

Polypoid Angiomyofibroblastoma Tumor of Nasal Cavity: Case Report

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ABSTRACT

We report an extremely rare case and localization of polypoid angiomyofibroblastoma tumour, a case report and review of the world literature concerning angiomyofibroblastoma tumour. We present the case of a 74-year-old man who underwent left anterior and posterior ethmoidectomy with extirpation of tumour mass from left nasal cavity, epipharynx and left sphenoid sinus. The prognosis for this group of tumour is good and patient didn't receive any kind of therapy except surgical treatment. To our knowledge, this is a rare report in the world literature of polypoid angiomyofibroblastoma tumour of nasal cavity. This case indicates that angiomyofibroblastoma tumour of nasal cavity and paranasal sinuses is a rare disease including its localisation which otolaryngologists should be aware of, and one which should be included in the differential diagnosis of tumours involving sinonasal tract.

Key words: polyps, angiofibroma, nasal cavity

Introduction

The fibro-myofibroblastic tumour and tumour-like tumour lesions that can arise from the oral cavity include nodular fasciitis, solitary fibrous tumor, myofibroma/myofibromatosis, inflammatory myofibroblastic tumor, and low-grade myofibroblastic sarcoma¹⁻⁴. Although subtle morphologic and immunohistochemical differences between these lesions do occur, some cases show morphological overlapping or hybrid features⁵⁻⁸, indicating they are histogenetically related lesions arising from common precursor cell^{5,8-12}. Accordingly, some authors doubt that these tumors represent distinct entities, suggesting that they could be better viewed as morphologic variations of the same disease^{9,12}. Even these variations of the previously described tumors were reported in oral cavity localisation and they are extremely rare, we report on an unusual tumor occurring as a polypoid mass in left nostril and epipharynx arising from posterior part of left ethmoid cells and left sphenoid.

Case Report

A 74-year-old man was referred to our hospital in October 2009 complaining of complete left side nasal obstruction. His nasal obstruction started 1 year ago and it was progressively worse especially last few months. His past medical history was significant for hypertension over the past 10 years and the family history was negative for possible congenital lesions.

Nasal endoscopy with anesthesia revealed deviation of the nasal septum and polypoid-like tumour also visualised with posterior rhinoscopy. Anesthesia of nose cavity did not help him at all of relieving his symptoms of nasal obstruction. The neck examination was clinically negative.

Computed tomography scans revealed a tumour mass in the posterior part of left nasal cavity with spreading in epipharynx, obstructing proximal 3/4 of left nostril and in cranial direction polypoid mass filling of ethmoid cells and left sphenoid sinus (Figure 1 and 2).

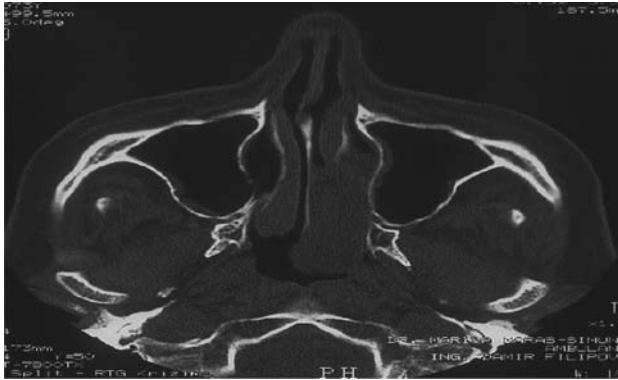


Fig. 1. Transversal computed tomography showing a tumour mass in left nasal cavity with spreading in epipharynx and partly in oropharynx obstructing proximal 3/4 of left hoana.

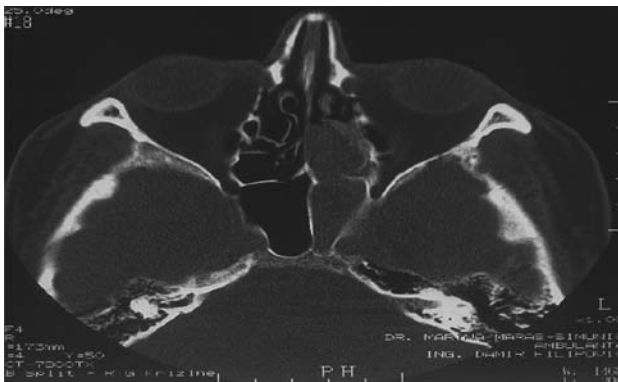


Fig. 2. Transversal computed tomography showing a tumour mass in left nasal cavity with spreading in left ethmoidal cells and in left sphenoidal sinus.

Under general anaesthesia, left anterior and posterior ethmoidectomy was performed with extirpation of whole tumour mass from left nasal cavity, epipharynx and left sphenoid sinus. Before that, reconstruction of nasal septum was performed for better visualising and removing of tumour mass. Endoscopically, on operation tumour mass was not looking like a polyp, and operation was performed with minimal bleeding.

Three pieces of tumour mass measuring 2x1 cm, 1x1 cm and 1x0.5 cm was sent for patohistological analysis. Microscopic examination showed circumscribed, oval lesion composed of uniform medium-sized oval cells with pale eosinophilic cytoplasm with oval nuclei and small nucleoli. Stromal part of the tumour was consisted of oedematous collagen with lot of small blood vessels. Mitotic activity was absent (Figure 3).

Immunostaining was preformed on the tumour showing expression of CD34, desmin and pan muscle actin. Positivity for α -smooth muscle actin was only observed in association with the walls of small blood vessels. The morphology and expression of CD34, skeletal muscle markers and desmin confirmed the diagnosis of angiomyofibroblastoma.

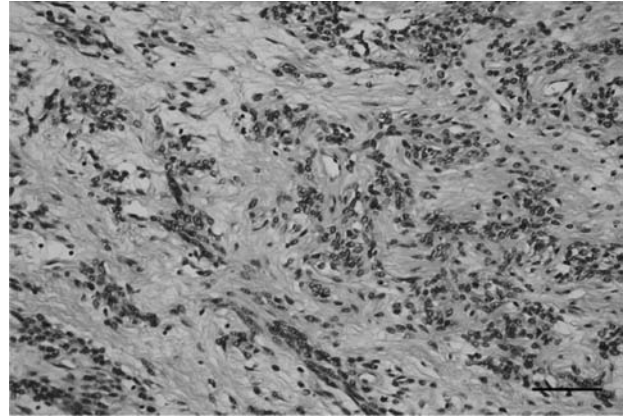


Fig. 3. Photomicrograph showing the tumour composed predominantly of a relatively uniform population consisting of small- to medium sized, round to oval- to epithelioid-shaped cells, pale eosinophilic cytoplasm and round to oval nuclei with finely dispersed chromatin and small nuclei, embedded in a prominent myxoid/ oedematous stroma rich with small blood vessels. (H&E; original magnification indicated by scale bar corresponding to 100 μ m).

Post-operative recovery was very good. After the consultation with oncologist the patient was reviewed monthly. Nine months after the operation, control CT was made

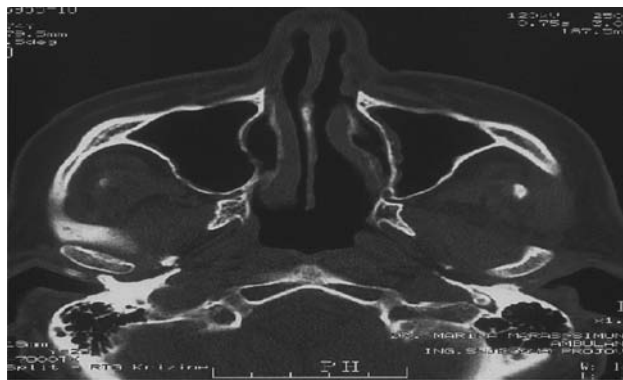


Fig. 4. Transversal computed tomography showing left nasal cavity, epipharynx and oropharynx free of tumour mass 9 months after surgery treatment.

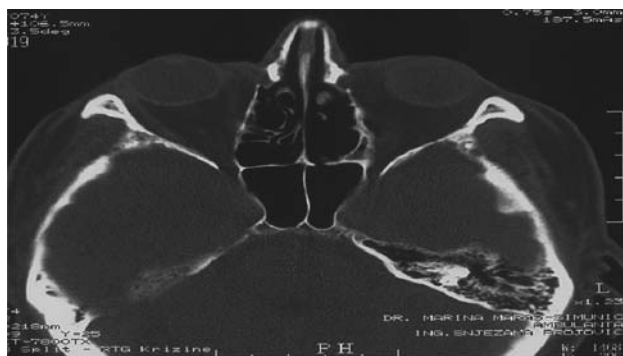


Fig. 5. Transversal computed tomography showing left ethmoidal cells and left sphenoidal sinus free of tumour mass 9 months after surgery treatment.

and there were no signs of either local tumour recurrence or distant metastasis (Figure 4 and 5). Clinical examination and fiberoendoscopy was also negative for tumour recurrence.

Discussion

The tumour that we presented and that was pathologically described as angiomyofibroblastoma is extremely rare tumour with only few cases documented worldwide. Previously documented these kinds of tumour were reported in oral cavity and we reported it for the first time in sinonasal area. It is very difficult to make an accurate diagnosis of this kind of tumour because of lack of standardized diagnostic criteria. Microscopic examination showed circumscribed, oval lesion composed of uniform medium-sized oval cells with pale eosinophilic cytoplasm with oval nuclei and small nucleoli. Stromal part of the tumour was consisted of edematous collagen with lot of small blood vessels. Mitotic activity was absent. Immunostaining was performed on the tumour showing expression of CD34, desmin and pan muscle actin. Positivity for α -smooth muscle actin was only observed in association with the walls of small blood vessels. The morphology and expression of CD34, skeletal muscle markers and desmin confirmed the diagnosis of angiomyofibroblastoma tumour. Although a longer follow-up period is necessary, the absence of atypical mitoses, nuclear pleomorphism, and necrosis suggests a favourable prognosis for the present case.

In our case follow up period was nine months with no evidence of recurrence of tumor. We suggest that treatment of polypoid angiomyofibroblastoma tumor should include complete resection of the tumour.

This is further supported by the fact that biological behaviour of angiomyofibroblastoma of the lower female genital tract or extragenital sites is usually benign, with

only a low recurrence after complete surgical excision. Although, fibro-myofibroblastic tumours usually have low mitotic activity, some cases- especially AMF and cellular angiofibroma (CAF) may show brisk mitotic activity (up to 11 mitoses per 10 high power fields)^{13–15}. Tumour that we report doesn't show any mitotic activity and accordingly small chances for local recurrence but because these stromal tumours are closely related lesions arising from a similar fibroblastic-like progenitor cell native to hormonally responsive specialized stroma¹², we should be aware of recurrence and future possible transformation and differentiation of AMF tumour.

Conclusion

We report a unique case of polypoid angiomyofibroblastoma tumour in sinonasal area location with characteristic radiological, histological and immunohistochemical features. These kinds of tumours are rare and difficult to diagnose. Otolaryngologist should be aware of this tumour, because, even these tumours are usually benign with only a low recurrence rate after complete surgical excision, some of them may show brisk mitotic activity and rarely they could have malignant transformation. That is one of the reasons why we suggest longer follow up period for this kind of tumour. Also, angiomyofibroblastoma should be included in the differential diagnosis of lesions involving the sinonasal tract. Additional new cases are necessary to better understand the clinical-pathologic features of this unusual »fibro-myofibroblastic lesion«, for which the descriptive term »polypoid angiomyofibroblastoma-like tumour« seems to be appropriate. To conclude, even this kind of tumour are showing expansion and attending to grow, they are not showing any kind of invasion process what is very important factor for overall prognosis and treating for AMF tumours.

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POLIPOIDNI ANGIOMIOFIBROBLASTOM – TUMOR NOSNE ŠUPLJINE: PRIKAZ BOLESNIKA

S A Ž E T A K

Cilj nam je bio prikazati izuzetno rijedak slučaj i lokalizaciju tumora nosne šupljine – angiomyofibroblastom te njegovu lokalizaciju. Pregledali smo i analizirali gotovo svu dostupnu svjetsku medicinsku literaturu vezanu uz ovaj izuzetno rijetki tumor. Prikazali smo 74 – godišnjeg muškarca kojemu je učinjena lijeva prednja i stražnja etmoidektomija s odstranjenjem tumorske mase iz lijeve nosne šupljine, epifarinksa i lijevog sfenoidnog sinusa. Prognoza za ovaj tip tumora je dobra te bolesnik nije primio nikakvu drugu terapiju osim kirurške. U zaključku, ovo je izuzetno rijetki tumor po svojim patohistološkim karakteristikama, kao i po lokalizaciji te bi ga svaki otorinolaringolog trebao imati u vidu kada razmišlja o diferencijalnoj dijagnozi tumora nosnih šupljina i sinusa da bi bolje planirao optimalno liječenje.