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CASE REPORT

Awake surgery for eloquent area glioma in a pregnant patient: a case report with 7-years follow up

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Abstract

Background: Awake glioma surgery in pregnant patients occurs rarely and is a huge challenge for the whole therapeutic team, requiring the cooperation of a neurosurgeon, an anaesthetist, a speech therapist and an obstetrician.

In this paper we present the case of a 31-year-old patient in 22 hbd with low grade glioma (LGG) of the left temporal lobe.

The patient was admitted to the outpatient clinic, having experienced transient speech disorders for about a week. An MRI examination revealed an extensive tumour in the left temporal region. The speech cortical centres were mapped using fMRI and the awake surgery was tailored. The surgery was performed under neuroleptanalgesia with dexmedetomidine and remifentanil and regional anaesthesia. The speech centres were located. The tumour was completely removed, revealing astrocytoma fibrillare WHO II. The patient's speech was continuously monitored, as well as the foetal vital functions. The course of the pregnancy was uneventful. In the 48th month after the first operation, the patient underwent a reoperation due to tumour recurrence with consecutive protonotherapy. Currently, 88 months after the first operation, the patient and child both remain in very good condition.

Conclusions: Few cases of glioma resection with intraoperative awakening in pregnant women have been described in the literature. The awake method seems to be an optimal treatment option.

Keywords: awake craniotomy, pregnancy, low grade glioma

Introduction

The coexistence of a primary brain tumour and pregnancy is rare and was estimated by Haas in 1986 to occur in 3–6/1,000,000 live births [1]. In recent studies, only case series are available and the incidence of primary brain tumours diagnosed during pregnancy is unknown [2–8]. In addition, there are no established treatment paths since they vary for every case, depending

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on the size of the tumour, potential histology, neurological symptoms and stage of pregnancy. Different options, including termination of pregnancy, premature caesarean section, close monitoring, surgical resection and/or radiotherapy, during or after pregnancy, are tailored to each patient [3,8]. Treatment of eloquent area tumours is even more challenging and requires e.g. intraoperative awakening and therefore the close cooperation between a neurosurgeon, an anaesthetist, a speech therapist and a gynaecologist. Currently, only a few cases of glioma resection in pregnant women, performed in awake surgery, have been described in the literature [9–14].

In this paper we present the case of a 31-year-old patient in 22nd week of gestation (hbd) with low grade glioma (LGG) of the left temporal lobe.

Case description

A 31-year-old pregnant woman in 22 hbd who had had transient motor speech disorders for about a week was admitted to an outpatient clinic. A brain MRI revealed an extensive tumour in the left temporal region with mass effect (Figure 1). The tumour was hypointense in the T1-weighted sequence and hyperintense in the T2-weighted and fluid-attenuated inversion recovery (FLAIR) sequences, with medium contrast enhancement and no diffusion restriction.

Due to the suspicion (suggested by the radiology consultant) of a possible high grade glioma and mass effect, surgery was recommended. The location of the speech cortical centres was mapped using fMRI and the awake surgery was tailored. The aim of the procedure was complete resection of the tumour, sparing speech function, while maintaining the safety of the pregnancy. The patient was fully informed about what to expect during the procedure.

The surgery was performed under neuroleptanalgesia with dexmedetomidine and remifentanil and regional anesthesia: ropivacaine and lidocaine. Sedation was controlled using the bispectral index (BIS) and EEG monitoring. The patient was in a semilateral position to prevent aortocaval compression. She was conscious throughout the entire surgical procedure with short periods of minor sedation for comfort during e.g. pinning, catheterisation or drilling. Magnesium was administered promptly before surgery. Sensorimotor monitoring was conducted. During the procedure the speech centres were located, safety margins were marked and the tumour was completely removed using a Cavitational Ultrasonic Surgical Aspirator (CUSA) in a piecemeal method.

In histopathology, a diagnosis of astrocytoma fibrillare (WHO gradeII) was made (WHO 2016 classification). The speech was continuously

monitored by the speech therapist. The vital functions of the foetus were checked by the gynaecologist. No abnormalities were detected in the postoperative foetal ultrasound. The patient was discharged on the fifth postoperative day in very good condition, with minimal reversable speech disturbances while speaking fast.

The further course of the pregnancy was uneventful and terminated on time (37 Hbd) by caesarean section due to the glioma diagnosis. The newborn was healthy. In the patient, all the speech disturbances had been resolved by the time of the birth. The patient was observed in the outpatient clinic and underwent a series of annual brain MRI scans.

Forty-six months after the first operation, the patient started to complain of stuttering when speaking and reported difficulties with word recall. An MRI scan revealed the recurrence of an exophytic tumour and post-contrast enhancement in one of the walls of the postsurgical cavity. Two months later (48th month) she underwent a reoperation, under general anaesthesia. Total excision was performed. There was no change in tumour histology in comparison to the first resection (astrocytoma fibrillare WHO II). Nevertheless, the oncologist referred the patient for protonotherapy, the course of which was uneventful.

In consecutive MRIs no recurrence of the tumour was found. Currently, 88 months after the first operation, the patient remains in very good condition. She complains of difficulties with finding words while speaking fast or under pressure. In everyday speech there is no noticeable deficit. Her 7-year-old child is developing well, with no neurological, health or developmental issues noted since birth. In the patient's opinion, no difference between her child and peers may be found.

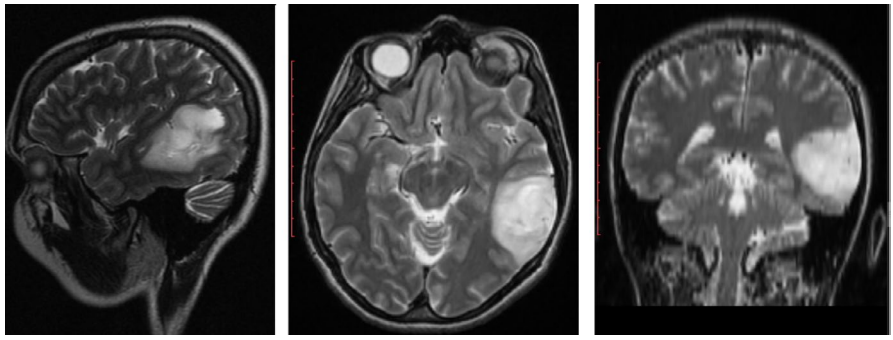

Figure 1. Preoperative MRI with left temporal lobe glioma T2 images

Discussion

Gliomas in eloquent areas of the brain pose a significant challenge due to the risk of postoperative deficits. Pregnancy further complicates management, considering the potential risk to the mother and foetus. The impact of pregnancy on glioma prognosis is inconclusive. Some authors consider the direct association between pregnancy, overall survival and tumour recurrence to be doubtful [4,7,8], whereas others point to the adverse effects of pregnancy on neoplasm [2,3,6,15]. Determining the prognostic impact of pregnancy, especially in LGG, is challenging. Pregnant patients with glioma can be divided into two groups: those newly diagnosed while pregnant and those whose tumour is diagnosed before conception [3,7]. Management, history and prognosis are different in these groups and depend on the exact histopathology, the occurrence of seizures, tumour volume, oncological treatment, recurrence, clinical deterioration and the molecular profile of the tumour [3,7,8]. Treatment varies significantly and must be tailored to each patient individually. In the first trimester or the early part of the second one termination is a possible option [3,5,8]. When maintaining pregnancy, management becomes particularly difficult due to the adverse effects of pregnancy itself (caused by hormonal and haemodynamic changes and increased levels of growth and angiogenic factors) and the wellbeing of the foetus [3,5,8,16]. In stable patients with favourable histology, expectant management until the end of the pregnancy is possible, otherwise appropriate surgical and/or radiotherapeutic and/or chemotherapeutic treatment is implemented [6,8,17]. For resection, the second trimester seems to be optimal [16,18]. Careful MRI monitoring should be performed, which impacts treatment options [3,5]. Rigorous obstetric supervision throughout the whole pregnancy is advisable [2,5].

Additional complications occur with eloquent area tumour location, where progression or resection may cause major deficits. Awake craniotomy allows real-time mapping and the preservation of critical brain functions, which is crucial in eloquent area glioma [19]. It encourages a maximal safe resection, which is crucial in terms of overall survival in glioma patients [20,21]. So far, only the safety of surgery during pregnancy under general anesthesia has been investigated. Recently, there have been single reports of successfully performed awake glioma resections in pregnant patients [9–11, 13,14,17,19,22,23] and one systematic review by Mofatteh et al. [12]. Maintaining optimal neurological health in the pregnant patient is crucial due to the future need to fulfill parental responsibilities [12]. Awake craniotomy protocol reduces exposure to the anaesthetic medication usually used in general anaesthesia [12], which could cause some harm [5,8,11,16,17,19,23]. In

awake procedures intravenous sedatives are used in addition to local anaesthesia, which plays the main role, thus diminishing the usage of intravenous drugs [12]. The potential adverse effects of sedative drugs on the foetus occur rarely and beneficial effects, when applied at appropriate concentrations, have been reported [5,8,11,16,17,22]. In the review by Mofatteh et al., propofol was used in five out of nine studies; dexmetomidine in five out of nine, remifentanil or fentanyl in eight out of nine, lignocaine plus bupivacaine in three out of nine and lignocaine plus ropivacaine in three out of nine; in one study sevoflurane was administered [23] with no adverse effects to the foetus or mother reported, despite the drug regimen. However, the foetus should be thoroughly monitored pre-, intra- and postoperatively [10–12,24].

Awake craniotomy in pregnant women requires the multidisciplinary cooperation of a neurosurgeon, an obstetrician, a speech therapist and an anaesthetist. Proper preparation for complications such as intraoperative seizures, agitation, the need for urgent intubation, complications with foetus, and even emergent caesarean section, seem to be crucial and alternative plans/scenarios should be elaborated [11–13,25]. The patient's resilience, compliance to procedures, proper preparation and cooperation with the whole surgical team are essential.

Due to the low incidence of gliomas in pregnancy, there are insufficient guidelines for treatment using various methods tailored to individual needs. Current reports of awake surgery in pregnant patients are rare and concern various histopathological cases [12]. Moreover, in recently published reports there is a lack of long-term follow up of patients and their children to investigate the consequences of glioma surgery during pregnancy using awake craniotomy. In our case, a 7-year follow up provides us with a thorough insight into the long-term effects on both mother and child, giving hope for high efficacy and the long-term safety of awake craniotomy during pregnancy for glioma surgery in eloquent areas.

Conclusions

Glioma in pregnancy remains a rare condition in neurosurgical practice. Nevertheless, due to the double risk for both the mother and the foetus, it is an urgent matter that needs to be resolved. Different aspects of treatment have been discussed in the literature including the necessity to perform awake resections. Unfortunately, due to the rarity of cases, there is still a lack of evidence to inform the guidelines. Nevertheless, the awake-awake-awake method seems to be a viable option for eloquent area glioma resections in pregnant patients, offering favourable long-term

outcomes for the woman and her child. This case underscores the importance of planning individualised treatment and comprehensive follow-up in managing such complex cases. Precise guidelines on pre-, intra- and postoperative treatment should be developed.

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