OCULAR MOTILITY DISORDERS WITH DIPLOPIA AS THE FIRST SYMPTOMS OF PARANASAL TUMOURS WITH ORBITAL INVASION

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SUMMARY

OCULAR MOTILITY DISORDERS WITH DIPLOPIA AS THE FIRST SYMPTOMS OF PARANASAL TUMOURS WITH ORBITAL INVASION – A CASE REPORT

Presentation of two case reports about our experiences with diagnostics of paranasal tumours in patients with diplopia or ocular motility disorders which were the first symptoms of these tumours. Furthermore, the following diagnostic and therapeutic procedure which has been conducted in cooperation with other hospital departments is presented. The first case report is an example of primary paranasal tumour, the second case report represents secondary infiltration of maxillary sinus and orbital invasion.

Methods: A case reports.

Key words: diplopia, paranasal tumours, orbital invasion, spinocellulary carcinoma

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INTRODUCTION

The causes of diplopia in tumours invading into the orbit from the surrounding structures may be either mechanical (dislocation of bulb and/or extraocular muscle by means of a tumorous process in the orbit) or neurogenic (paresis of oculomotor nerves, in particular due to pressure in the region of the apex of the orbit). Within the framework of differential diagnostics upon examination of a patient with diplopia, paranasal tumours do not rank in first place, but should nevertheless not be overlooked.

When examining a patient with diplopia we pay attention to the position and aspect of the eye. It is necessary to evaluate deviation (heterotropia) of the bulb (deviation in the position of the eye due to paralytic strabismus not only due to a malfunction of the afflicted muscles, but above all due to hyperfunction of antagonists of the paretic muscles). We evaluate primary and secondary deviation. We also examine the motility of the bulb in all directions of gaze and determine the correlation between subjective perception of diplopia and the deviation of the eye. We must also pay attention to any applicable compensatory positioning of the head.

Upon evaluating the position of the eye we assess the dislocation of the bulb on the frontal level and also in the sagittal axis. We therefore determine whether this concerns exophthalmos or enophthalmos, axial or paraxial.

Within the framework of differential diagnostic consideration in examining diplopia, we also focus attention on the appearance of the surrounding area of the eye (swellings, retraction of the eyelids, ptosis). We also should not overlook evaluation of nystagmus, which need not necessarily be evident in primary position, and appears or is accentuated in extreme gaze positions. On the anterior segment of the eye we focus on injection, chemosis and pupillary response, we also examine sensitivity of the cornea. On the fundus we are interested especially in the appearance of the papilla of the optic nerve, upon protrusion of the bulb we also look for draping of the retina. It is also always appropriate to examine the perimeter. We complete the examination with a palpation examination of the orbital entry in order to exclude resistance.

Of other symptoms not directly connected with the eye, we evaluate and inquire about sensory malfunctions in the innervation region n. V., paresis n. VII., nasal blockage and secretion, disorders of mobility, stability, feeling of dizziness, nausea, tinnitus, hearing disorders.

From the anamnestic data we inquire in the personal and family anamnesis with regard to vascular diseases, diabetes, demyelinating diseases, neurological disorders (e.g. epilepsy, myasthenia gravis), disorders of the thyroid gland and tumorous pathologies (5, 9, 10, 11, 14).

Case report no. 1

A man aged 62 years reported to our centre due to swelling of the eyelids of the left eye, persisting for approx. 14 days, and a feeling of blurred and double vision. He had never undergone any therapy, but had not been to the doctor for 30 years, does not even have a general practitioner, smokes 30-40 cigarettes per day and drinks beer almost every day.

From the examination I present only the pathological findings:

In primary position the left eye (LE) is in hypertrophy and mild exotropia, in an alternative covering test the right eye

(RE) draws downwards and outwards, LE upwards and outwards. Motility of the left eyeball is limited especially downwards and temporally. The patient states diplopia in the entire left half of the visual field, upon gaze max. downwards and max. upwards, states maximum disparity of images in upper left. Images are shifted vertically and horizontally. The patient repeatedly states that the peripheral image is from the RE. On the Worth light test he sees 5 lights. The ocular apertures are asymmetrical, in the LE the ocular aperture is narrower 6 mm, in the RE 9 mm, anisocoria, pupil of RE narrower, LE wider, they respond to illumination, the left pupil slowly. Papilla in left eye with wider excavation than in right. We sent the patient for CT of the head. In differential diagnostics we considered the tumorous process in the left orbit (tumour, pseudotumour)/ tumour invading into orbit from surrounding area/ inflammatory affliction of paranasal cavities with incrushing of contents into orbit (less probable, patient does not manifest symptoms of such extensive inflammatory pathology)/ endocrine orbitopathy (less probable, patient has never undergone treatment of thyroid gland, has no blepharal symptoms or protrusion of the bulb).

On CT of the head pathological matter of the left maxillary sinus were identified, spreading into the left ethmoid sinus, left nasal cavity and left orbit, forcing out the left eyeball, usurping the adjacent skeleton – v.s. tumour, dense content of right maxillary sinus – v.s. sinisitis maxillaris I.dx.

At a follow-up examination with the result of the CT after an interval of 14 days the patient perceives disruptive double vision following lifting of the eyelid. To targeted questioning the patient states suffering from a cold persisting for approx. 1-2 months, without infusion with blood, feeling of blocked nose. Slight deterioration of vision occurred to 5/6 no. with +3.0D sf., J.no.2 with + 6.0D sf.,

In primary position bulb of LE is clearly dislocated upwards and outwards and slightly in exotropia, eyelid covers entire pupil, motility of left bulb limited especially downwards and temporally, patient states diplopia in entire left half of visual field and in max. downward and max. upward gaze, ocular apertures asymmetrical, in LE ocular aperture narrower – 5 mm, in RE 9 mm, in palpation lower eyelid with edema and hard matter in depth of eyelid, anisocoria, pupil of RE narrower, LE wider, respond to illumination, in left slowly.

Due to suspicion of a tumour the patient was sent for examination to ORL, where MR of the head was indicated.

According to MR of the head there is a pathological formation of spherical shape, irregular contours, size approx.

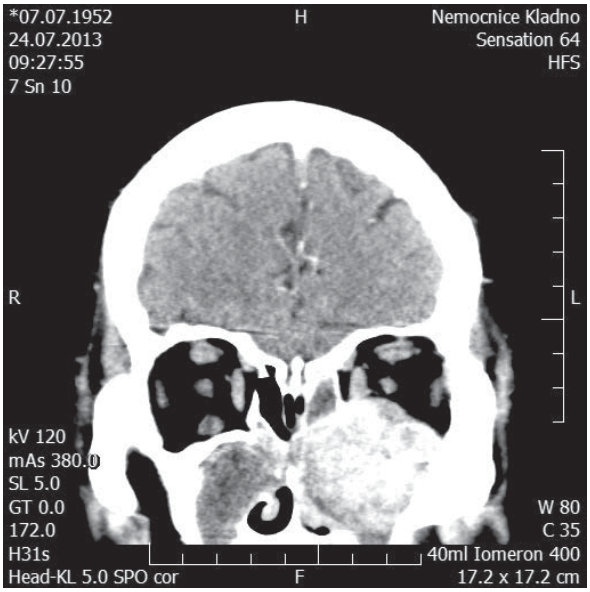

Fig. 1 CT of head – coronary cross-section

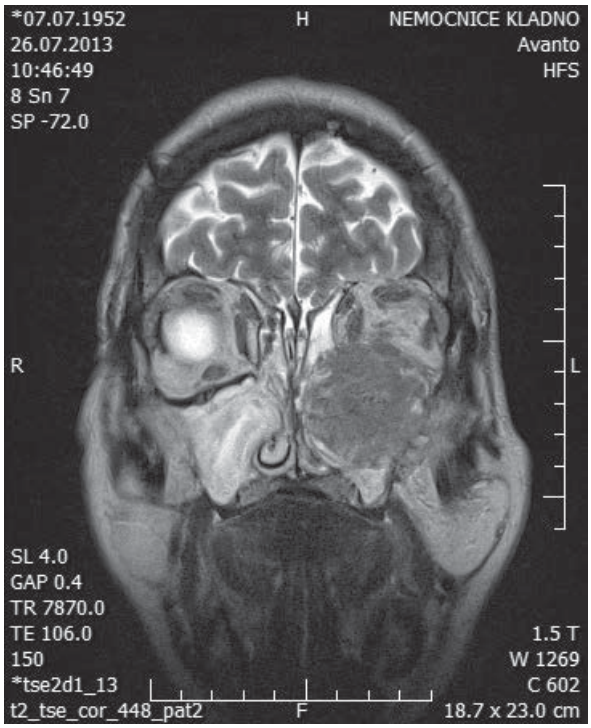

Fig. 2 MR of head – coronary cross-section

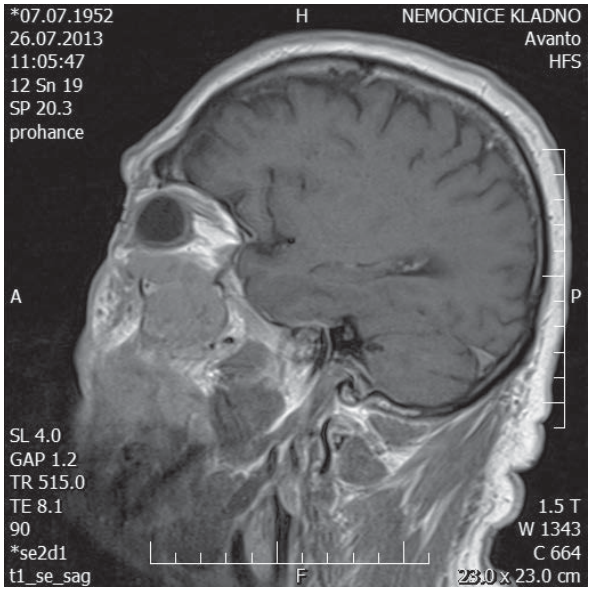

Fig. 3 MR of head – sagittal cross-section

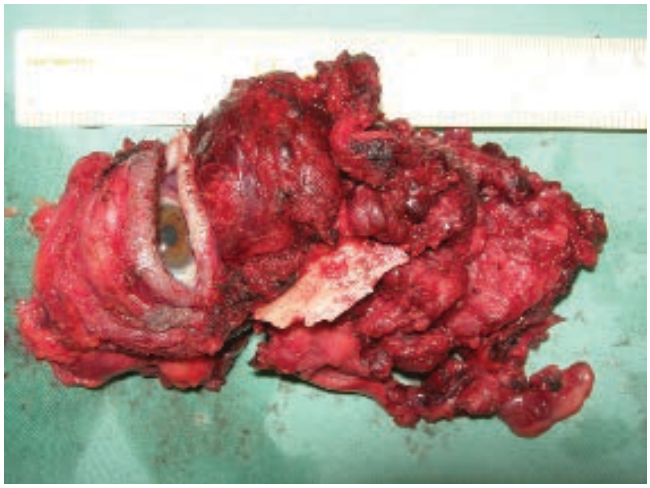

Fig. 4 Resected area

43x45x48 mm, T2 and T1 isosignal and post-contrast with relatively evenly saturating, entirely filling left maxillary sinus, destroying surrounding bones (wall and ceiling of left maxillary sinus, walls of left ethmoids, partially also nasal septum, v.s. infiltrating small wing of left cuneiform), spreading into left nasal cavities, ethmoid cavities, left orbit, fossa infratemporalis with infiltration of pterygoid muscle, ventrally soft tissues of left cheek. The base of the left orbit

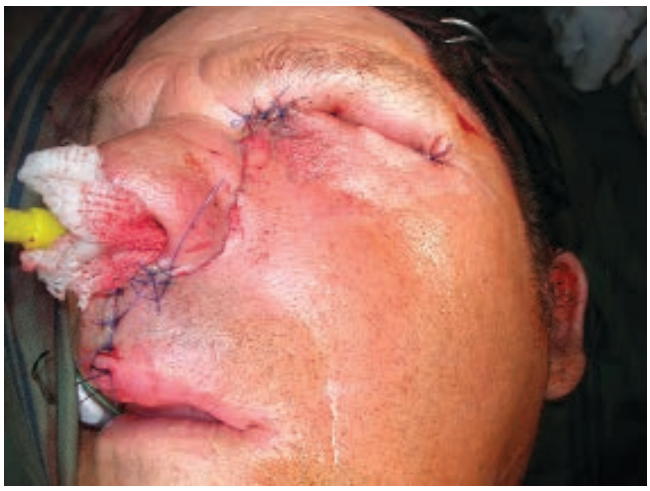

Fig. 5 Finding after end of surgical procedure

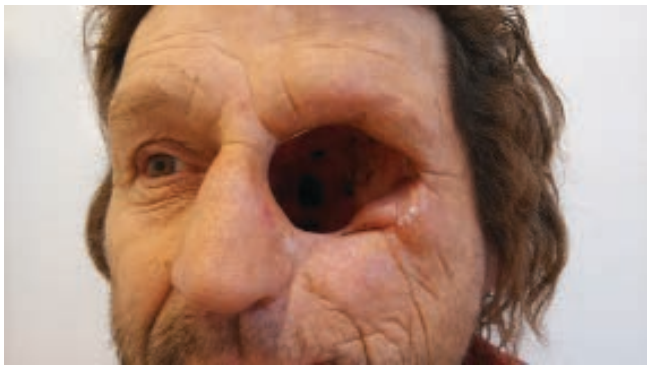

Fig. 6 Postoperative cavity two years after procedure

Fig. 7 Patient with affixed epithesis

is destroyed and pathological matter is spreading into the lower part of the left orbit, with a close relationship to the m. rectus inferior, dislocating it and the eyeball cranioventrally. Peripherally it protrudes from the nose into the nasopharynx.

Pathological content of right maxillary sinus and wall changes of frontal cavity – v.s. sinusitis.

Intracranially only numerous small non-specific deposits in white matter frontally bilaterally, v.s. post-ischemic or post-inflammatory.

Colleagues from ORL department sent the patient for examination and proposal for surgical procedure to the otorhinolaryngology and head and neck surgery clinic at the Motol University Hospital. A sample was first of all taken from the patient for a histological examination, and one month after the determination of the diagnosis on CT of the head the definitive surgical solution was performed (maxillectomia subtotalis, exenteratio orbitae sin., plastica rhinobasis, dissectio nodi lymphatici colli l. sin. I-V).

Histologically this concerned carcinoma spinocelularis sinus maxillaris et orbitae I. Sin. PT4pN0. Subsequently the patient underwent chemotherapy and radiotherapy, according to a control CT of the head one year after the procedure he was without signs of recurrence. At present, after more than 2 years the patient is still without signs of recurrence, subjectively without pain or complaints. The postoperative cavity is pacific, the patient has post-radiation changes externally on the neck. The patient covers the postoperative cavity alternately with a bandage and an epithesis, which objectively fulfils its purpose, but subjectively the patient is not entirely with the colour of the epithesis, which he considers to be too dark, especially in the winter months, since it was made in the summer when he was markedly suntanned.

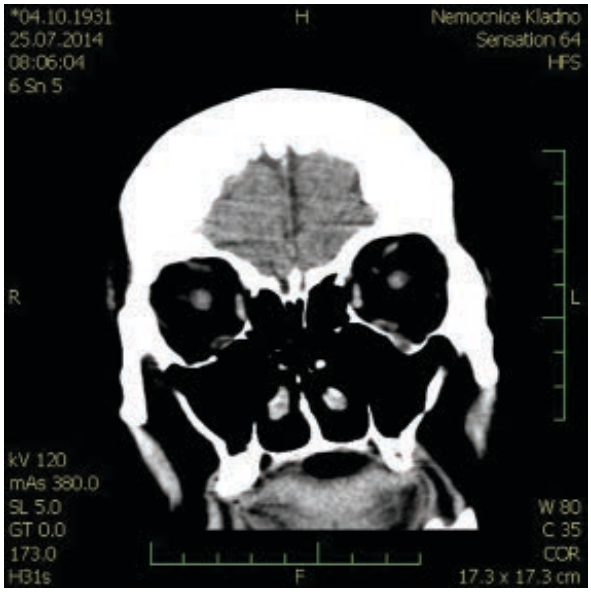

Fig. 8 CT of head – coronary cross-section

Case report no. 2

A female patient aged 83 years, in our care from May 2014, was sent to us by an outpatient ophthalmologist due to lagophthalmos in the left eye and hydrophobic lesion of the cornea. Paresis n. VII. originated following
removal of a recurrence of a spinocellular carcimona on the left cheek in the region above the maxillary sinus on the interface with the lower eyelid (first operation 1 October 2013, re-operation 30 October 2013). At the time of the first examination at our centre the patient also stated pain in the innervation region of the 2nd trigeminal branch, which she however connects with the extraction of a tooth in March 2014. At our department partial tarsorrhaphy was performed on the left eye, the patient attends regular follow-up examinations, the finding on the cornea is stabilised. At present the patient is in the care of a neurologist for paresis n. VII. and neurological pain in the left cheek.

At a planned follow-up examination in June 2014 we determined deterioration of vision in the LE to movement, certa, finding on the anterior segment without change, newly dragging abduction of LE, eye does not draw over central line.

For this reason we sent the patient for a control neurological examination with suspected paresis n. VI., in which CT of the head was indicated. On CT of the head only minor older ischemic changes in the right eye were described in the region of the basal ganglia and periventricularly, suspected st.p. Otitis media chron. I. sin., bone deposit in occipital bone rather giving impression of benign lesions, plasmocytoma less probable, recommended laboratory exclusion.

At the turn of July and August 2014 a droop of the upper eyelid of the LE appeared, as a result of which the patient sought a neurologist. At our follow-up examination in August 2014 the patient stated worsening droop of the upper eyelid, which covers the entire pupil. We further determined pronounced deterioration of motility of the left bulb, which was virtually immobile, discrete movement only to elevation and depression. Scar on interface of lower eyelid

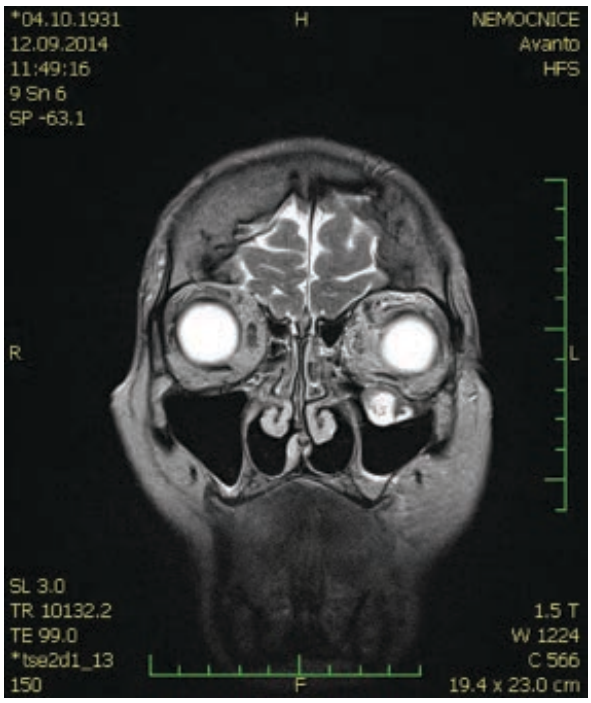

Fig. 9 MR of head – coronary cross-section

and cheek on left side pacific, without signs of recurrence of skin tumour.

We indicated MR of the head due to progressing ptotis of the upper eyelid and total ophthalmoplegia. According to the description of MR of the head, a pathological deposit is bulging into the base of the left orbit, propagating from the hypodermis by the lower part of the left eye socket, encroaching into the maxillary sinus and causing a defect of the bone ceiling of the left maxillary sinus – base of orbit, bulging partially into base of orbit extraconally, dislocating the lower extraocular muscle cranially, the deposit encroaches up to half of the depth of the orbit, its total size is approximate-

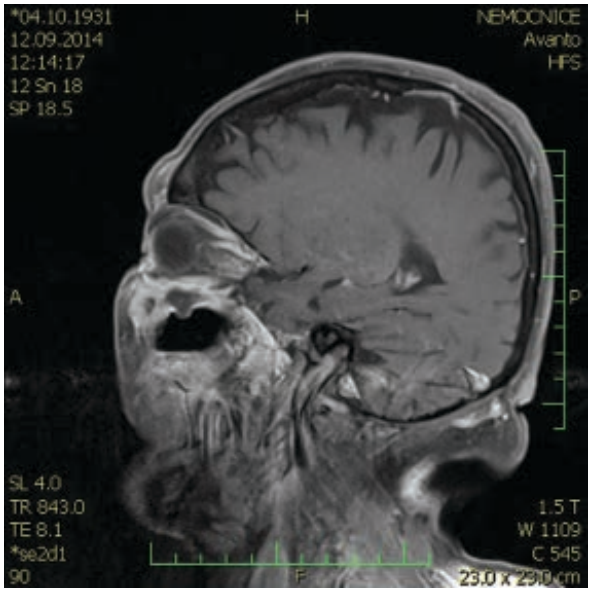

Fig. 10 MR of head – sagittal cross-section

ly 14x16x35 mm, irregular shape, T2 heterogeneous signal, centrally predominantly T2 hypersignal, T1 hyposignal, post-contrast there is pronounced opacification of deposit, central parts are not opacified. Suspected atrophy of stem n. V. left.

The patient was examined by a dermatologist and oncologist at the otorhinolaryngology and head and neck surgery clinic at the Motol University Hospital, a surgical procedure was not indicated. In November 2014 a control MR of the head was performed. Upon comparison with MR from September 2014 the pathological deposit in the region of the left cheek is in progression – propagating into the base of the left orbit and the left maxillary sinus, intracranially, into the masticating muscles on the left side and v.s. infiltrated and parotid gland on the left side. The scope of infiltration of the bone is also larger, intracranially the adjacent gyry of the left temp. lobe are slightly forced out with v.s. irritation of meninges in this region.
With regard to the progression of the finding, palliative radiotherapy was indicated. The patient came to an ocular follow-up examination after the end of radiotherapy at the end of January 2015, at which the finding was dominated by pronounced asymmetry of the face with ptosis of the left eyebrow by approx. 1.5 cm, total ptosis of the upper eyelid, bulb of LE entirely immobile, and also there was newly present post-radiation erythema of the eyelids and left cheek. The finding on the cornea was stable.
At the end of April 2015 there was a progression of the finding, ulceration of the skin above the maxillary sinus appeared, perforation between the maxillary sinus and nasal cavity, and the other with the oral cavity. The patient died 11 months after the origin of the first oculomotor malfunction with a deep defect of the cheek into the maxillary sinus.

DISCUSSION

Both primary and secondary orbital tumours represent a problem which requires a multi-disciplinary approach to therapy. In the Czech and Slovak professional ophthalmological literature we find a small number of articles devoted to primary tumours of the orbit, but the statistical data about secondary tumours is very sparse. According to a thirteen year study by Slovak authors published in 1994, malignant tumours comprised 43% of tumours of the orbit. In their cohort 30% of orbital tumours were secondary, represented most frequently by menigiomas (10%), carcinomas of the eyelids (basalioma and epidermoid carcinoma) (10%), whereas tumours invading from the paranasal cavities represented only 1.4% (3). Krásný et al. came to a similar conclusion with regard to the number of malignant tumours in a ten year study of orbital tumours. According to a study from 2008, lymphomas formed 45%, other malignant tumours concerned carcinomas of the orbitopalpebral region in 31.5% of cases, growth of conjunctival melanoma into the orbital region in 13% of cases, basalioma was diagnosed in 7.5% and sarcoma in 2.5%. Secondary tumours were not evaluated in this study (7). A large group of tumorous processes in the orbit is formed by tumours imitating formations, according to Krásný 40% of tumorous formations of the orbit were formed by an inflammatory pseudotumour (7). In the case of affliction of the paranasal cavities and orbit simultaneously, within the framework of differential diagnostic consideration we should not overlook the non-tumorous but nevertheless life and sight threatening pathology Wegener granulomatosis (newly referred to as granulomatisis with polyangiitis), in which orbital complications appear in approx. 15% of those afflicted.
Tumours of sinonasal localisation are relatively rare (in the Czech Republic their incidence is stated at 0.5-0.6/100 000 of the population), but their invasion into the orbit is relatively frequent, because for a long time tumours are asymptomatic or manifest symptoms similar to an inflammatory pathology of the nasal and paranasal cavities. Imitation of inflammatory symptoms often leads to erroneous or delayed diagnosis. By contrast with inflammatory pathologies, however, similar symptoms of tumours in the majority of cases are strictly unilateral. As a result patients frequently report for treatment in an advanced stage of the pathology, in which the tumour is invading the surrounding tissues and structures – orbit, brain and meninges or fossa pterygopalatina. Invasion into the orbit is present in 10-15% of patients (2, 6).
In the majority of patients the first manifestations include unilateral nasal obstruction (45%), exophthalmos (36%), dislocation of the bulb and diplopia (22%), pain in the face or head (20%), unilateral epistaxis or secretion from the nose (22%), also deterioration of sense of smell, swelling of the eyelid, intumescence or swelling in the face and malfunction of sensitivity in the innervation region of the 2nd trigeminal branch (2).
The median age is 40 years, the incidence is slightly higher in men. Nicotine addiction and abuse of alcohol are stated in more than 50% of patients (2).
Of malignant tumours of the nose and paranasal cavities we most often encounter spinocellular carcinoma (70%), the most frequent localisation of which is the lateral wall of the nasal cavity and the maxillary sinus. Regional metastases occur in approximately 20% of cases, remote metastases form a squamous carcinoma in 10% of patients (6).
Spinocellular carcinoma invades into the orbit most frequently. In addition to this tumour, other types of paranasal malignant tumours may invade into the orbit – malignant melanoma, nasal glioma, adenocarcinoma, neuroendocrine carcinomas, lymphoma and also tumours invading into the cavities from other parts of the head and face – rhabdomyosarcoma (most frequently from the nasopharynx), skin tumours or carcinomas of the saliva glands (2).
According to the conclusions of Chu Y. et al., invasion into the orbit is slightly more common in carcinomas of the nasal cavity than in carcinomas of the paranasal cavities; here their comparison differs from the majority of previous studies, which may be influenced by the type of study (here the main criterion of the examined cohort is either precisely orbital invasion or it is a secondarily examined symptoms – according to the authors of this article the majority of previous studies focused on nasal and paranasal tumours and not on naso-orbital tumours, as in the case of the study by Chu Y. et al.) (2).

At the time of diagnosis, in 70% of cases or more we encounter an advanced tumour in stage T3 (tumour infringes upon any of the following structures: bone of posterior wall of maxillary sinus, subcutaneous tissue, facial skin, base or medial wall of orbit, fossa infratemporalis, lamellas of sphenoid bone (processus pterygoideus) of cuneiform, sinus ethmoidales) and T4 (tumour infringes upon content of orbit including base and medial wall, including apex and/or any of the following structures: lamina cribrosa, base of skull, nasopharynx, sinus sphenoidalis, sinus frontalis) (2, 6).
In our patient from the first case report, a number of characteristic symptoms of a tumour of the paranasal cavity invading into the orbit were confirmed immediately. The first manifestations, to which the patient nevertheless did not attribute sufficient significance to justify visiting a doctor, were impaired breathing through the left nostril and secretion from the nose. The first manifestation that caused the patient to visit a doctor was double vision in virtually all directions of gaze. During our examination dislocation of the bulb and swelling of the eyelid was determined. These symptoms also correlate with statistically stated frequent ocular manifestations of the pathology. The patient has been a heavy smoker for several years and states abuse of alcohol on a virtually daily basis.
Histologically spinocellular carcinoma of the maxillary sinus in advanced stage pT4pN0 was verified, which ranks among the most common malignant tumours of the paranasal cavities and is the most common tumour which behaves locally invasively and grows into the orbit.
Upon an analysis of diplopia, contrary to expectations the patient stated the peripheral image as the image of the right eye. I explain this perception with reference to the large dislocation and deviation of the bulb of the left eye outwardly. The right eye is not capable of occupying such an extreme position in a leftward gaze in order to reach the same position as the left eye.
Another conspicuous symptom in the patient's eye was anisocoria, which was evident at the first examination. I attribute its origin to pressure of the parasympathetic part of the lower branch of n. III. in the apex of the orbit, which led to an infringement upon the parasympathetic innervation of the pupil.
I included case report no. 2 for a comparison of subjective complaints, symptoms and therapeutic procedure with the first case. The dermatological finding in the place of the scar during the time of our observation before determination of the diagnosis, and for several months afterwards, was without signs of recurrence, but expansion of the spinocellular carcinoma continued across the hypodermis and maxillary sinus into the orbit and further intracranially. The question arises as to whether the pains in the region of the left cheek were actually connected with the extraction of the tooth, or whether they were rather a manifestation of the propagation of the tumour. As Palmar states, chronic idiopathic pain in the face and paresthesis in the innervation region n. infraorbitalis may be easily underestimated symptoms of a tumour of the maxillary sinus (12). Within two months the motility disorder progressed from restriction of abduction to total ophthalmoplegia and ptosis of the upper eyelid. The patient did not observe diplopia due to her practical blindness in the left eye. Due to partial tarsorrhaphy, assessment of exophthalmos was difficult. Although expansion of the tumour to the apex of the orbit is not described on MR of the head at the time of origin of total ophthalmoplegia and ptosis, with regard to the scale of the oculomotor disorders we envisage this.
The treatment of these secondary tumours of the orbit covers surgical treatment, radiotherapy or a combination thereof, and chemotherapy usually as an additive therapeutic method. The surgical options are lower, central or total maxillectomy, extended exenteration of the orbit, craniofacial or coronary resection, resection of the tumour from lateral rhinotomy or a combination of these procedures (2, 6, 8).
Selection of the most suitable therapeutic procedure is always a multi-disciplinary question. Treatment is planned and conducted in the co-operation of an otorhinolaryngologist, orbital surgeon, oncologist, diagnostic radiologist, pathologist and if applicable neurosurgeon and dermatologist. We must naturally always take into account the patient's overall condition of health (13). The various therapeutic procedure in this co-operation was planned also in our cases and was chosen with substantial differences, in order to be of maximum benefit for the patient with regard to age, condition of health and size of the tumour, and in the case of case report no. 2 in order to be as sparing as possible for the patient. For this reason a surgical procedure was abandoned and the patient underwent only palliative radiotherapy.
With reference to the substantial mutilation of the face by extensive surgical procedures, an important factor is also subsequent care leading to an improvement of cosmetic appearance and return to a regular daily regime with a sufficiently high quality of life (1, 2). Preoperatively it is appropriate to plan cosmetic covering of the defects in the face, which contributes to an improvement of the patient's psychological condition and motivation. In addition to reconstructive plastic procedures, silicon prostheses and eye prostheses serve this purpose. At present the development of modern technologies enables the manufacture of precise copies of various parts of the human body, which upon an ordinary view are virtually indistinguishable from the surrounding tissue. As a result, for some patients the choice of an epithesis becomes a more appropriate solution not only from a therapeutic perspective (further surgical procedures are not required, the free trepanation cavity uncovered by a flap of skin enables earlier identification of any applicable recurrence of the tumour), but also from a cosmetic perspective. Epitheses may be fixed to the body either with the use of glue or fixed to the frame of glasses, and the patient places the epithesis on the face together with the glasses. The last option for affixing epitheses is fixation using titanium screws and magnets, which however is not appropriate for patients with malignant tumours, because these materials may limit control CT and MR examination in order to exclude recurrence upon the generation of artefacts (1, 4). In our patient the epithesis is held very well in place upon fixation with glue, in its shape it corresponds to the other half of the face, nevertheless somewhat surprisingly for us the patient is not entirely satisfied because he feels that the colour of the epithesis is not the same as that of the surrounding skin, especially in the winter months, when he

feels it is more suitable to cover the trepanation cavity with a bandage. However, objectively the resulting cosmetic effect with the epithesis is more than satisfactory.

CONCLUSION

Upon examination of patients with diplopia, motility disorder and dislocation of the bulb we should keep in mind the issue of expansively spreading tumours from the paranasal cavities, because precisely these ocular symptoms rank among the first that cause patients to seek medical attention and are usually a sign of a very advanced pathology. Within the framework of differential diagnostic consideration, in addition to knowledge of the topological-anatomical ratios on the orbit and surrounding area, also important is anamnestic data relating to nasal blockage and secretion from the nose, to which patients may not attach sufficient significance to mention themselves, as well as information about pain in the innervation region of the trigeminal nerve.

The therapeutic procedure should always be chosen individually upon multi-disciplinary co-operation. In deciding we must take into consideration the fact that we are faced with a patient with a very serious, potentially life threatening pathology, which we wish to treat radically, but on the other hand the majority of surgical procedures leading to total resection of tumours are highly demanding and substantially mutilate the patient's face. As a result, before the procedure we should familiarise the patient with the possibilities of subsequent cosmetic solution, in which new horizons open up for us with the use of ever more sophisticated and anatomically credible silicon epitheses.

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