Intraoperative neurophysiological monitoring in paediatric Chiari surgery—help or hindrance?

Fahid T. Rasul1 & Samir A. Matloob1 & Nikolaos Haliasos2& Ivana Jankovic3 & Stewart Boyd3 & Dominic N. P. Thompson4

1 Department of Paediatric Neurosurgery, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK
2 Department of Neurosurgery, Queens Hospital, Romford, UK
3 Department of Neurophysiology, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK
4 Consultant Paediatric Neurosurgeon, Great Ormond Street Hospital for Children NHS Foundation Trust, London WC1N 3JH, UK

Abstract

Introduction. The role of intraoperative neurophysiological monitoring (IONM) during surgery for Chiari I malformation has not been fully elucidated. Questions remain regarding its utility as an adjunct to foramen magnum decompression surgery, specifically, does IONM improve the safety profile of foramen magnum decompression surgery and can IONM parameters help in intraoperative surgical decision-making. This study aimed to describe a single institution experience of IOM during paediatric Chiari I surgery.

Methods. The methodology comprised a retrospective review of prospectively collected electronic neurosurgical departmental operative database. Inclusion criteria were children under 16 years of age who had undergone foramen magnum decompression for Chiari I malformation with IONM. In addition to basic demographic data, details pertaining to presenting features and postoperative outcomes were obtained. These included primary symptoms of Chiari I malformation and indications for surgery. MRI findings, including the presence of syringomyelia on pre-and post-operative imaging, were reviewed. Details of the surgical technique for each patient were recorded. Only patients with either serial brainstem auditory evoked potential (BAEP) and/or upper limb somatosensory evoked potential (SSEP) recordings were included. Two time points were used for the purposes of analysing IONM data; initial baseline before skin incision and final at the time of skin closure.

Results. Thirty-seven children underwent foramen magnum decompression (FMD) with IONM. Mean age was 10.5 years (range 1–16 years) with a male:female ratio 13:24. The commonest clinical features on presentation included headaches (15) and scoliosis (13). Twenty-four patients had evidence of associated syringomyelia (24/37 = 64.9%). A reduction in the SSEP latency was observed in all patients. SSEP amplitude was more variable, with a decrease seen in 18 patients and an increase observed in 12 patients. BAEP recordings decreased in 13 patients and increased in 4 patients. There were no adverse neurological events following surgery; the primary symptom was resolved or improved in all patients at 3-month follow-up. Resolution or improvement in syringomyelia was observed in 19/24 cases.

Conclusions. Our data shows that FMD for Chiari malformation (CM) is associated with changes in SSEPs and BAEPs. However, we did not identify a definite link between clinical outcomes and IONM, nor did syrinx outcome correlate with IONM. There may be a role for IONM in CM surgery but more robust data with better-defined parameters are required to further understand the impact of IONM in CM surgery.

Keywords Chiari . Neurophysiology . Intraoperative monitoring . Foramenmagnumdecompression

Introduction

Intraoperative neurophysiological monitoring (IONM) is now considered a standard of care for many spinal procedures. Its role in Chiari surgery has received scant attention, and current data appears to be contradictory. There are two possible reasons why IONM might be of benefit during foramen magnum decompression surgery. Firstly is to improve the safety of surgery, by reducing the risk of adverse neurological outcomes. This benefit has already been demonstrated in the context of surgery for intramedullary spinal cord tumours and spinal deformity surgery. Secondly, it might be possible to use IONM parameters to guide the extent of surgical intervention.

Whilst foramen magnum decompression (FMD) is the commonest operation performed for Chiari malformation (CM) a plethora of variations of surgical technique are currently used. Of particular current interest is whether or not dural opening should be routinely performed? Surgery can be extradural (involving bony decompression only (including/excluding C1 laminectomy)) or intradural with further modifications (expansion duraplasty, arachnoid dissection). FMD aims to relieve the pressure on neural structures at the craniovertebral junction whilst simultaneously re-establishing normal CSF flow dynamics across the foramen magnum. A large survey of paediatric neurosurgeons showed a preference for posterior fossa decompression with duraplasty [1, 2]. Evidence suggests that this technique improves 75–90% of symptomatic CM paediatric patients [3]. A recent systematic review suggested that duraplasty was associated with better clinical improvement, longer hospital stay and increased incidence of post-operative complications. The authors concluded, however, that the overall quality of evidence was low and that further prospective trials were required to answer this question more robustly [4]. There is recognised morbidity associated with dural opening, and some authors have concluded that duraplasty is linked to an increased incidence of CSF complications [5]. There are now increasing reports suggesting that CM symptoms can be improved (including improved syringomyelia) by limiting the operation to a bony decompression only [6]. To date, there has been no randomised control trial investigating the different types of FMD for Chiari malformation, and thus there is no uniform surgical approach to this pathology. The choice of technique largely reflects surgeon preference rather than evidence-based practice. Some surgeons have attempted to tailor their technique according to various criteria, for example reserving dural opening for cases where there is associated syringomyelia or using intraoperative ultrasound to look for improved tonsillar movement or improved CSF flow after bony decompression [7]. Whether IONM parameters could be used to inform the extent of surgery that is required during Chiari surgery is a novel concept that has been less well addressed.

A limited number of adult and paediatric studies have investigated the role of IONM in Chiari surgery with variable results [8–12] (Table 1). Barzilai et al. reported stable somatosensory evoked potentials (SSEPs) and motor evoked potentials (MEP) during surgery in 21 pediatric patients. Though 3 patients experienced some attenuation during positioning, this did not lead to any clinical effects. Zadel et al. have reported the largest series to date and found that most brainstem auditory evoked potential (BAEP) changes occurred following bony decompression. This may be an important consideration to guide the extent of surgery. Anderson et al. reported their results from a small prospective cohort with results in keeping with those of Zadel et al. noting that BAEP changes occurred mainly following bony decompression, and like Barzilai et al., they found that some changes occurred during positioning particularly neck flexion. On the other hand, Roser et al. reported no significant changes in either SSEP or MEP during Chiari surgery even though 92.8% of their group showed clinical improvement or stability. From the published data, there is thus no proven role for IONM in Chiari surgery; however, with technological improvements and greater experience of IONM, this may change in the future.

Methods

The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement was used in the preparation of this section of the manuscript [13]. Informed consent was not sought, as this was a retrospective study.

Setting and participants

The study was conducted at Great Ormond Street Hospital for Children, London, UK (GOSH).

Patients offered foramen magnum decompression surgery are those with MR evidence of CM ± syrinx with clinical symptoms or signs considered by the senior author to be preferable to the CM, in whom no additional cause for Chiari I malformation can be identified in patients with CMand syringomyelia scoliosis was considered a sign even if no additional symptoms were present. All cases are recorded on a prospectively maintained database, and this database was searched over a 7-year period between 1 January 2012 and 31 December 2018 to identify all children aged 17 years or less who underwent FMD with IONM for CM by a single surgeon.

Variables and data sources: A retrospective case note review was performed for each child to extract data on their presenting symptoms, radiological investigations, clinical outcome and intraoperative SSEP and BAEP readings at specific time points during surgery.

The following information was recorded: demographic data (age, gender), clinical signs and symptoms on presentation, radiological evidence of CM ± syrinx, operative technique (bony decompression only or dural opening and duraplasty).

Outcome measures included post-operative symptoms and radiological evidence of syrinx change on whole-spine MRI 3 months after surgery.

Surgical technique

All patients were placed in the prone position with slight head flexion (Fig. 1). A midline incision and exposure of sub-occipital bone and C1 lamina were performed (Fig. 2). Sub-occipital craniectomy followed by C1 laminectomy was performed in all patients. In patients who underwent dural opening, the dura was opened in a T shape, with inferior lateral releasing incisions to maximise decompression of the cerebellar tonsils (Fig. 3). A synthetic expansion dural patch was applied (Duraguard ® or suturable Duragen ®) for watertight dural closure. Ultrasound (USS) was used to confirm that the inferior limit of the tonsils was visible after bone removal. USS findings were not routinely used to determine the extent of the surgical procedure. Bone decompression only with dural splitting was typically reserved for younger patients (less than 5 years) or in children with symptoms of headache only and no associated syrinx.

Intraoperative neurophysiological monitoring

Intraoperative monitoring was carried out with serial BAEP and upper limb SSEP recordings. Two time points were analysed for the purposes of this study, firstly at skin incision (after positioning) and secondly at skin closure. Since body temperature can affect IONM recordings, we documented core temperature at the same two time points. The stimulation/monitoring system was XLTEK Protektor™. Sub-dermal needle electrodes were placed at scalp positions A1 and A2 with a sub-dermal corkscrew electrode (‘Neuroline’, Ambu) at the vertex as reference for BAEP and at C3’, C4’ and Erb’s point all referred to a mid-frontal sub-dermal corkscrew for SSEP. Electrode impedances were < 2 kΩ. See Table 1 for settings for evoked potentials. For BAEP, the peak latencies of Wave V, and the I-V interpeak latencies were compared; the peak latencies of ‘N20’ were used for SSEP. Table 2 summarises the settings for evoked potentials.

Results

Thirty-seven children satisfied the inclusion criteria for this study. The mean age was 10.5 years (range 1–16 years) and the male:female ratio was 13:24.

Presentation

Clinical Patients presented with a variety of signs and symptoms including headaches, scoliosis, sensory/motor disturbances in the upper limbs, bulbar dysfunction, gait disturbance, sleep apnoea and speech delay. Often patients presented with multiple symptoms. A summary of clinical features on presentation is included in Fig. 4.

Radiological Twenty-four patients had CM with MRI evidence of a syrinx (24/37 = 64.9%); 13 patients had CM with no evidence of syrinx (13/37 = 35.1%).

Treatment

Surgery Twenty-nine patients underwent FMD + expansion duraplasty; 8 patients underwent FMD and dural splitting.

IONM results

SSEP data was available for 33/37 patients (89.2%). Twelve patients had an increase in their SSEP amplitude at the end of surgery compared with pre-incision readings (12/33 = 36.4%). Eighteen patients had a decrease in their SSEP amplitude (18/33 = 54.5%). Three patients had no change in their SSEP amplitude (3/33 = 9.1%). Thirty-one patients had a reduction in their SSEP latency by the end of surgery (31/33 = 93.9%), and 2 patients had no change in SSEP latency (2/33 = 6.1%). No patient had an increase in SSEP latency at the end of surgery. We obtained BAEP data on 19 patients (19/37 = 51.4%). Thirteen patients had a reduction in their BAEP at the end of surgery compared to pre-incision readings (13/19 = 68.4%). Four patients had an increase in their BAEP by the end of surgery (4/19 = 21.1%). Two patients had no change in BAEP by the end of surgery. We obtained core body temperatures pre-incision and at skin closure on 29 patients (29/37 = 78.4%). Twenty-seven patients had a higher core body temperature at the end of surgery (27/29 = 93.1%). Of those 27 patients, the observed temperature increase was > 0.5 °C in 21 patients. Figures 5 and 6 show the effects of surgery on SSEP latency and SSEP amplitude respectively, and Fig. 7 shows the effect of surgery on BAEP.

Outcomes

Clinical: All patients who presented with symptoms prior to undergoing FMD reported symptomatic improvement or resolution at 3 months post-operatively. There were no cases of worsened neurological symptoms. In all 13 patients who presented with evidence of scoliosis, there was an improvement or no further progression of their scoliotic deformity at the latest clinical and radiological follow-up.

Radiological: Twenty-four patients had radiological evidence of a syrinx in association with CM. Complete resolution of syrinx was observed in 7 patients (7/24 = 29.2%); improvement in the size of syrinx was observed in 12 patients (12/24 = 50%). In the other 5 patients, the syrinx appeared stable post-operatively.

Correlating radiological markers of improvement with IONM

On the further assessment of radiological markers of improvement, we analysed our sub-group of 19 patients in whom the syrinx improved or resolved. In this group, we obtained SSEP data in 17 patients. Of these 17 patients, 12 showed a reduction in SSEP amplitude at the end of surgery compared with pre-incision values (12/17 = 70.6%). In 16 of these patients, the SSEP latency decreased (16/17 = 94.1%) and in patients whom it did not decrease, the latency remained static. We obtained BAEP data in 14 of these 19 patients. In those patients, we observed a reduction in the BAEP latency in 6 patients (6/14 = 42.9%), an increase in BAEP latency in 3 patients (3/14 = 21.4%) and no change in the remaining 5 patients (5/14 = 35.7%).

Discussion

The principal findings of our study are as follows:
(1) FMD was effective at improving patient symptoms. No patients with an associated syrinx experienced radiological progression of the syrinx following surgery.
(2) SSEP latency decreased in 93.9% of patients.
(3) More than 50% of patients had a decrease in their SSEP amplitude.
(4) BAEP readings decreased in 68.4% of patients.
Our results suggest that FMD surgery has an effect on SSEPs and BAEPs but the exact nature of this effect is not well defined. On further analysis of our data, we attempted to correlate clinical outcomes with neurophysiological data. Since there were no cases of worsened neurological symptoms, it was difficult to correlate symptomatic changes with changes in SSEP amplitude or BAEP latency. There was a high percentage of patients in whom a reduction in the SSEP latency was observed (31/33 patients = 93.9%). It might be reasonably supposed that this is a consequence of releasing pressure on the sensory nuclei and pathways at the level of the foramen magnum. Reduction of pressure inside the syrinx cavity following FMD might be an additional mechanism for the improvement in SSEP latency in those cases with CM and syringomyelia; however, it is perhaps surprising that such effect would occur so immediately the following decompression. There are studies that have demonstrated very early resolution of syrinx following surgery and so this remains a plausible explanation. Change in latencies was not strikingly different between the two groups. The median values of the shortening in SSEP latency are more than double for the unimproved group (8.3 vs 3.1), with a notable outlier. The median difference in BAEP latency between the two groups was in the same direction (6.4 vs 5.65), but this is not significant. Five patients who underwent surgery showed stable appearances with no progression of their syrinx post-operatively.

Previous studies that have investigated the role of IONM in CM surgery have not shown similar results. Roser et al. investigated 39 patients undergoing FMD for CM with IONM[12]. They reported no significant changes in the SSEP latency before and after surgery. This is in contrast to our observations that SSEP latency decreased in a high percentage of patients (93.9%). Zamel et al. analysed BAEPs in 80 patients during CM surgery [9]. They reported that the majority of changes occur following bony decompression. We did not observe consistent improvements in BAEPs in our population; however, we did not perform neurophysiological readings at the same time points during surgery as were used in their study. In our study, we looked only at the pre-incision readings and the readings immediately after skin closure. Therefore, we cannot comment directly on the effect of bony decompression. Some studies have commented on the effect of positioning on IONM. Anderson et al. studied 11 patients prospectively [10]. One of the key messages in their study was that neck flexion during positioning puts neurologic structures at risk according to their BAEP and SSEP data. This has also been suggested by Barzilai et al. who observed attenuation of SSEP and MEP during positioning though this did not lead to any clinical effect [8]. In our study, we did not perform any IONM during positioning so it is not possible for us to comment on this aspect of FMD surgery.

Limitations

There were several limitations in our study. There are no defined thresholds for SSEP and BAEP values that denote an effect in CM surgery. We are not aware of guidelines on how to interpret the results of IONM in CM surgery. Also, there is difficulty in obtaining sufficient evidence using identical techniques [14]. Is there a certain percentage change that should occur before we alter our surgical strategy? In spinal surgery, there are better defined neurophysiological values that can guide surgery. For example, if SSEP amplitude drops by 50% or latency increases > 10%, this is considered significant [15] and a surgeon should take heed of this real-time information to inform their surgical strategy. No such values exist

for IONM in CM surgery. How do we know, therefore, if/when to stop or to change surgical strategy? IONM is used as a guide for surgeons to improve surgical technique/strategy and this ultimately is aimed at reducing morbidity associated with surgery. In an era fraught with medicolegal considerations, IONM results may be cited as evidence in litigation cases. It is therefore important that the potential implications of IONM changes during Chiari surgery become better defined. The current paucity of data regarding IONM in CM surgery means that there are no specific SSEP and BAEP values or percentage changes that have been shown to have a definitive association with altered clinical outcome. This may not mean that neurophysiology is irrelevant but, rather, that our understanding and interpretation currently is limited due to lack of high quality data.

Our data includes only two time points in surgery. We looked at the SSEPs before skin incision and again after skin closure. This provides a relatively crude marker of the potential neurophysiological changes associated with key steps of surgery during FMD. If sensory and motor latencies and amplitudes change during surgery, it is important to know when this occurs and why. This is the real value of IONM in any spinal surgery. Ideally, real-time feedback would be provided so that any changes can be acted upon at the time–for example optimising spinal cord perfusion in response to a reduction in motor amplitudes. According to previous data, positioning is an important aspect to consider for IONM and the effect of bony decompression on IONM values may guide surgical approaches. We did not perform neurophysiological monitoring at either of these time points so our data may be of more limited value in this respect. Intraoperative decision-making about when to perform duraplasty and when to limit surgery to bony decompression was not objectively analysed, and this may have had some impact on the effect and understanding of the role of IONM in CM surgery. IONM is a time-consuming task that prolongs surgery and is associated with a cost impact. Can we justify its use in CM surgery if there is no discernible effect? To date, no study has proven its efficacy in this scenario though there is a real paucity of literature in this field. Is IONM in CM surgery a hindrance or a potential help is a question that merits further investigation?

Conclusions

Our data suggests that FMD for CM is associated with changes in SSEPs and BAEPs. However, we have not been able to identify a definite link between clinical outcomes and neurophysiological monitoring. There may be a role for IONM in CM surgery but more robust data with better defined parameters are required to further understand the impact of IONMin CM surgery.

Compliance with ethical standards

Conflict of interest.

The authors declare that they have no conflict of interest.

Ethical Approval.

The study was registered as a service evaluation with the Great Ormond Street Hospital for Children NHS Foundation Trust.

Informed consent.

Informed consent was not sought as this was a retrospective study.

Table1.

Table 1 Key studies assessing the role of IONM in FMD for CM

Citation	Year of publication	Study design	Number of subjects	Adult or paediatric study population	Key results	IONM helpful?
Barzilai et al.	2016	Retrospective cohort	21	Paediatric	19 cases = Stable SSEP and MEP. 3 cases = attenuation during positioning – nil clinical effect	Yes
Zamel et al.	2009	Retrospective cohort	80	Paediatric and adult	BAEP improvements occur mainly following bony decompression; less so following duraplasty	Yes
Anderson et al.	2003	Prospective observational	11	Paediatric and adult	BAEPs improved in all patients mainly following bony decompression, clinical improvement in all patients. Risk of altered SSEP and BAEP during positioning and neck flexion mainly	Yes
Chen et al.	2012	Consecutive patients over 1 year	13	Paediatric	SSEP improved in all patients, clinical improvement in all patients	Yes
Roser et al.	2016	Retrospective cohort	39	Adults	No significant change in SSEP or MEP between baseline and final measurements. 92.8% patients remained clinically stable or showed clinical improvement	No

Table 2. Settings for recording Evoked Potentials.

Parameters	BAEP	SSEP
Stimulus	0.1ms auditory clicks, 70-80dB HL	0.2-0.5ms 20-30mA currents
Stimulation rate	10.1 – 11.1 Hz	3.7 Hz
Stimulus site	R & L ears alternately	R & L median/ulnar nn. alternately
Number of stimuli averaged	1000	200
Epoch	10ms	50ms
Bandpass	100Hz – 3KHz	20Hz – 3KHz

HL = hearing level;

Conclusions

Our data suggests that FMD for CM is associated with changes in SSEPs and BAEPs. However, we have not been able to identify a definite link between clinical outcomes and neurophysiological monitoring. There may be a role for IONM in CM surgery but more robust data with better defined parameters are required to further understand the impact of IONMin CM surgery.

Compliance with ethical standards

Conflict of interest.

The authors declare that they have no conflict of interest.

Ethical Approval.

The study was registered as a service evaluation with the Great Ormond Street Hospital for Children NHS Foundation Trust.

Informed consent.

Informed consent was not sought as this was a retrospective study.

Figures

Figure 1. Patient positioning – prone.

Figure 2. Exposure of sub-occipital bone and C1 lamina

Figure 3. Dural opening and decompression of tonsils

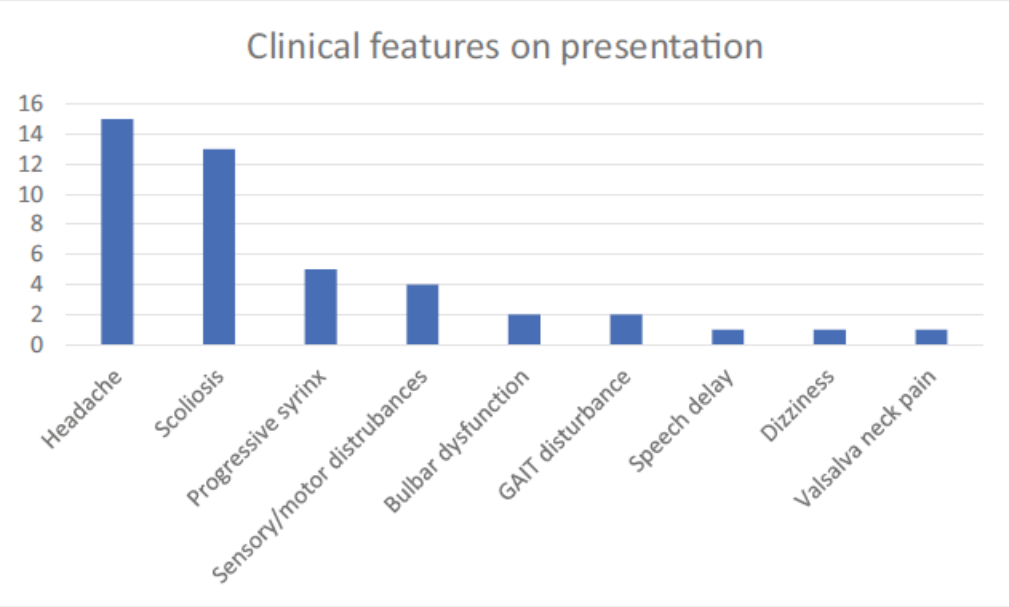

Figure 4. Clinical features on presentation.

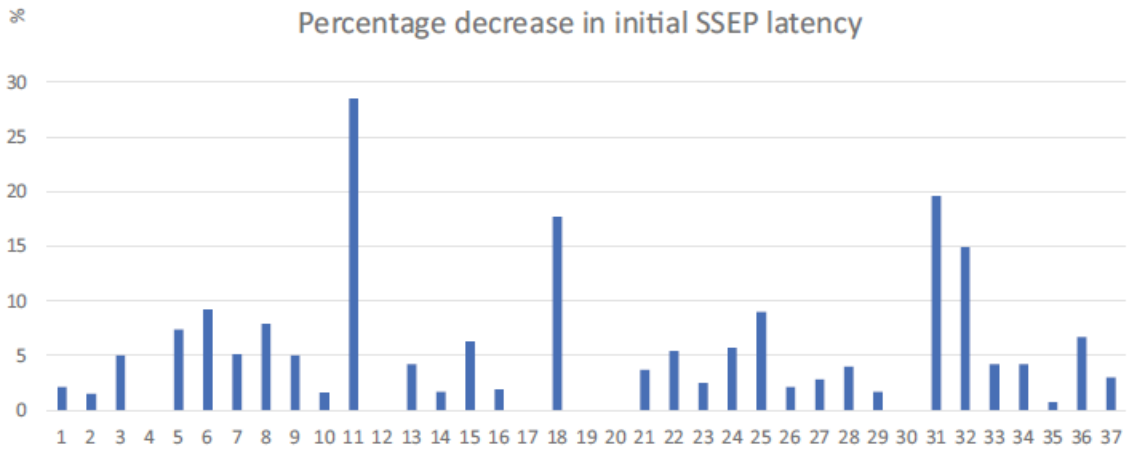

Figure 5. Percentage decrease in initial SSEP latency.

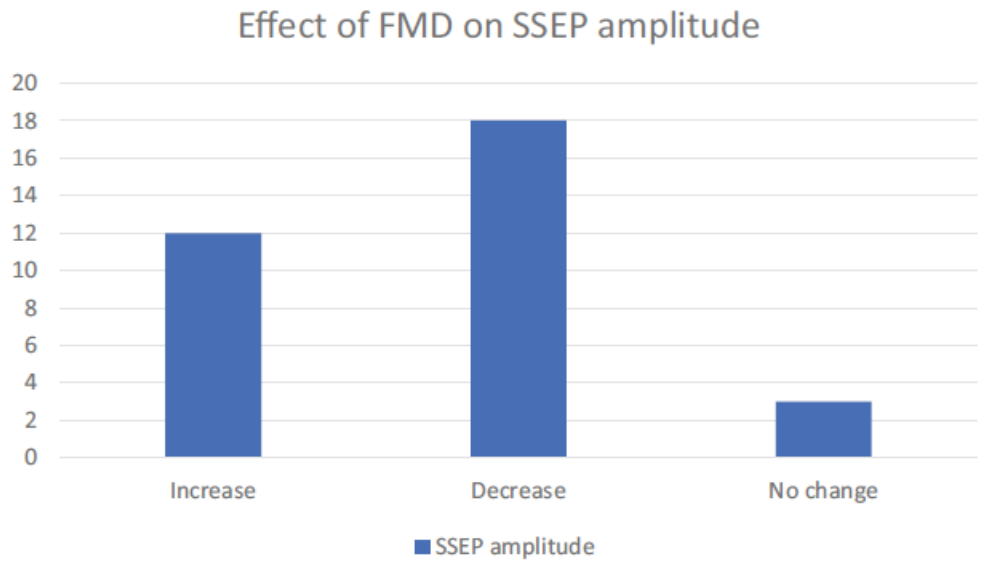

Figure 6. Effect of FMD on SSEP amplitude

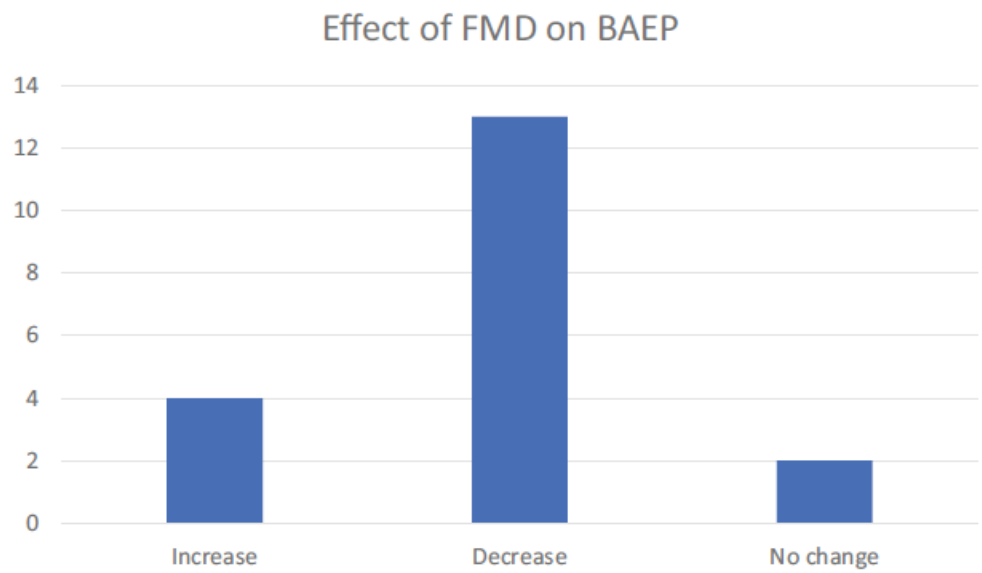

Figure 7. Effect of FMD on BAEP I-V interpeak latency.