

CASE REPORT

Cavernous haemangioma of the left nasal cavity and maxillary sinus

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ABSTRACT

BACKGROUND. Cavernous haemangiomas of the nasal cavity and paranasal sinuses are very rare tumors with only few reported cases in the literature. Clinically, cavernous haemangiomas may be locally aggressive and destructive by virtue of the pressure exerted. The most common locations of these benign lesions include the face, the scalp, the orbit, the oral and nasal cavities.

MATERIAL AND METHODS. Two similar cases of sinonasal cavernous haemangiomas are presented: a 67-year-old woman with a huge cavernous haemangioma arising from the left maxillary sinus and a 59-year-old woman with a cavernous haemangioma originating from the mucosa of the left middle nasal meatus. The diagnostic algorithm and the surgical management – combined approach (transnasal endoscopic and Caldwell-Luc surgical technique) for the first case and minimally invasive transnasal endoscopic technique for the second case, are discussed.

CONCLUSION. Sinonasal cavernous haemangioma is a rare tumor that can lead to diagnostic confusions in what symptomatology and endoscopic appearance are concerned. The CT-scan is a useful diagnostic tool in evaluating the local tumor extension and the surgical procedure type to be chosen for the therapeutic management. Endoscopic sinus surgery alone or combined with external approaches are successful surgical techniques, which can be used in the management of these tumors.

KEYWORDS: cavernous haemangioma, nasal cavity, maxillary sinus, nasal endoscopic surgery

INTRODUCTION

Haemangiomas of the nose and paranasal sinuses are vascular benign tumors, classified by Batsakis as capillary, cavernous, mixed and proliferative¹. Clinically, cavernous haemangiomas may be locally aggressive and destructive by virtue of the pressure exerted. Despite their malignant-type evolution, these tumors do not undergo malignant transformation.

Almost half of the haemangiomas are found in the neck and head regions, their most common locations including the face, the scalp, the orbit, the oral and nasal cavities. First described as entities by Engels, cavernous haemangiomas of the nasal cavity and paranasal sinuses are very rare, with only few cases reported in the literature². They have been described as arising from the inferior turbinate^{3,4}, the vomer⁵, the perpendicular plate of the ethmoidal bone⁶ or the maxillary sinus⁷.

Nasal haemangiomas have to be differentiated from

any intranasal benign or malignant tumor, such as nasal polyps, inverted papilloma, olfactory neuroblastoma, lymphoma, haemangiopericytoma, haemangioendothelioma, arteriovenous fistula, lymphangioma, melanoma, adenocarcinoma, squamous cell carcinoma or metastatic malignancies, etc.

The author reports two cases of sinonasal cavernous haemangiomas operated in 2012 and 2013 in the ENT Department, Braila Emergency County Hospital. In these particular two cases, nasal polyposis coexisted with cavernous haemangioma.

CASE REPORT

Case 1

A 67-year-old woman was admitted in our ENT Department in February 2012 for recurrent epistaxis, left nasal obstruction, postnasal drip, facial swelling and

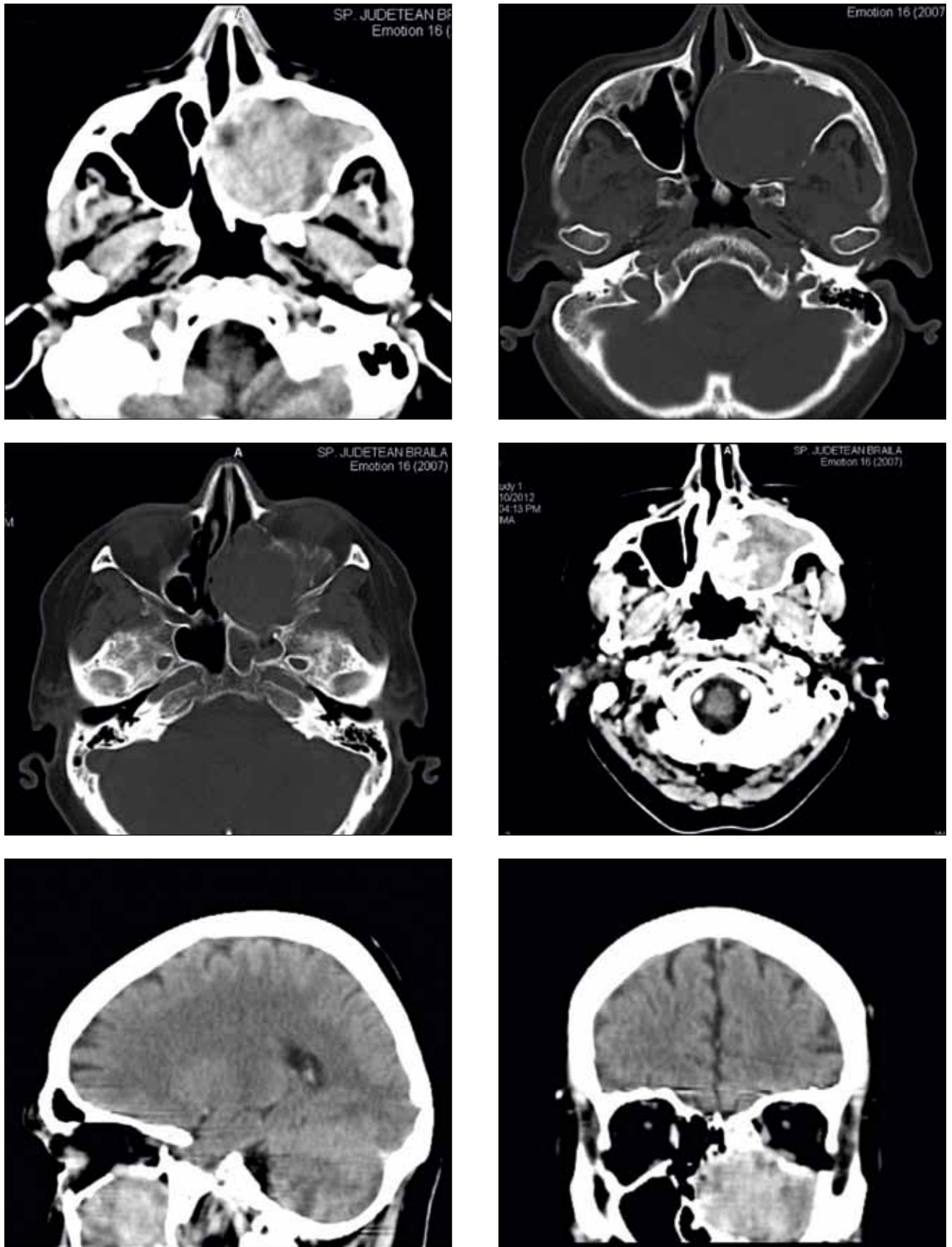


Figure 1 Crano-facial CT scan (axial, coronal and sagittal slices) - circumscribed tumoral mass originating in the left maxillary sinus, involving the ethmoid cells, remodeling the medial wall of the left maxillary sinus, lamina papyracea, cribriform plate and nasal septum



Figure 2 Intraoperative view - combined surgical approach: transnasal endoscopic surgery (a) and Caldwell-Luc approach (b); macroscopic aspect of the tumor (c)

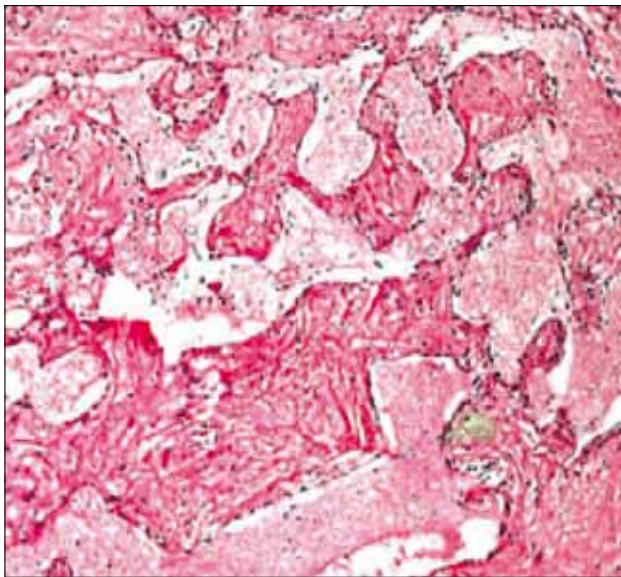


Figure 3 Histopathologic examination – cavernous haemangioma (large blood-filled spaces lined with flattened endothelium)



Figure 4 Cranio-facial CT scan, axial slice, performed 12 months after the surgery - tumor free

headache. The symptomatology started 15 months previously with progressive left nasal obstruction, accompanied by several episodes of anterior nasal bleeding since December 2011. Anterior rhinoscopy revealed a reddish tumor mass filling the left nasal fossa. The patient performed a craniofacial CT-scan examination that showed a $5 \times 4 \times 4$ cm well-circumscribed tumor, originating in the left maxillary sinus, with partial obliteration of the ethmoidal, frontal and sphenoidal air cells on the same side (Figure 1).

Complete tumor resection was achieved by combined approach - transnasal endoscopic surgery and Caldwell-Luc procedure (Figure 2) under general anesthesia. The tumor site of origin at the level of the left maxillary sinus was confirmed intraoperatively. The bleeding was controlled with a coagulation-suction device and the left nasal fossa was packed with Merocel, which was removed 2 days later without bleeding recurrence.

Histopathologic examination of the tumor showed large blood-filled spaces lined with flattened endothelium, and the positive diagnosis was cavernous haemangioma (Figure 3). The patient was discharged 4 days after surgery. Clinical examination and nasal endoscopic examination did not reveal any residual disease at 4 weeks postoperatively. No sign of recurrence was observed after a 12-months follow-up (Figure 4).

Case 2

A 59-year-old woman was admitted in January 2013 in our ENT Department, with history of progressive nasal obstruction, starting from October 2012, but without any previous episodes of nasal bleeding. Also, the patient related headache, nasal discharge, postnasal drip and facial swelling. Anterior rhinoscopy combined with nasal endoscopic examination, performed after nasal decongestion, revealed a red-violaceous

tumor mass occupying the left osteomeatal complex, accompanied by a nasal polypoid mass (Figure 5).

The cranio-facial CT-scan showed a circumscribed tumor mass originating in the left middle meatus, with invasion of the left maxillary sinus and partial opacification of the anterior ethmoid cells (Figure 6), without any signs of bone erosions at the level of the orbit or the skull base.

Complete excision of the tumor was achieved by transnasal endoscopic sinus surgery (Figure 7) under general anesthesia. Intraoperatively, we discovered the site of origin of this tumor at the level of the mucous membrane of the left middle nasal meatus. The anatomopathological examination showed the same characteristics as those found in the first case, the histopathological diagnosis being also cavernous haemangioma.

No signs of residual disease were found at the endo-



Figure 5 Nasal endoscopic examination performed after nasal decongestion - nasal poliposys and red-violaceous tumor mass extruding from middle meatus

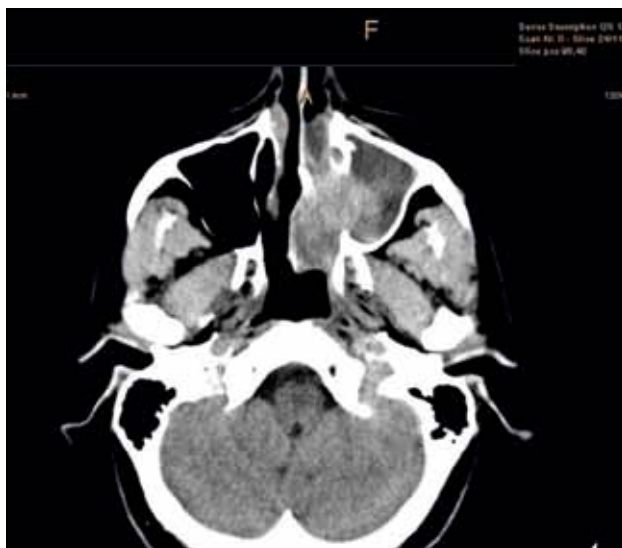
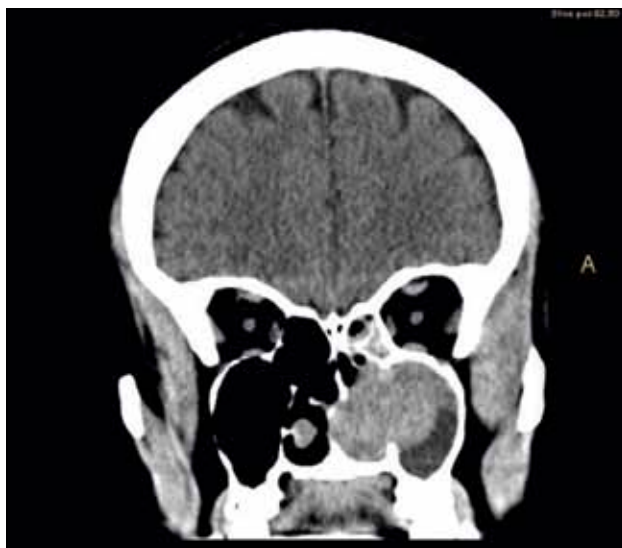


Figure 6 Cranio-facial CT-scan (coronal, sagittal and axial slices) - circumscribed tumor mass originating in the left middle meatus, with invasion of the left maxillary sinus and partial opacification of the anterior ethmoid cells.

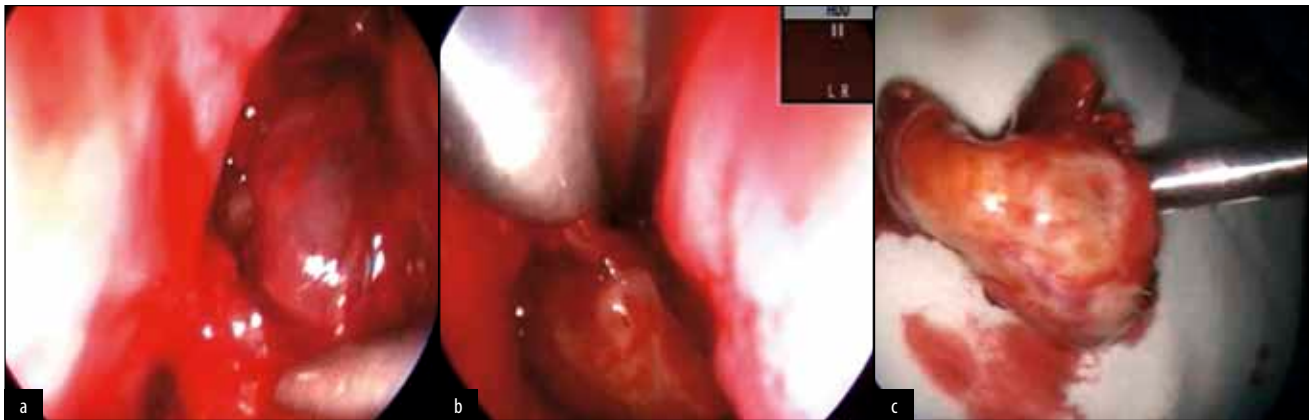


Figure 7 Endonasal endoscopic tumor removal (a, b) and the macroscopic aspect of the cavernous haemangioma (c) – intraoperative view.

scopic examination performed 6 weeks postoperatively. Until now, the follow-up of this patient is too short to make any remarks regarding possible recurrence of the disease.

DISCUSSIONS

Cavernous haemangioma of the paranasal sinuses is a benign vascular tumor, associated with recurrent epistaxis, nasal obstruction, rhinorrhea, facial pain or central facial deformity. It has a slow-growth course, with tendency for bone erosion due to its compressive effect ranging from simple erosion to complete destruction^{8,14}.

Nasal mucosal haemangiomas should be differentiated from haemangiomas that arise from the nasal bones or the maxilla, which are primary osseous lesions; the symptoms and surgical approach of the later are completely different. Microscopically, cavernous haemangiomas appear as large endothelium-lined vascular spaces. In some cases, the thrombi that form into these vascular spaces can calcify and are identified at CT-scan as phleboliths^{15,16}.

Different classification systems for haemangiomas were presented in the literature, histological subtyping classification being the most widely accepted. Thus, depending on the dominant vessel size at microscopy, haemangiomas are divided into capillary, cavernous and mixed types¹.

Sex incidences are prevalent in women and the mean age at presentation of cavernous haemangiomas of the nasal cavity is around 40 years⁹.

Considering the fact that the presence of a bleeding mass in the nasal cavity is a symptom often associated with various malignant or benign lesions, the positive diagnosis is made by the histopathological examination of the surgical specimen.

One of the largest studies presented in the literature by Osborn in 1959 reviewed 51 patients with haemangiomas of the nose, seen over an 11-year period,

and he found that only two were of the cavernous variety. The tumor had unilateral localization and was described as a slowly growing haemorrhagic mass, frequently red, violaceous or purple, sometimes coated with necrotic tissues. Pain was not a characteristic symptom for nasal haemangiomas⁷.

Because of their high vascularization, in order to obtain a histopathological diagnosis, the preoperative biopsy of haemangiomas, is not an easy task and must be performed with great care to avoid severe bleeding. It is recommended to perform an imaging investigation (CT or MRI scans) prior to any biopsy.

Contrast CT-scan examination usually reveals the anatomic location of the tumor and its extension, the characteristic image being a soft tissue density circumscribed mass, enhancing after injection of i.v. contrast substances. Usually, the underlying bone is normal, but it may be remodeled by adjacent long-standing pressure of the expanding mass^{8,9}. A characteristic feature of cavernous haemangiomas is the occurrence of phleboliths. The MRI exam may reveal an isointense lesion on T1-weighted sequences and a hyperintense lesion on T2-weighted sequences, compatible with low-flow vascular structures¹⁴. Characteristically, cavernous haemangiomas do not contain large vessels and, therefore, the MRI scan can differentiate them from the signal voids associated with the hypervascularity typical of other vascular malformations^{9,10}.

Management of the maxillary hemangioma usually includes preoperative embolization in order to avoid hemorrhage, followed by open or endoscopic resection of the tumor¹⁷. In their study, Kim et al¹¹ presented two cases where surgery did not result in severe bleeding. This can be explained by the fact that haemangiomas are venous malformations supplied by small to medium sized vessels and may also reflect a low or non-arterial circulation. Partial resection of this tumor is also advisable in order to preserve adjacent vital structures¹⁷. Haemangiomas are considered to be resistant to radiotherapy; however, some authors con-

sider that, performed before surgery, it may reduce tumor's vascularization¹⁸.

In my two reported cases, preoperative angiography was not performed, because it was not available in our Institution and the patient refused to be referred to another medical center, due to social and financial reasons.

The surgical technique used depends on the location of the tumor and its extension. There are some authors according to whom ligation or cautery of the feeding vessels, or cryotherapy, corticosteroid treatment, sclerosing solutions and YAG LASER resection may be useful⁸. Many surgical techniques have been suggested, such as the lateral rhinotomy, midfacial degloving, Caldwell-Luc, trans-palatal or trans-antral approach and the Le Fort I osteotomy procedure, all of them being indicated in those cases with large invasive tumors. The transnasal endoscopic approach has been proposed as the technique of choice in haemangiomas of the nasal cavity and paranasal sinuses^{8,13}.

In the two cases presented, the minimally invasive transnasal endoscopic approach, used alone or combined with Caldwell-Luc procedure, proved to be efficient in order to provide an adequate exposure and visualization of the lesion, bleeding control and complete removal of the tumor.

CONCLUSIONS

Sinonasal cavernous haemangioma is a rare tumor that can lead to diagnostic confusions in what symptomatology and endoscopic appearance are concerned. In this regard, the second reported clinical case, due to the absence of epistaxis and the association of nasal polyposis, could create confusion regarding preoperative diagnosis and intraoperative risks evaluation.

The CT-scan is a useful diagnostic tool in evaluating the local tumor extension and the surgical procedure type to be chosen for the therapeutic management.

Endoscopic sinus surgery alone or combined with external approaches are successful surgical techniques that can be used in the management of these tumors.

REFERENCES

1. Batsakis J.G. - Tumors of the head and neck: clinical and pathological considerations. Baltimore, MD: Williams and Wilkins, 1979;p.291-312.
2. Engels T., Schorner W., Felix R., Witt H., Jahnke V. - Kavernoses Hamangiom des Sinus maxillaries. HNO, 1990;38(9):342-344.
3. Webb C.G., Porter G., Sissons G.R.J. - Cavernous hemangioma of the nasal bones: an alternative management option. J Laryngol Otol., 2000;114:287-9.
4. Fahmy F.F., Back G., Smith C.E., Hosni A. - Osseous hemangioma of inferior turbinate. J Laryngol Otol., 2001;115:417-8.
5. Nakahira M., Kishimoto S., Miura T., Saito H. - Intraosseous hemangioma of the vomer: a case report. Am J Rhinol., 1997;11:473-7.
6. Graumüller S., Terpe H., Hingst V., Dommerich S., Pau H.W. - Intraosäres Hämangiom der Lamina perpendicularis ossis ethmoidalis. HNO, 2003;5:142-5.
7. Osborn D.A. - Hemangiomas of the nose. J Laryngol Otol., 1959;73:174-9.
8. Iwata N., Hattori K., Tsujimura T. - Hemangioma of the nasal cavity: a clinicopathological study. Auris Nasus Larynx, 2002;29:335-9.
9. Dillon W.P., Som P.M., Rosenau W. - Hemangioma of the nasal vault: MR and CT features. Radiology, 1991;180:761-5.
10. Itoh K., Nishimura K., Togashi K., Fujisawa I., Nakano Y., Itoh H., et al. - RM imaging of cavernous hemangioma of face and neck. J Comput Assist Tomogr., 1986;10:831-6.
11. Kim H., Kim J.H., Kim J.H., Hwang E.G. - Bone erosion caused by sinonasal cavernous hemangioma: CT findings in two patients. AJNR, 1995;16:1176-8.
12. Hayden R.E., Luna M., Goepfert H. - Hemangiomas of the sphenoid sinus. Otolaryngol Head Neck Surg., 1980;88:136-8.
13. Jungheim M., Chilla R. - Der interessante Fall Nr. 64 = The monthly interesting case - case no. 64. cavernous hemangioma. Laryngorhinootologie, 2004;83:665-8.
14. Jammal H., Barakat F., Hadi U. - Maxillary sinus cavernous haemangioma: A rare entity. Acta Otolaryngol., 2004;124:331-3.
15. Enzinger F., Weiss. - Soft tissue tumor. 2nd edition. St Louis: Mosby, 1988.
16. Archontaki M., Stamou A.K., Hajioannou J.K., Kalomenopoulou M., Korkolis D.P., Kyrizakis D.E. - Cavernous haemangioma of the left nasal cavity. Acta Otorhinolaryngologica Italica, 2008;28:309-311.
17. Sciarreta V., Pasquini E., Frank G., et al. - Endoscopic treatment of benign tumours of the nose and paranasal sinus: A report of 33 cases. AMJ Rhinol., 2006;20:64-71.
18. Ahad A., Christi R.P. - Haemangioma of the maxilla. IJOL, 1977;29:194.