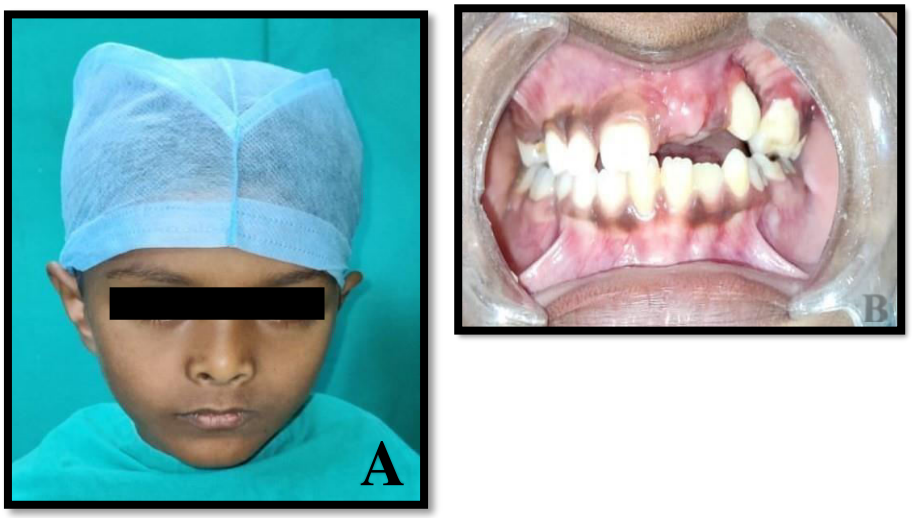

FIG-10:6 Month post-operative images (A) Extra-oral image (B) intra-oral image

Discussion:

Juvenile Ossifying Fibroma (JOF) represents a rare fibroosseous lesion affecting the jaws, distinguished by its onset at an age less than 15 years, distinct radiological features, tumour location, and a notable recurrence rate (5).Historically, confusion existed in the classificationoffibro-osseoustumours,withossifyingfibroma(inadultsorjuveniles)andcemento-ossifyingfibroma considered as separate entities based on histology, the former being classified as odontogenic and the latter as non-odontogenic (6). The term JOF encompasses two distinct histopathologic variants within the craniofacial skeleton: Psammomatoid Juvenile Ossifying Fibroma (PJOF) and Trabecular Juvenile Ossifying Fibroma (TJOF) (7). PJOF predominantly manifests in the sinonasal and orbital bones, while TJOF primarily affects the jaws, with a predilection for the maxilla (8).

The development of these jaw lesions is linked to abnormalities in the basal generative mechanism, which is essential for proper root formation (4). Reports indicate the presence of non-random chromosomal break points at Xq26 and 2q33, resulting in (x,2) translocations (9). Treatment for PJOF typically involves complete surgical resection with a margin of safety. In most cases, abroad access approach is necessary to achieve these objectives (10,11).

However, in well-delimited tumours with a small proportion, smaller access methods such as intra-oral approaches and endonasal endoscopy may be feasible. (12) It is important to note that the use of smaller access carries the disadvantage of a reduced likelihood of achieving complete removal with safety margins in more extensive and invasive lesions (13,14).

The characteristic feature of the psammomatoid subtype of this fibro-osseous lesion is the existence of eosinophilic spherical structures scattered within a fibrous stroma. This stroma is comprised of plump spindle-shaped cells arranged either in strands or whorls. These unique spherical structures are termed psammoma-like bodies, displaying variation in appearance but typically featuring a central basophilic area and a peripheral eosinophilic fringe (15)

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest: There are no conflicts of interest

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