Case Report

CENTRAL CAVERNOUS HEMANGIOMA OF THE MAXILLA- A CASE REPORT

Ramniwas M. Kumawat, Sarika L. Dindgire
Government Dental College & Hospital, Nagpur, Maharashtra, India

Corresponding author: Dr. Ramniwas M. Kumawat BDS, MDS, Department of Oral Pathology and Microbiology
Room no 215, Government Dental College and Hospital, Nagpur 440003, Maharashtra, India
Mob: +919766915091, Email address- drramniwask@gmail.com

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ABSTRACT

Hemangioma of the maxilla is a rare lesion with a variable and atypical radiographic appearance. A Hemangioma of bone is an uncommon tumour, comprising less than 1% of bone lesions in a series of 3,987 patients reviewed by Dahlin. Although these tumours are benign, they should be approached with great caution as demonstrated by numerous reports of morbidity and mortality. There is a tremendous variation in the features of central haemangiomas of the jawbones. Because of this variation, no pathognomonic sign exists that can be used to make a definite clinical diagnosis of these tumours. In addition, confusion with other conditions may occur since haemangiomas may mimic other lesions clinically, radiographically, and in some cases histologically. This report describes the features, diagnosis, and treatment of central hemangioma of the maxilla encroaching the maxillary sinus which is quite rare entity.

Key words: Central hemangioma, Cavernous hemangioma, Maxillary sinus.

الملخص العربي

وَرَمٌ وعائِيٌّ كَهْفِيّ مركزي في الفك العلوي، تقرير حالة

رامامنياوس م. كوماوات، ساريكا ل. ديندجاير
قسم أمراض الدم وعلم الأحياء الدقيقة، كلية الأسنان والمستشفى الحكومي، ماهرتشرا، الهند
للإتصال يراسل الكاتب الأول على العنوان أعلاه

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إن الورم وعائي طهفي في الفك العلوي هي آفة نادرة تتميز بمظاهر متغيرة وغير نمطية بالتصوير الشعاعي. وورم وعائي من كما أن الوعاء الكهفي العظمي هو ورم غير مألوف، يشكل أقل من 1٪ من أورام العظام في سلسلة من 3987 مريض تمت مراجعتها من قبل داهلين. على الرغم من أن هذه الأورام حميدة، يجب أن التعامل معها بحذر شديد حسب العديد من التقارير عن معدلات الاعتلال والوفيات. هناك اختلاف كبير في ملامح الأورام الوعائية المركزية لعظام الفكين. وبسبب هذا الاختلاف، لاتوجد علامات واصمة يمكن الإعتماد عليها في تشخيص سريري مؤكد لهذه الأورام. وبالإضافة إلى ذلك، يمكن الخلط بينها وبين غيرها من الأورام حيث أن الأورام الوعائية الكهفية قد تحاكي غيرها من الورام سريرياً، وأشعياً، وفي بعض الحالات تشريحياً. هذا التقرير يقدم وصفاً لخصائص، وتشخيص، وعلاج ورم وعائي كهفي مركزي للفك العلوي زاحف على الجيب الفكي، وهي حالة نادرة جداً.

الكلمات المفتاحية: ورم وعائي مركزي، ورم وعائي كهفي ، الجيب الفكي.

INTRODUCTION-

A haemangioma is a proliferation of blood vessels that creates a mass resembling a neoplasm. The central (intraosseous) type is frequently found in the vertebrae and skull and rarely develops in the jaws. When occurring i the jaw, the lesion produces a hard non tender slow-growing swelling [1]. The teeth, which is in the vicinity of the tumour may be loosened and bleeding may occur from the gingiva around the necks of the affected teeth [1]. The teeth have increased mobility and may exhibit a pumping action such that, when depressed in an apical direction, the teeth rapidly resume their original position. The lesion may pulsate, and a bruit may be detected on auscultation. Some haemangiomas may be present without any sign or symptom [2]. These lesions of the bone have been referred to as "the great mimicker" because they can produce many different radiographic images. Most commonly, lesions show a multilocular radiolucency with small (honeycomb appearance) or large (soap-bubble appearance) loculations. Phleboliths appearing as small rounded or sausage shaped radiopacities having concentric structure with small radiolucent dot in the centre may be seen. Lesions may also present as a unilocular radiolucency [3]. Management of haemangiomas and the treatment of choice depend on several factors including the age of the patient and the size and extent of the lesions, as well as their clinical characteristics. Some congenital lesions may undergo spontaneous regression at an early age [4].

If superficial lesions are not an esthetic problem and are not subject to masticatory trauma, they may be left untreated [5]. Small and superficial lesions may be completely excised with relative ease. However, excision of more deeply seated lesions usually involves a wider surgical approach, which may result in a disfigurement that is difficult to accept for the treatment of these lesions. In addition, emergency surgery may become mandatory when arterial bleeding arises from intraosseous hemangiomas of the jaw following simple tooth extraction [6]. Various treatments have been used in the management of haemangiomas, including oral corticosteroids, intralesional injection of fibrosing agents, interferon α-2b, radiation, electrocoagulation, cryosurgery, laser therapy, embolization and surgical excision [7], [9]. The report describes the features, diagnosis, and treatment of unusual case of central haemangioma of the maxilla with extension into the maxillary antrum.

CASE REPORT

A young female 16 year-old patient presented with a chief complaint of a swelling in the upper left anterior region of the jaw since 4-5 months. The swelling was asymptomatic from the beginning. On extraoral examination, the face was asymmetrical due to a single intrabony swelling measured approximately 3 cm x 2 cm, situated on the left side of maxilla, which was bony hard in consistency, non-tender on palpation and overlying skin appeared to be normal, Fig. 1.A.

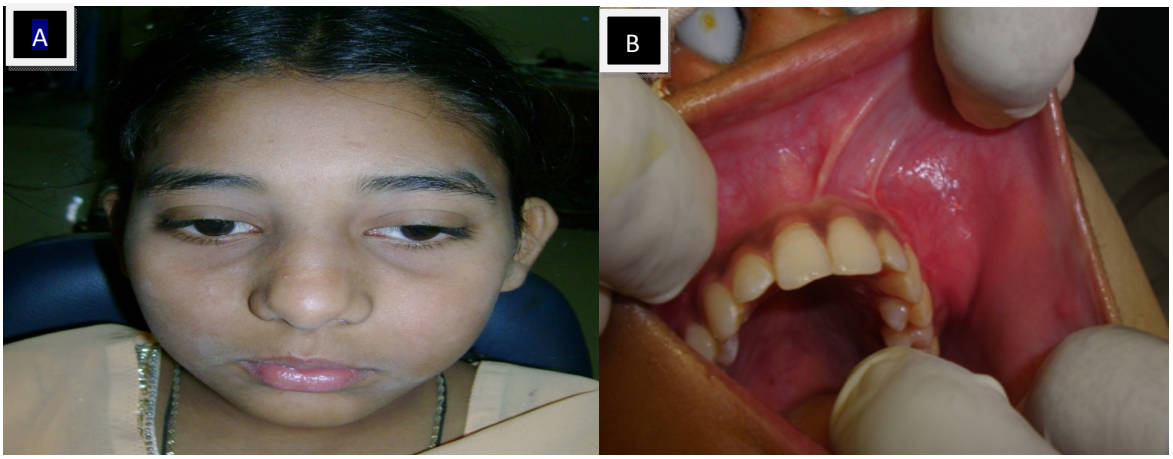

Fig.1.A.Extraoral view showing swelling on left side of face; B. Intraoral view shows Obliteration of left buccal vestibule

On intraoral examination a single diffuse intrabony swelling of size approx. 3X2 cms extending and obliterating left buccal vestibule. Expansion of buccal cortical plate in region of 23, 24, 25, 26 without involvement of palate. Slight displacement and loss of contact point between 25, 26 teeth was seen. Grade I mobility with 23, 24, 25, 26, Fig. 1. B. Radiographic examination shows mixed lesion causing displacement of 25, loss of distinct lamina dura of 25 and mesial aspect of 26, borders are ill defined with blending into surrounding normal bone, loss of distinct trabecular pattern, Fig. 2. A. CT scan reveals expanding bony lesion involving inferior and lateral walls of left maxillary antrum and alveolar process. The lesion was encroaching on sinus, ground glass appearance was noted, Fig. 2. B.

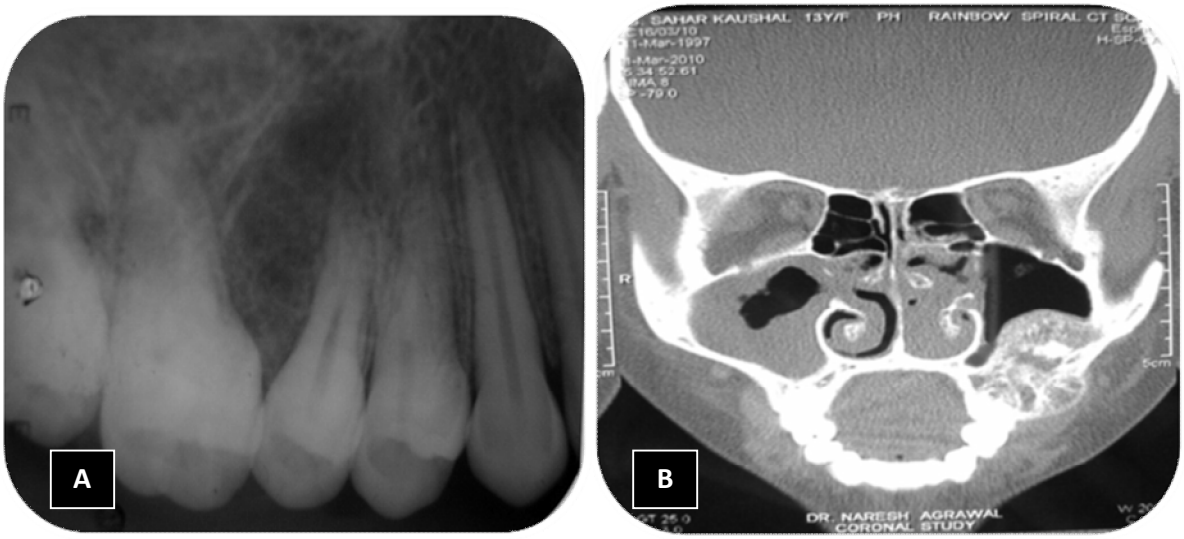

Fig.2.A. IOPA showing mixed radiolucent- radiopoque lesion involving 24, 25, 26.Displacement of 25 and 26 seen. B. CT scan showing expanding bony lesion involving inferior and lateral walls of left maxillary antrum and alveolar process, encroaching the maxillary sinus.

Based on all these features provisional diagnosis of fibrous dysplasia was given. Incisional biopsy was carried out. On H and E stained section shows lesional tissue composed of mature laminated bony trabeculae in fibro- vascular connective tissue stroma. Bony trabeculae are large and interconnected at places, trabeculae shows presence of osteocytes within the lacunae and osteoblastic rimming, Fig. 3. A. The diagnosis of fibro- osseous lesion was given. Based on provisional diagnosis, lesion was surgically curettaged along with bone contouring and surgeon gave the history of severe profuse bleeding intraoperatively. Histopathologically H and E stained section shows numerous small and large, dilated and engorged thick and thin walled endothelial lined blood vessels with extravasated RBCs. At places small proliferating blood vessels were seen with extravasated RBCs. [Fig. 3 B.] Final diagnosis of central cavernous hemangioma was established. The patient's post- operative course was uneventful for 9 months.

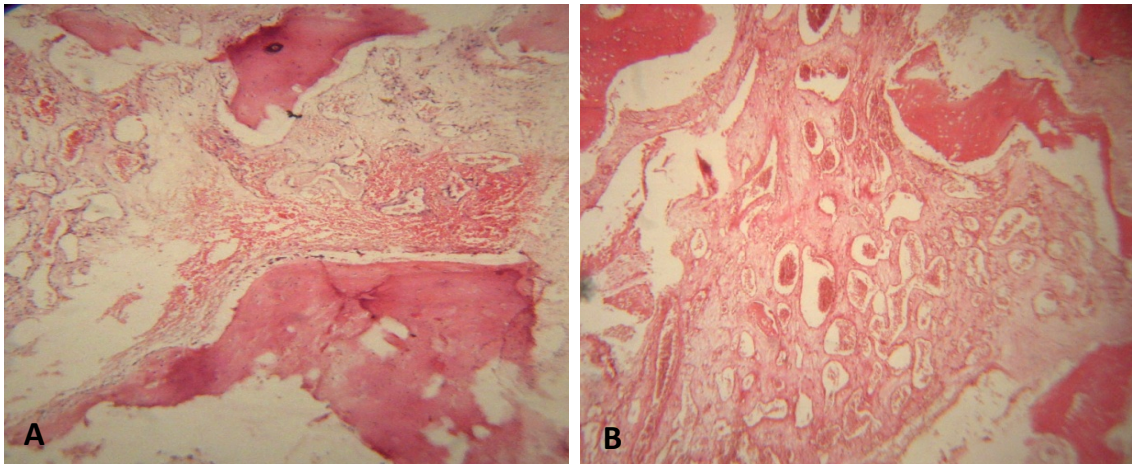

Fig. 3 A. Incisional biopsy showing mature laminated bony trabeculae and fibrovascular connective tissue stroma B. Excisional biopsy showing dilated and engorged endothelial lined blood vessels

DISCUSSION-

Haemangiomas of the jaws are quite rare. When they occur two thirds are found in the mandible, with a female: male ratio of 2:1 and peak incidence in the second decade of life [10, 11]. In the mandible, the greatest frequency of occurrence has been the body region but condylar tumors have also been reported [1]. But this case was in left side of maxilla along with involvement of left maxillary antrum. The origin of central haemangioma is debatable. Signs and symptoms include discomfort, oozing or pulsatile bleeding from the gingiva around the teeth in the region of the lesion, bluish discoloration of gingiva, mobile teeth, derangement of arch form, accelerated exfoliation and agenesis of teeth. Highly expansile lesions cause sensation of pulsation, audible bruits on extension into the soft tissue and blanching on pressure. Occasionally patients have paraesthesia in the region of the lesion [11,12]. In our case, clinical examination revealed asymptomatic swelling present on the left side of the maxilla. The swelling was bony hard, painless to palpation and covered by normal mucosa. Central haemangioma with areas of increased radiopacity can be mistaken for ground glass appearance of fibrous dysplasia. The other radiolucent patterns of central haemangioma can be misdiagnosed as intermediate stages of fibrous dysplasia but this lesion does not show unburst appearance. Histopathologically, the surgical specimen showed cavernous haemangioma involving the medullary space of the maxillary bone.

DIFFERENTIAL DIAGNOSIS

Radiographic differential diagnosis includes osteosarcoma, fibrous dysplasia, central giant cell granuloma, ameloblastoma, odontogenicmyxoma, multiple myeloma and aneurysmal bone cyst. Osteosarcoma produces similar sunburst appearance. It may be entirely radiolucent, mixed radiolucent–radiopaque or quite radiopaque. There may be asymmetric broadening of periodontal ligament space and onion skin growth of periosteal bone. Irregular margins of a lesion raise a question of malignant tumour like osteosarcoma [11]. Central haemangioma with areas of increased radiopacity can be mistaken for ground glass appearance of fibrous dysplasia. The other radiolucent patterns of central haemangioma can be misdiagnosed as intermediate stages of fibrous dysplasia but this lesion does not show sunburst appearance. Multilocular regions of rarefaction that accompany expansion and thinning of the cortex may mimic a central giant cell granuloma of the jaws. However some investigators believe that loculations produced by a central haemangioma are smaller with fine fibrillar network. Cystic lesions are more difficult to diagnose radiographically and an aneurysmal bone cyst may be suspected, but radiographically this lesion does not resorb the adjacent teeth [13].

CONCLUSIONS

Haemangioma of the maxilla is a rare lesion with a variable and atypical radiographic appearance. The differential diagnosis of an expansile paranasal sinus mass with destructive features should include haemangioma, and angiography is

indicated in the work-up of such cases. Embolization of the vascular supply of the tumour prior to surgery is advised. Computed tomography is a valuable aid in staging the extent of the tumour and planning reconstructive surgery.

REFERENCES

1. Marwah N, Agnihotri A, Dutta S. Central hemangioma: an overview and case report. Pediatr Dent. 2006;28:460-6.
2. Whear NM. Condylar haemangioma--a case report and review of the literature. Br J Oral Maxillofac Surg. 1991;29:44-7.
3. Zlotogorski A, Buchner A, Kaffe I, Schwartz-Arad D. Radiological features of central haemangioma of the jaws. Dentomaxillofac Radiol. 2005;34:292-6.
4. Tröbs RB, Mader E, Friedrich T, Bennek J: Oral tumors and tumor-like lesions in infants and children. Pediatr Surg Int 2003, 19:639-645.
5. Silverman RA: Hemangiomas and vascular malformations. Pediatr Clin North Am 1991, 38:811-834.
6. Kocer U, Ozdemir R, Tiftikcioglu YO, Karaaslan O: Soft tissue hemangioma formation within a previously excised intraosseous hemangioma site. J Craniofac Surg 2004, 15:82-83.
7. Onesti GM, Mazzocchi M, Mezzana P, Scuderi N: Different types of embolization before surgical excision of haemangiomas of the face. Acta Chir Plast 2003, 45:55-60.
8. Burstein FD, Simms C, Cohen SR, Williams JK, Paschal M: Intralesional laser therapy of extensive hemangiomas in 100 consecutive pediatric patients. Ann Plast Surg 2000, 44:188-194.
9. Deans RM, Harris GJ, Kivlin JD: Surgical dissection of capillary hemangiomas. An alternative to intralesional corticosteroids. Arch Ophthalmol 1992, 110:1743-1747.
10. Alves S, Junqueira JL, de Oliveira EM, Pieri SS, de Magalhães MH, Dos Santos Pinto D Jr, et al. Condylar hemangioma: report of a case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006;102: 23-7.
11. Nagpal A, Suhas S, Ahsan A, Pai K, Rao N. Central haemangioma: variance in radiographic appearance. Dentomaxillofac Radiol. 2005;34:120-5.
12. Bunel K, Sindet-Pedersen S. Central hemangioma of the mandible. Oral Surg Oral Med Oral Pathol. 1993;75:565-70.
13. Beziat JL, Marcelino JP, Bascoulergue Y, Vitrey D. Central vascular malformation of the mandible: a case report. J Oral Maxillofac Surg. 1997;55:415-9.