Case report

A Rare Presentation of Central Giant Cell Granuloma of the Maxillary Sinus

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ABSTRACT

Objective: Central giant cell granuloma (CGCG), also known as giant cell reparative granuloma, is a non-cancerous proliferative lesion of unknown aetiology. It is a localized osteolytic lesion with the diverse biological behavior of aggression which most commonly affects the mandible and other jaw bones. It is a rare condition and its characteristic clinical or radiological features are still not well defined. It resembles to some neoplasms and can easily be misdiagnosed with Antro- Choanal Polyp, Angiofibroma, Squamous Cell Carcinoma and Inverted Cell papilloma.

We are reporting a case of rare presentation of CGCG arising from the maxillary sinus. This is a case report of a 15 year old boy who presented with the episodes of recurrent epistaxis and nasal obstruction. Diagnosis of giant cell granuloma was made on the basis of age presentation, rare location, and histological findings of excised specimen, which revealed central giant cell granuloma. This case helps to demonstrate the wide variation in the clinical and radiological features of CGCG and highlights the significance of histological features of this lesion.

Key Words: Central giant cell granuloma, recurrent epistaxis, Nasal obstruction.

Introduction

Central giant cell granuloma (CGCG) is an interosseous lesion comprising of fibrous tissue which is believed to have many foci of haemorrhages, collections of multinucleated giant cells and often trabeculae of woven bone.[1,2] CGCG is a non-cancerous proliferative lesion of unknown cause which most commonly involves mandible and rarely maxilla with infiltrating giant cells[1,3] CGCG is considered local reparative reaction of bone due to its destructive nature.[4] Intramedullary haemorrhage or trauma are the possible contributing factors.[5] Giant cell granuloma though owing a benign course is often confused with giant cell tumour. However for giant cell tumour distinguishing factor is its occurrence in ages of 25 to 45 years.[1,6] Moreover, giant cell tumour usually involves long bones which recurs even after curettage showing the aggressiveness of the tumour and its potential for malignant transformation.[7,8] CGCG has a lower recurrence rate and no cases of malignant transformation or metastasis has been reported.[9,10] Clinical presentation of CGCG of the mandible is variable and difficult to predict. Depending on the clinical and radiological features, it is categorized as nonaggressive and aggressive lesion.[11] Usually, the central lesions present with no signs and symptoms clinically other than a diffuse swelling over the affected area.[11,12] Unilocular or a multilocular radiolucent lesion with diffuse or irregular borders, sometimes leading to expansion of cortex, displacement of teeth, or root resorption can be found radiographically.[11] Still characteristic radiographic and clinical signs are not well defined and it can be misdiagnosed for various malignant and non-malignant conditions. Hence, the diagnosis of CGCG solely depends on histopathology. Histologically, CGCG is characterised by vascular connective tissue and osteoclastic natured multinucleated giant cells and spindle shaped collagenized stromal cells.[11] The cells are evenly dispersed and clustered around haemorrhagic areas.[14] Histologically, the tumour also consists of vascularized network of stromal cells and multinucleated giant cells meagrely interspersed with collagenous fibrils but in contrast to CGCG, it shows irregular and uneven distribution with presence of plump tumour cells in stroma.[13,15] This is verified by the case reported here that is presented with clinical features which lead to differential diagnosis from antro-choanal polyp, angiofibroma, squamous cell carcinoma to inverted cell papilloma. The last two mentioned are unlikely.[15]

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Case report

A male patient 15 year of age was referred from Gojra to Faisal Hospital Peoples Colony Faisalabad with history of recurrent severe epistaxis and nasal obstruction for last one year. On examination there was whitish blog of secretions in the left side of nasal cavity giving the impression of nasal polyps. On posterior Rhinoscopy the mass was seen occupying left choana. Intra oral examination shows pushing of the soft palate anteriorly and there was no swelling over the palate or sub-labial region. No numbness or paresthesia over the cheek was noted and there is no loosening of the teeth of upper jaw. Apart from this, rest of the ENT examination was unremarkable. The patient also gave the history of frequent hospital admissions with the complaints of intractable epistaxis for which he got the treatment frequently in the form of

nasal packing off and on. At that time there were two possibilities. One of them was antrochoanal polyp and the other was angiofibroma. The unlikely possibilities considered were squamous cell carcinoma and inverted cell papilloma. The patient was advised CT-Scan and routine examinations including CBC, Bleeding profile and LFTs. On CT scan there was an extensive lesion in the maxillary sinus which was widespread, breaching the medial wall of maxillary sinus and occupying the nasal cavity and approaching toward the nasopharynx. We planned excision using trans-antral approach. The intra-operative findings favoured the diagnosis of angiofibroma as there was massive bleeding during surgery. Almost 2 pints of blood loss was noted. Inverted cell papilloma and squamous cell carcinoma were less likely diagnosis considering the young age of the patient, clinical presentation of severe epistaxis, origin of the lesion from maxillary sinus and growing towards the nasopharynx, duration of the lesion and intra operative findings.[15] A diagnosis of CGCG was achieved by histopathological report of the excised specimen. Histopathological report reveals spindle ovoid to round histocytes with well vascularized fibrous stroma and woven bone lined by osteoclast, suggestive of giant cell lesion. All this morphology and age of the patient favours central giant cell granuloma.

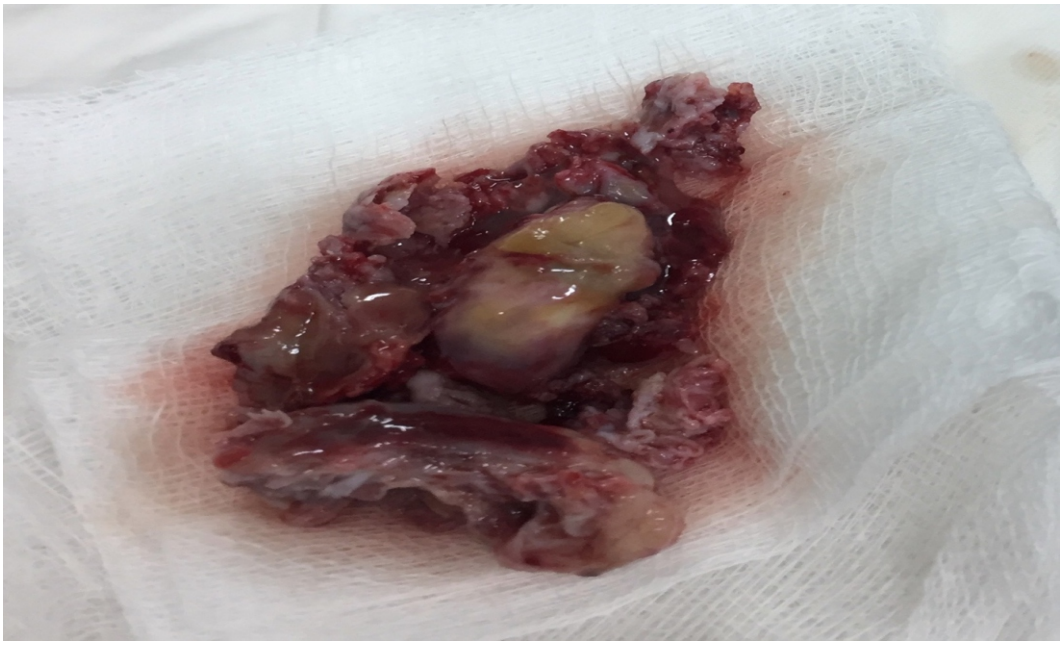

Figure 1: Mass Removed from Nasal Cavity and Maxillary Sinus.

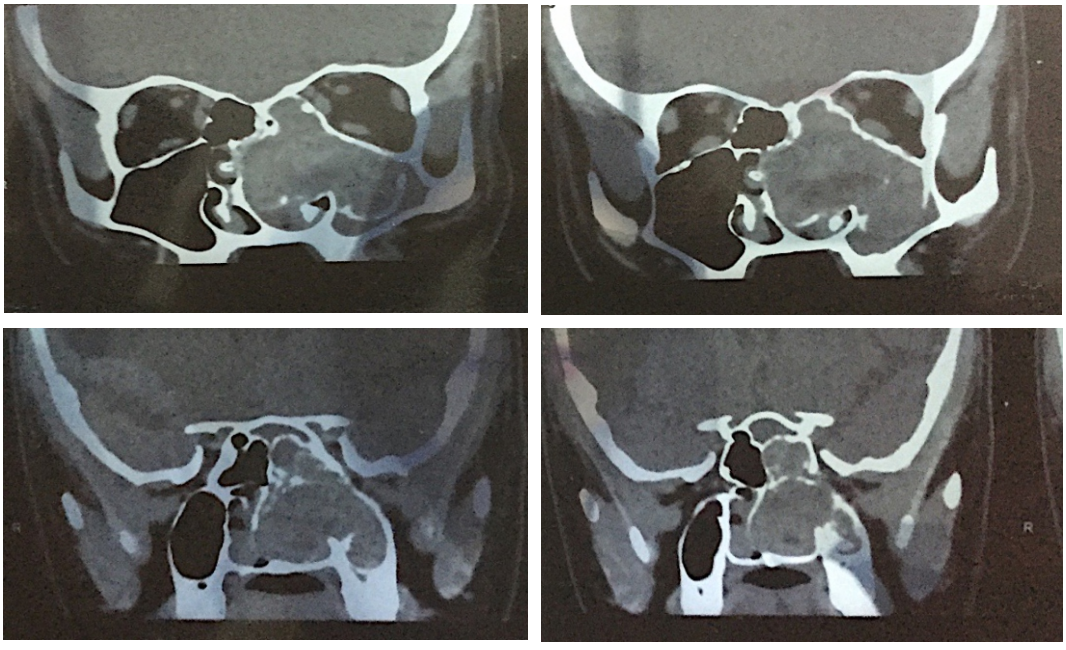

Figure 2: CT scan coronal view

Discussion

Central giant cell granuloma is believed as a non-cancerous proliferative condition whose etiology is still not known, however evidences are available showing it can be secondary to trauma.[1,2] It commonly develops in in children and young adults aged less than 30 years with more predilection in females than males.[16] Central giant cell granuloma occurs in any of the facial bones and cranial vault. Most common presenting site for this granuloma is mandible and rarely in maxilla.[17] Mandibular granuloma usually presents infront of right sided first molars and is seen often to cross the midline. The clinical presentation of CGCG is not uniform and it has an unpredictable course.[16] It is believed to be quite variable ranging from asymptomatic slow growing painless swelling with noticeable facial asymmetry, to aggressive nature of the lesion that manifests with pain.[2,11,17] Alternatively, this finding can be disclosed accidentally while doing jaw radiography for other purposes. Palpation of the suspected bony area may elicit tenderness. Teeth may lose their firmness to the point of attachment but maintain their vitality.[11] The present case, however, involved the maxilla. Maxillary CGCG is likely to present with asymptomatic facial swelling as the cortical bone here is thin and provides little resistance to growth. Similar report of maxillary CGCG was reported by Tsichlaki A in 2012. Tsichlaki A reported the presence of maxillary CGCG in a 45 years old female, who presented with five months history of nasal obstruction.[17] The age of presentation was unusual as it is more common in first three decades of life.[16] We reported the presence of CGCG in 15 years old boy, who presented with episodes of recurrent epistaxis and nasal obstruction for 1year. On the bases of the presenting age, rare location and histological features of the specimen of patient we diagnosed it as CGCG. Kapoor R reported a case of maxillary CGCG in 2016. He reported the case of maxillary CGCG in 20-year-old female patient presenting with swelling of the left side of the face for 1-year. Our report and previous reports discussed above are showing the wide variation in the clinical features of patients of CGCG. A diagnosis of giant cell granuloma should depend upon histological features as the radiological pictures CGCG presentation is also not uniform. It appears from unilocular or to multilocular on radiographs.[11,16] It may be well-defined as well as can be poorly defined and can show variable expansion with damage to cortical plate.[11] Case reported by Chavva S, shows ill-defined borders with evident cortical destruction and migration of associated teeth.[11] The radiological appearance of the lesion cannot be considered as diagnostic for granuloma solely as it can be confused with many other lesions of jaw. Hence, the ultimate diagnosis depends upon histopathology.[9] Histologically giant cell granulomas show numerous multinucleated giant cells and mononuclear cells (fibroblast and histiocyte-like cells and monocyte-macrophages) within a prominent fibrous stroma.[11] Evidences shows multinucleated giant cells exhibiting characteristics of the osteoclasts phenotype.[15] These findings are in favour of our reported case as we found almost similar histological findings. Our histopathology report of excised specimen reveals collections of spindle ovoid to round histocytes with well vascularized fibrous stroma. Woven bones lined by osteoclast were also noted in specimen of the patient. Our report concludes giant cell rich lesion. All morphological changes favour central giant cell granuloma. These characteristic features of CGCG were also reported by previous researches.[1,11,14] In most of the cases this granuloma presents as a single, painless radiolucent expansion. Some lesions are seen to be more devastating even on surrounding bones.[18] The management of CGCG depends upon presentations of the lesion and on radiographic findings. Generally, curettage is done for localized and well-defined lesions with a low rate of recurrence.[18,19] In widespread lesions, which involves cortex perforation on radiographs, radical excision and partial maxillectomy is inevitable. Adjunct to surgery, medical management includes steroids or calcitonin that is believed to inhibit the function of giant cells and halt the osteoclastic activity.[18,19] However, alpha Interferon appears to be fruitful for managing aggressive CGCG due to its anti-angiogenic effects.[17] Alpha Interferon also encourages bone formation through stimulation of osteoblasts and pre-osteoblasts and inhibit bone resorption.[19] Intravenous Bisphosphonates are given on priority basis with hopeful results.[19] Follow-up at regular interval is mandatory to rule out any occurrence. Recurrences are rare and are more common in the maxilla.[18] Presentation of this lesion is quite variable and challenging for diagnosis. So we recommend considering CGCG in the differential diagnosis of the growths of the maxillary sinus. In our case the clinical findings and behaviour is quite different resembling the antrochoanal polyp and angiofibroma.

Conclusion

Diagnosis of CGCG should be made on the basis histopatholical findings, as clinical and radiological features are widely varied among patient to patient.

Author Contribution

All authors contributed equally and are responsible for material provided.

Conflict of Interest and Funding Disclosure

They have no conflict of interest and funding support.

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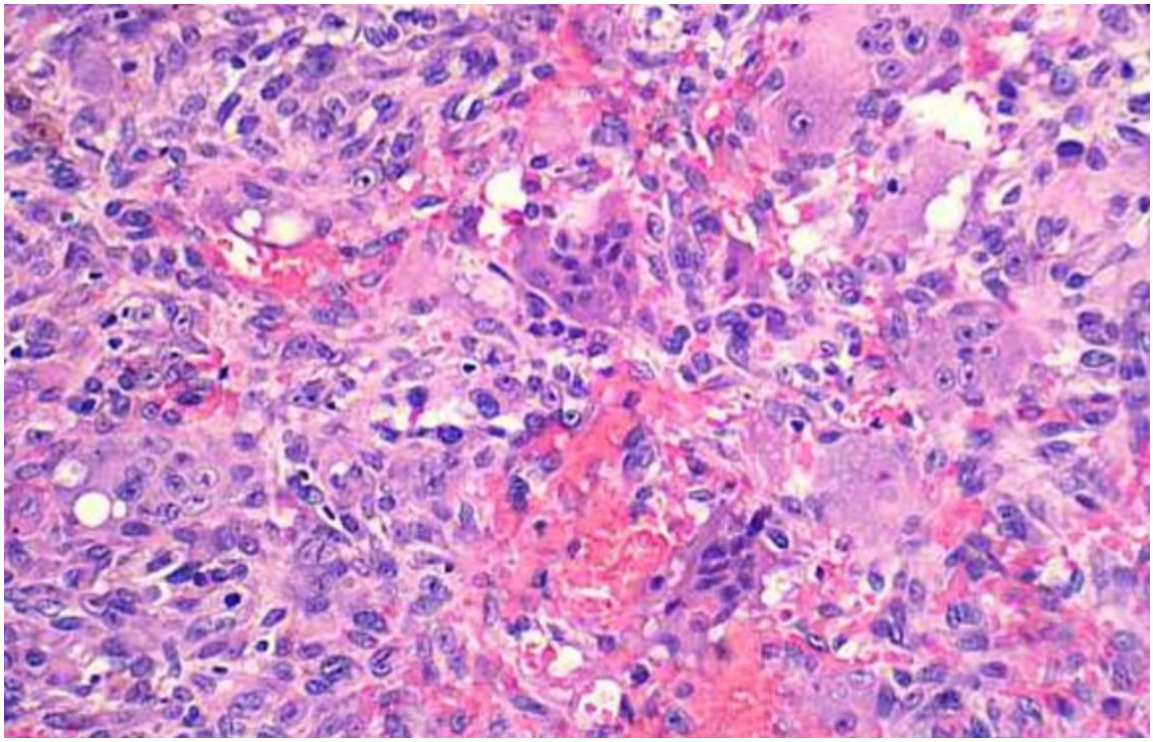

Figure 3a: Histopathology slide showing giant cell granuloma features.

Spc Nature: BIOPSY
Spc Site: NASAL CAVITY

History: Sessile polypoidal mass in the left side of nasal cavity, bleeds on touch.
Angiofibroma and inflamed antrochoanal polyp?.

Gross: Specimen container is labeled with the patient's name and medical record number.
Received in formalin are multiple soft tissue fragments collectively measuring 6.5 cm x 4.0 cm x 3.0 cm.
Serial sectioning of multiple fragments showing tan white cut surface, firm in consistency.
Representative sections are taken and submitted in blocks (A-D).

Micro: Sections show collection of spindle ovoid to round histiocytes with well vascularized fibrous stroma.
There is woven bone lined by osteoclasts.

IMMUNOHISTOCHEMICAL STAIN:
Androgens receptor: Negative

Diagnosis: **NASAL CAVITY, BIOPSY:**
Giant cell rich lesion (see note).

Note: Morphology favors central giant cell granuloma. Correlate with radiological findings.

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Fig. 3b: Histopathology Report of the specimen shown in fig. 2.