

Coexistence of Glomangioma and Yolk Sac Tumour in a Child: A Case Report

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ABSTRACT

Glomus tumours (GTs) primarily arise from glomus bodies that are located in the dermis layer of skin. However, they can be encountered ectopically in most parts of the body. As a result of researches done in a 17-month-old male patient who presented to us with complaint of an increasingly growing swelling, he was diagnosed with a yolk sac tumour. Chemotherapy was started and then, he was operated. GT was found inside the tumoural mass. Our patient, who is the first case according to our knowledge, where the concurrence of yolk sac and glomus tumours was reported, has been discussed in the light of literature.

Keywords: Glomus bodies, Yolk sac tumour

CASE REPORT

A 17-month-old male patient presented to our hospital with complaint of swelling in hip and inability in sitting. Swelling had occurred about one month ago, it had enlarged increasingly and it had become a condition that prevented him from sitting. During his physical examination, a mass was identified in the sacrococcygeal region. During magnetic resonance imaging (MRI) studies which were done, the mass was seen to invade into the gluteal muscle, posterior to the rectum and it was reported as a teratoma. In thoracic tomogram, a nodular image which was compatible with metastasis in the lower lobe of the right lung was seen. Alpha fetoprotein was 68309 ng/mL.

It was diagnosed as a yolk sac tumour during a histopathologic investigation of the material which was done, which was received by doing a tru-cut biopsy. The patient was discharged after 3 he took courses of BEP (Bleomycin, Etoposide, Cisplatin) treatment. A post-operative tumoural mass necrosis and the presence of a GT inside this mass were identified. In the histopathologic examination, glomus tumour was found to consist of uniform, round or oval cells with round or oval nuclei and pale eosinophilic cytoplasm. Tumour cells were arranged in sheets or nests patterns, and they were found to surround vessels. The tumour cells were stained by PAS and they were diffusely positive for SMA [Table/Fig-1]. The MIB-1 index was about 1-2%. Glomus tumour was diagnosed on the basis of these features. Chemotherapy of the patient was continued. The patient is still being followed up by us without any problem.

DISCUSSION

Glomus bodies are neuromyoarterial receptors that surround cutaneous arteriovenous anastomoses and are located in reticular dermis. Histologically, they include a shunt system with an efferent venule and an arteriole which are connected with an anastomosis. Even though these anastomoses are considered as the source of tumour, the other possible origins such as neural crest and pericytes cannot be excluded, because tumour is also seen there where glomus bodies such as nerve, bone, stomach, mediastinum, rectum, mesentery and tongue are not found [1].

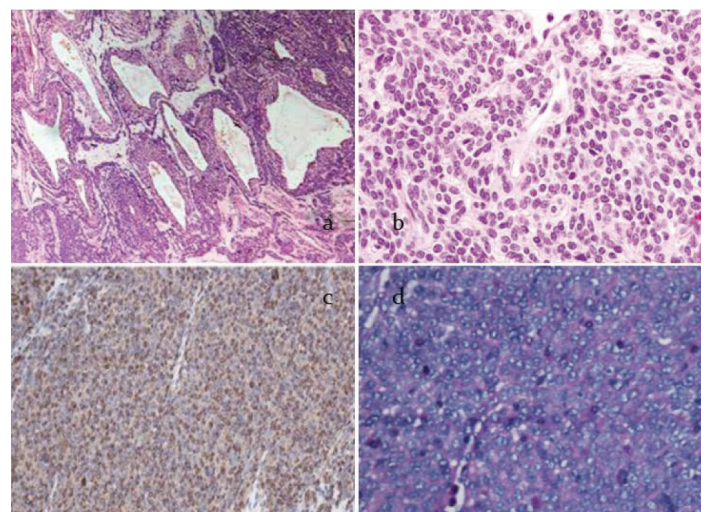
GTs are single and nonfunctional lesions seen in the mid-adult age group. They are more frequently seen in women than in men [2]. Approximately 10% of tumour, which is sporadic in fact, is familial

and it shows autosomal dominance [3]. Among such cases, 10% are bilateral or multicentric [4].

Clinical symptoms are tinnitus, pain, difficulty in swallowing, hoarseness, pulsatile mass, headache, nasal obstruction, hearing loss and anaemia, which are based on type of mass [5]. They are neuroendocrine tumours [2] and they may also lead to a hypertensive crisis during surgery [6]. Our patient was a 17-month-old male who presented to us with a complaint of a swelling in hip. However, this swelling was dependent on a yolk sac tumour and not on a Glomus tumour.

Tumour has a benign nature mostly, but it has also been reported that it cause malignant changes by 3-10% and metastasis by 2% [5]. Metastatic spreads frequently occur in regional lymph nodes [2].

Methods which are mostly applied for making a diagnosis are ultrasound and MRI [7]. In our case, the main mass was reported as a teratoma by MRI and Glomus tumour inside it, which was unable to be seen. Angiography is particularly important in determination of the presence of multicentric tumour and its vascularity [8]. Measuring



[Table/Fig-1]: (a) Dilated blood vessels and glomus cells (Hematoxylin-eosin, original magnification, X 100); (b) Round nuclei and pale eosinophilic cytoplasm of the tumors (Hematoxylin-eosin, original magnification, X 400); (c) Tumor cells showing cytoplasmic positivity for SMA (SMA, original magnification, X 200) (d) Tumor cells showing positivity for PAS (PAS, original magnification, X 400)

excretion of vanillyl mandelic acid, that is a breakdown product of catecholamine in functional tumours, may be useful for making a diagnosis [6].

Primary treatment which is given for these tumours is surgery. Radiotherapy is suggested for incomplete resection cases and for inoperable Glomus jugulare and rarely for carotid body tumours [9] or in addition to surgery, to control metastasis [10] and for the other hand, for bilateral cases where unilateral X and XII cranial nerves are sacrificed [8]. In our case, a GT was identified inside the mass which was excised by doing a surgical procedure which was performed after initially giving chemotherapy. While structure of yolk sac tumour gets necrotized completely with chemotherapy, it has been noticed that glomus tumour is protected. Follow-ups of our patient whose treatment was completed, continue without any problem.

REFERENCES

- [1] Iqbal A, Cormack GC, Seerri G. Hereditary multiple glomangiomas. *Br J Plast Surg.* 1998; 51: 32-7.
- [2] Pellitteri PK, Rinaldo A, Myssiorek D, Gary Jackson C, Bradley PJ, Devaney KO, et al. Paragangliomas of the head and neck. *Oral Oncol.* 2004; 40: 563-75.
- [3] Barnes L, Eveson JW, Reichart P, Sidransky D, editors. *Pathology and genetics of head and neck tumours* (World Health Organization classification of tumours). Lyon, IARC Press, 2005.
- [4] Manolidis S, Shohet JA, Jackson CG, Glasscock ME. Malignant glomus tumors. *Laryngoscope.* 1999; 109: 30-4.
- [5] Karas DE, Kwartler JA. Glomus tumors: a fifty-year historical perspective. *Am J Otol.* 1993; 14: 495-500.
- [6] Schwaber MK, Glasscock ME, Jackson SG. Diagnosis and management of catecholamine secreting glomus tumors. *Laryngoscope.* 1984; 94: 1008-15.
- [7] Wortsman X, Jemec GBE. Role of high-variable frequency ultrasound in preoperative diagnosis of glomus tumors. *Am J Clin Dermatol.* 2009; 10: 23-7.
- [8] Biller HF, Lawson W, Som R, Rosenfeld R. Glomus Vagale Tumors. *Ann Otol Rhinol Laryngol.* 1989; 98: 21-6.
- [9] Powell S, Peters N, Harmer C. Chemodectoma of the Head and Neck: Results of Treatment in 84 Patients. *Radiation Oncology Biol Phy.* 1992; 22: 909-24.
- [10] Cuhruk Ç, Aktürk T, Saatçi M, Yılmaz O. Carotid Body Tümörleri: *Türk Otorinolarenoloji. XX: Ulusal Kongresi* Girne, 1989;116-7.

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