Intraosseous orbital haemangioma

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We present a case of rare orbital roof haemangioma infiltrating the medial and lateral walls of the orbit, zygomatic bone and lesser and greater wings of the sphenoid. The tumour was totally excised by the frontolateral approach, followed by craniofacial and orbital roof reconstruction.

key words: haemangioma, orbital roof, intraosseous

Cranial haemangiomas are rare benign tumours comprising about 0.2% of all intraosseous neoplasms and less than 5% of them develop in the orbital bone. We present a case of large haemangioma of orbital roof infiltrating lateral orbital wall, orbital margin zygomatic bone, lesser and greater wings of the sphenoid.

CASE REPORT

A 57-year-old female mathematics teacher complained of periodic headache in right frontal area and difficulty in writing marks in the class register for 5 months. The difficulty lay in writing the marks not in a proper line, but one line over the proper surname. The patient’s daughter noticed deformation in the area of right orbit in the form of a tumour of the upper eyelid with eye ball dislocation downwards. General physical examination revealed unmovable palpable tumour in the right orbital margin. The right eyeball was lower than the left one, fixed in a slight divergent and vertical squint. Ophthalmologic examination revealed immovable palpable tumour in the right orbital margin. The right eyeball was lower than the left one, fixed in a slight divergent and vertical squint. Ophthalmologic examination revealed immovable palpable tumour in the right orbital margin. The right eyeball was lower than the left one, fixed in a slight divergent and vertical squint. 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cularity of the tumour, and this tumour was vascularised from a frontal branch of the right facial artery. Embolisation was not performed due to the small size of the vessels feeding the tumour.

**SURGICAL PROCEDURE**

Following fronto-temporal craniotomy, the tumour was excised with the unaffected right frontal bone with orbital margin, 2/3 of orbital roof, part of zygomatic bone, lesser and greater wings of the sphenoid and part of temporal bone in temporal fossa (Fig. 2). The tumour infiltrated frontal dura which was excised together with the tumour. The damaged dura was stuck with „iodura“ using „Beriplast-P“ glue. Cranioplasty was performed during the same surgical procedure, applying appropriately modelled „Codubix“ prosthesis.

The patient was discharged from hospital free of neurological symptoms and with improvement of right eyeball mobility. CT scan performed six months later showed almost normal cerebral and orbital picture. „Codubix“ prosthesis implanted in the layer of cranial bone gave good cosmetic effect.

**HISTOLOGY**

The specimen examined by pathologists revealed orbital bones with red bluish tumour in the central part measuring 45 × 35 × 25 mm. A bone cortex around the tumour was thin and fragile. On the cut surface, the tumour was reddish and porous, suggesting vascular neoplasm. Bone tissue surrounding the tumour was normal and of normal density, that is hard and poorly supplied with blood.

Microscopically, the tumour consisted of thin-walled, wide vascular canals lined by endothelial cells, partly filled with blood. They were separated by thin stroma with numerous collagen fibrils and few pericytes. Small bone trabeculae were found between the vascular stroma (Fig. 3). The diagnosis of intraosseous cavernous haemangioma was made.

**DISCUSSION**

Intraosseous cranial bone haemangioma is very rare and comprises 0.2% of bone neoplasms [2, 3, 9]. Frontal and parietal bones are most frequently affected [9]. Less then 5% of these tumours are found in orbital roof, and only 25 cases have been published so far [3, 7, 9]. The first case of such haemangioma was described by Rowbothan in 1942 (Rowbothan 1942). The tumour may be observed at any age but is most frequent in the fifth decade (31.9%) equally in both sexes [9]. Cranial bone haemangioma most commonly occurs in one orbital bone and may spread onto surrounding bones such as zygomatic bone, sphenoid bone and ethmoid bone. One of the large tumours described in the literature infiltrated frontal bone, orbital rim and larger wing of the sphenoid bone [5].

The most typical clinical manifestations are: proptosis and diplopia, ophthalmoplegia, diminution of vision, nasal obstruction, epistaxis, swelling around the orbital rim.

In the case of intraosseous haemangioma, basic X-ray and CT scan showed reduced density of the bone surrounded by higher density zone [2, 7, 9]. Intensive vascularisation characteristic for osseous haemangioma is presented in MRI [2, 7].
Pathogenesis of haemangioma is not clear. Injury is thought to be the cause of tumour development [9]. However, the tumour is suggested to be a hamartoma of mesenchymal origin which differentiates into vascular elements which may be stimulated by injury, ischaemia or other factors, and angioblastic response in the bone tissue.

Histologically there are two types of cranial haemangiomas: cavernous and capillary. Cavernous haemangioma is the most frequent and consists of vascular areas between endothelial spaces and of osteoblasts and osteoclasts in the injured bone, suggesting permanent process of bone restructuring (remodelling) [2, 7]. The capillary type is rare, characterised by numerous tortuous pathways of small blood filled spaces. Some haemangiomas are mixed types most common in calvaria.

Haemangiomas are very slow-growing bone tumours and should be differentiated from other bone tumours such as: fibrous dysplasia, aneurysmal bone cyst, dermoid cyst, meningioma, osteoma and — rarely — osteogenic sarcoma. Haemangiomas are in the majority diagnosed only after surgery or biopsy [2, 7, 9].

Surgery is the treatment of choice in the case of orbital roof haemangioma. Surgical procedure strategy lies in total excision of the tumour around unaffected tissue in order to avoid bleeding from this well vascularised neoplasm. In our case fronto-lateral approach was applied (similarly to Spetzler) to excise orbital roof together with orbital rim, a part of lateral orbital wall, and frontal part of zygomatic bone [8]. Presurgical embolisation was not applied, due to small feeder vessels from external carotid artery. Fronto-lateral approach preserves olfaction and optic nerve, and visualises orbital structures in orbital apex. Radical surgery consisting of tumour excision together with bone excision causes a large defect of facial part of the cranial bone. Cranio-plastic surgery is necessary to protect from injury and due to aesthetic needs. We used Tricomed „Codubix” prosthesis after proper spatial modification enabling orbital roof with rim and cranium frontal part restoration [1, 4].

CONCLUSION
Cranial haemangiomas are usually benign tumours rarely found in orbital roof.
Total tumour excision is a procedure of choice.
Fronto-lateral approach enables radical treatment of this type of neoplasm with safe preservation of orbital structures.

REFERENCES