MINI REVIEW

Rhinology, 48, 146-149, 2010

A benign maxillary tumour with malignant features*

Rosario R. Ricalde[1], Aimee Caroline E. Lim[1], Ramon Antonio B. Lopa[1], Jose M. Carnate, Jr.[2]

[1] Department of Otorhinolaryngology, University of the Philippines – Philippine General Hospital, Manila, Philippines
[2] Department of Pathology, University of the Philippines – Philippine General Hospital, Manila, Philippines

SUMMARY

Non-specific biopsy results such as chronic inflammation, hemorrhage, necrosis can be frustrating to the clinician. This is especially true if the patient presents with clinical features suggestive of an aggressive tumour. This is a review of the clinical features, diagnostic dilemmas and surgical management of a benign maxillary mass with malignant features - a disease called hematoma-like mass of the maxillary sinus (HLMMS). Our experience with five cases will also be cited.

Key words: organized haematoma, organizing haematoma, haemangioma, haematoma, haematoma-like mass, maxillary sinus, epistaxis

INTRODUCTION

There are times when a patient presents with clinical features suggestive of a malignancy - epistaxis, nasal obstruction, prominent cheek bulge, proptosis, palatal bulge, blurring of vision of a few months' duration. Imaging in the form of CT scan and MRI is the next rational step. The CT scan and MRI results show malignant features such as bony erosion and a heterogeneously enhancing mass. At this point, the novice clinician is confident that the tumor is aggressive and most likely a malignancy. To confirm the initial assessment and determine management, a biopsy is done. The outcome shows chronic inflammation, hemorrhage and necrosis – all benign non-specific results that are not expected given the clinical and radiological features. As a consequence, multiple biopsies are performed before definitive management is actually performed in a futile attempt to reconcile the clinical and radiological data as well as a histopathology report. Such tumours exist and have been called maxillary sinus haematoma, organized haematoma, organizing haematoma, haematoma-like mass of the maxillary sinus. To the authors' best knowledge, this is the first time that this disease is being reported in the Philippines. We review the literature and cite our local experience with five patients diagnosed with this benign tumour with malignant features.

DISCUSSION

A non-neoplastic haemorrhagic lesion exhibiting mucosal swelling and bone destruction can develop in the maxillary sinus. This has been reported by Tadokoro in the Japanese literature as far back as 1917 and was called blood boil of the maxillary sinus [(1)]. Other diseases reported that seem to be synonymous to the blood boil of the maxillary sinus are organized haematoma, organizing haematoma, and haematoma of the maxillary sinus [(2-6)]. Yagisawa et al. proposed the term haematoma-like mass of the maxillary sinus (HLMMS) to unify all above synonyms and to resolve confusion in terminology [(1)], a term the authors likewise prefer.

Until now only a few cases have been reported, with Song et al. of Korea presenting the most cases at 20 [(6)]. In the review of Lim et al. on spontaneous maxillary sinus haematoma, 12 cases were cited [(7)]. In the Philippines, we have diagnosed and successfully managed 5 cases. Table 1 shows the summary of the clinical and radiological features and management of our cases.

Patients had a wide age range varying from the 1st decade to the 7th decade of life, averaging at 50s, and slightly more common in males [(1-8)]. Common initial symptoms were epistaxis, nasal obstruction and cheek swelling (Figure 1) [(1-8)]. All of these symptoms were also present in our five cases.

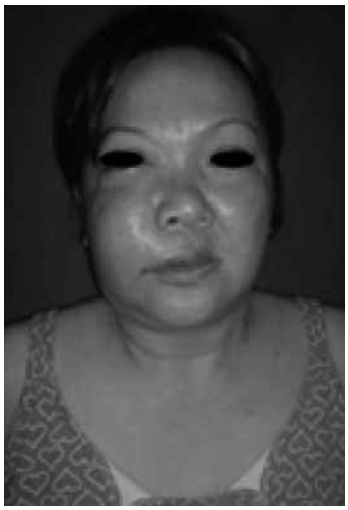

Figure 1. Patient showing a cheek swelling.

A predisposition to spontaneous bleeding is a likely risk factor.

*Received for publication: March 11, 2009; accepted: August 28, 2009

DOI:10.4193/Rhin09.046

Table 1. Summary of clinical and radiological features and management.

CASE	AGE/ SEX	INITIAL SYMPTOM	CT SCAN / MRI FINDINGS	BIOPSY RESULT	INITIAL DIAGNOSIS	SURGERY	FINAL HISTOPATH
1	F/68	Cheek swelling	CT Scan - Heterogenous density in the left maxillary sinus which extended into the ethmoid, sphenopalatine fossa, medial wall of the orbit, skull base and frontal lobe. There was erosion of the medial pterygoid plate, alveolar ridge and medial wall of the right maxillary sinus	Mixed capillary and cavernous haemangioma	Infection	Caldwell-Luc	Fibrocollagenous and necrotic tissue with chronic inflammation
2	M/59	Epistaxis	CT scan - heterogenous non-enhancing mass occupying the right maxillary, ethmoid and sphenoid sinuses, the right nasal cavity and the right orbit with erosions through the nasal septum, medial wall of the right orbit with proptosis of the right eye	Inflammatory nasal polyp	Meningioma vs cancerous new growth	Denker's	Inflammatory nasal polyp with haemorrhage and necrosis
3	F/49	Epistaxis	CT scan - progressive enlargement of the left maxillary sinus with involvement of the left nasal cavity, ethmoid and sphenoid sinuses, left masticator space, beginning osseous erosion of the left orbital wall	Chronic non-specific inflammation, necrotic tissue	Nasal polyposis	Denker's	Chronic inflammation
4	M/48	Epistaxis	CT scan - expansile right maxillary mass, extending to the nasal cavity and ethmoidswith erosion of the medial maxillary wall.	Chronic inflammation	Intranasal mass t/c carcinoma	Denker's	Haemangioma
5	F/48	Proptosis	CT scan - non-homogenous maxillary mass extending to the right ethmoid sinuses. Lateral and superior bone erosion of ethmoid air cells, medial maxillary wall and floor of the orbit.	Acute inflammation with necrotic debris	t/c maxillary sinus carcinoma	Excision via gingivobuccal approach	Haematoma

Hypertension and diabetes were other factors identified in our cases. One had a significant history of facial trauma, while another was a professional deep-sea diver. Only one patient had no identifiable co-morbid condition or risk factor.

The pathophysiology of a haematoma-like mass of the maxillary sinus starts with the accumulation of blood within the maxillary sinus. This may be either from an obvious tendency for bleeding such as a history of trauma, bleeding diasthesis and vascular disease, or spontaneous bleeding due loss of mechanical integrity of the arterial supply to the maxillary sinus, as in ruptured aneurysm or inflammatory erosion of the arterial wall [3,5]. The collection of blood then exceeds mucociliary clearance within the maxillary sinus leading to retention of blood and formation of a fibrous capsule around the haematoma preventing reabsorption, allowing recurrent intracapsular bleeding, progressive expansion and local bony erosion [3].

Two imaging studies are of value in diagnosing this disease –

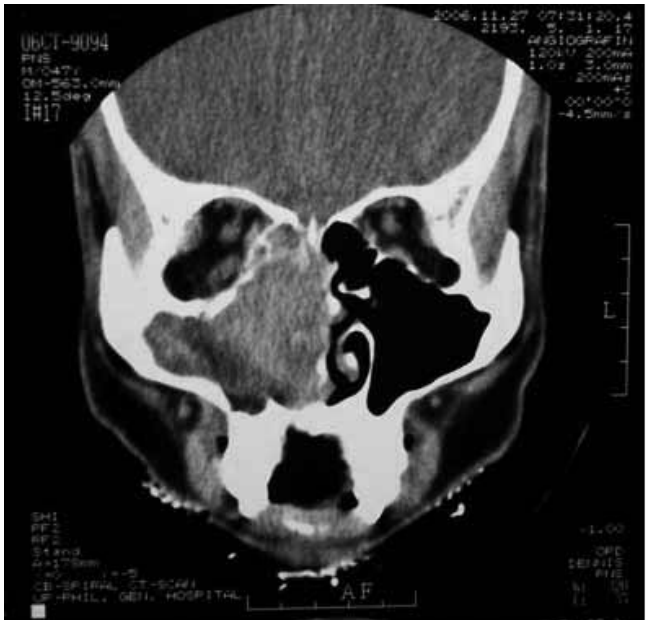

Figure 2. Coronal CT scan showing a heterogeneously enhancing expansile right maxillary mass with bony erosion.

CT and MRI with contrast. On CT, the mass has variable enhancement density with thinning or destruction of surrounding bone [3-5,8,9]. Figure 2 shows the CT scan of one of our cases. On MRI, the mass is well demarcated from surrounding structures and heterogenous in signal intensity on both T1WI and T2WI [5,9,10]. According to Yagisawa et al., CT and MRI provide sufficient information to differentiate these lesions from malignant tumours [1].

All our patients had a paranasal sinus CT taken, revealing a unilateral, heterogeneously enhancing, infiltrating, expansile mass. In retrospect, the bony erosion might have been pressure induced and that radiologically, the lesions might not be neoplastic.

No malignant cells or microbes were seen and cultures for bacterial, fungal and acid fast bacilli were negative. This ruled out infection and neoplasm.

Table 2. Histological criteria according to several authors.

Yagisawa et al., 2006 [1]	Dilated vessels, haemorrhage and fibrin exudation.
Unlu et al. 2001 [2]	Lesions consisted of amorphous eosinophilic, relatively acellular fibrinous mass with some intact erythrocytes. In addition, marginal areas of the lesion showed fibrin deposition beneath the normal respiratory epithelium.
Tabee and Kacker, 2002 [3]	Benign reactive focal spindle cell proliferation in the background of organized haematoma and fibrosis.
Yoon et al., 2006 [4]	Old haematoma with hemosiderin pigment, fibrous tissue with fibrin material and some fibroblast proliferation, and vascular proliferation.
Lee et al., 2003 [5]	Amorphous fibrin masses mixed with haematoma and neovascularization.
Song et al., 2007 [6]	Peripheral wall consists of dense fibrous tissue, neovascularization, and extravasated red blood cells. Fibrosis and angiogenesis were organized in the haematoma background.

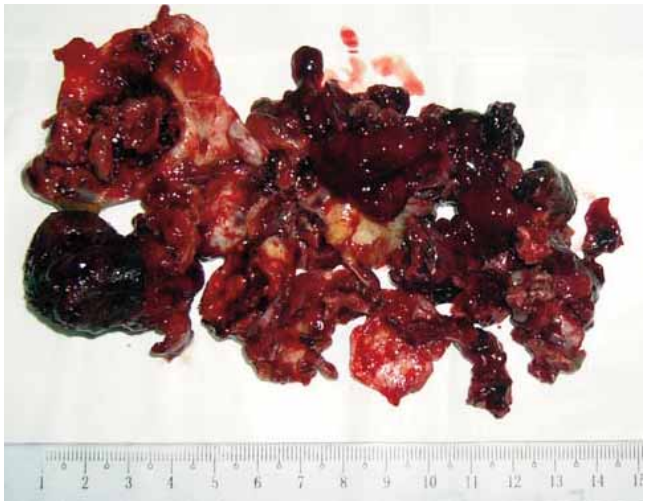

Figure 3. Gross specimen showing friable, fleshy, meaty red mass with blood clots.

The initial biopsies of acute or chronic inflammation, necrosis, inflammatory nasal polyps, haematoma, and haemangioma were not consistent with the clinical and radiological picture. Commonly, the problem with a biopsy is the limited volume of tissue obtained for histopathological examination. Few sections, possibly not representative of the whole lesion, may be the only specimen available for review. Since the mass is expansile, inflammation and polyp formation are expected and the sections biopsied may be of these parts while the true features of the mass are missed as it is located deeper and more central. Due to the diagnostic dilemma, management was delayed as biopsies had to be repeated and readings reviewed.

All patients eventually had definitive surgery either via the Caldwell-Luc or Denker's approach. Grossly, the masses were friable, fleshy, meaty red with blood clots (Figure 3). All specimens had a fibrous friable capsule, which was easily excised, suctioned and hard to preserve. This is consistent with the gross findings in other reports [1-5].

A review of the histopathological material from the five cases, by one of the authors who is a pathologist, revealed that all specimen had dilated vessels and or neovascularization, fibrin exudation or deposition in layers and haemorrhage (Figure 4). These were consistent with the 3 histopathological characteristics presented by Yagisawa et al. (1), which is an admixture of 1) haemorrhage, 2) dilated vessels, and 3) fibrin exudation. Table 2 shows the histopathological criteria adapted by different authors.

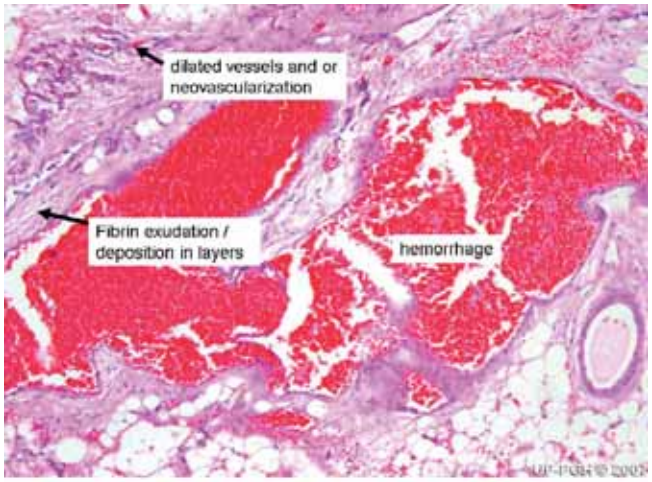

Figure 4. Low-power magnification with H &E stain showing dilated vessels with congestion, neovascularization, fibrin exudation, organization and haemorrhage.

In our experience, it is always prudent to biopsy a clinically and radiologically destructive lesion to rule out a neoplasm. However, when several attempts have been made with the same benign findings, a complete excision may be necessary which will be both diagnostic and curative. A Caldwell-Luc or Denker's approach is a sufficient method.

CONCLUSION

Recurrent epistaxis, nasal obstruction, facial swelling, proptosis and an intranasal mass and imaging studies of an expansile mass with variable enhancement density, thinning or destruction of surrounding bone on CT, well-demarcated with heterogenous signal intensity on both T1WI and T2WI on MRI, should raise suspicion for a benign tumour such as HLMMS. Complete excision via Caldwell-Luc or Denker's approach is both diagnostic and curative. A histopathology of haemorrhage and necrosis, fibrin exudation and neovascularization, and a negative culture for organisms should confirm the diagnosis. This entity should be a consideration in all cases of a clinically and radiologically destructive maxillary sinus tumour. Haematoma-like mass of the maxillary sinus (HLMMS), as a unifying diagnosis should be adopted.

REFERENCES

1. Yagisawa M, Ishitoya J, Tsukuda M. Haematoma-like mass of the maxillary sinus. Acta Otolaryngol. 2006. 126: 277-281.
2. Unlu HH, Mutlu C, Ayhan S, Tarhan S. Organized haematoma of the maxillary sinus mimicking tumor. Auris Nasus Larynx. 2001; 28: 253-255.
3. Tabaee A, Kacker A. Haematoma of the maxillary sinus presenting as a mass – a case report and review of literature. Int J Pediatr Otorhinolaryngol. 2002; 65: 153-157.
4. Yoon TM, Kim JH, Cho YB. Three cases of organized haematoma of thaemaxillary sinus. Eur Arch Otorhinolaryngol. 2006; 263: 823-826.
5. Lee BJ, Park HJ, Heo SC. Organized Haematoma of the Maxillary Sinus. Acta Otolaryngol. 2003; 123: 869-872.
6. Song HM, Jang YJ, Chung YS, Lee BJ. Organizing haematoma of the maxillary sinus. Otolaryngol Head Neck Surg. 2007; 136: 616-620.
7. Lim M, Lew-Gor S, Beale T, Ramsay A, Lund VJ. Maxillary sinus haematoma. J Laryngol Otol. 2008; 122: 210-212.
8. Lee HK, Smoker WR, Lee BJ, Kim SJ, Cho KJ. Organized Haematoma of the maxillary sinus: CT findings. Am J Roentgenol. 2007; 188: 370-373.
9. Nishiguchi T, Nakamura A, Mochizuki K, Tokuhara Y, Yamane H, Inoue Y. Expansile Organized Maxillary Sinus Haematoma: MR and CT Findings and review of Literature. Am J Neuroradiol. 2007; 28: 1375-1377.
10. Kim EY, Kim HJ, Chung SK et al. Sinonasal organized haematoma: CT and MR imaging findings. AJNR Am J Neuroradiol. 2008; 29: 1204-1208.

Rosario R. Ricalde, MD
Department of Otorhinolaryngology
University of the Philippines – Philippine General Hospital
Taft Avenue Ermita
Manila
Philippines 1000

Tel: +63-2-681 9506
Fax: +63-2-681 9505 or +63-2-631 1914
E-mail: chitricalde@yahoo.com