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ORIGINAL ARTICLE

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Abstract

Objective: To find physiologically arousing stimuli and labile physiological channels in a non-verbal adolescent with severe and multiple congenital disabilities, who did not have a reliable means of communication.
Methods: The client was repeatedly presented with visual and audiovisual stimuli, representing variations of six contextual factors over three sessions in a one month period. For each stimulus, reactions were detected in the client's four peripheral autonomic nervous system signals using a rule-based classification algorithm.
Results: During the presentation of audiovisual stimuli, the number of physiological reactions significantly differed from that observed in baseline ($\chi^2 = 3.93$, $p = 0.0476$). Aural stimuli articulated in an unfamiliar voice, and aural stimuli containing anticipatory patterns were also physiologically arousing. Fingertip temperature was the client's most labile physiological signal.
Conclusions: The results of this case study suggest that physiological data may complement caregiver acumen in deciphering the reactions of non-verbal clients with severe and multiple disabilities.

Introduction

The communication challenge

People with severe and multiple disabilities are often aware of events and stimuli in their immediate environment, but as a result of their limited physical abilities, are unable to interact with their surroundings through consistent and meaningful expressions or movements [1]. These individuals may therefore appear completely passive; they do not have the ability to deliberately and voluntarily convey their intentions, but rather rely completely on others (e.g. parent, nurse, or caregiver) to discern and interpret their needs. The inability to physically express personal intentions may be secondary to neurological conditions such as cerebral palsy, degenerative neuromuscular disorders such as muscular dystrophies and amyotrophic lateral sclerosis, autoimmune diseases such as multiple sclerosis, and strokes, especially in the brain stem [2]. In order to provide this population with some degree of independence, design, and development of access pathways has become an important topic in rehabilitation research.

An access pathway is the actual sensors or input devices by which an expression of functional intent (e.g. a movement or physiological change) is transduced into an electrical signal [3]. In other words, it is the conduit which connects an assistive technology, such as augmentative and alternative communication, to its user. Unfortunately, most of the commercially available alternative access pathways (e.g. mechanical switches, eye gaze systems, and inertial sensors) require at least one repeatable movement [3], and hence for many individuals, cannot help to resolve this communication challenge.

An alternative communication solution

While many people with severe and multiple disabilities often do not have the ability to voluntarily control their skeletal muscular system, their sympathetic sudomotor function may remain intact [4]. The individual's sympathetic activity is manifested in the central and peripheral signals of the autonomic nervous system (ANS). Signals of the peripheral ANS are modulated by affective and startling stimuli [5–8], and have been used to monitor human emotion [9–12]. An emerging physiological signal-based access pathway for people with severe disabilities is one that works based on voluntary control of peripheral ANS signals. It has been shown that by performing mental activities such as music imagery or mental arithmetic, one can voluntarily generate reactions in his/her peripheral ANS signals [13, 14].

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Motivation and objectives

This paper represents a foundational step towards the development of a communication solution for a non-verbal adolescent with severe and multiple congenital disabilities [15]. The first step in developing a physiological access pathway is the identification of stimuli that elicit meaningful and reproducible changes in an individual's physiological signals. Biofeedback monitoring may provide an objective way of detecting change-inducing stimuli [9, 11]. Thus, the primary objective of this study was to identify stimuli that cause significant reaction in the client's physiological signals. This purpose is analogous to stimulus preference screening in case studies with microswitches [16–18]. Lancioni et al. [19] defined preferred stimuli as those that that seemed to produce positive reactions (e.g. alert or smiling) by the client. We emphasize that in the context of the present case study, preferred (arousing) stimuli are stimuli that elicit meaningful and reproducible change in the client's physiological signals.

The secondary objective of this study was to identify the client's most labile physiological channels (i.e. physiological signals that contain the greatest number of reactions to arousing stimuli). Addressing these objectives will help to answer the question about whether or not physiological signals can be used to determine suitable stimuli for a potential physiological access solution for an individual with severe disability.

Materials and methods

Peripheral ANS signals

We used four peripheral ANS signals (electrodermal activity (EDA), fingertip temperature, heart rate, and respiration) as a means of identifying arousing stimuli. EDA is a measure of the sympathetic cholinergic function that induces changes in the skin's resistance to electrical conduction. Changes in EDA can be spontaneously or reflexively evoked by a variety of internal or external arousal stimuli [20]. These stimuli cause an increase in EDA of more than $0.05\,\mu S$ within a 10-s interval – a characteristic change known as an electrodermal reaction (EDR) [21]. These reactions occur with a latency of 1.3–$1.65\,s$ subsequent to stimulus presentation for persons without physical disabilities and between 1.5 and $2\,s$ for subjects with disability such as cerebral palsy [4, 22]. Recently, these electrodermal signals have demonstrated potential as an access pathway [13].

Fingertip temperature fluctuations are a combination of long-term changes due to homeostatic thermoregulation, and transient changes in cutaneous microcirculation [23]. These transient changes are often a result of the contraction and dilation of arteriovenous anastomoses (AVAs), which are densely innervated by sympathetic nerve fibres, and hence reflect the state of an individual's ANS [23]. The direction of these changes is dependent on the overall body temperature – the same stimuli can elicit both vasoconstriction and vasodilation [24].

The effects of startling or arousing stimuli on human cardiac signals have been often reported in the literature. For example, Lacey and Lacey [25] showed that infrequently occurring aural tones embedded in a sequence of standard auditory stimuli generated bradycardia, or slowed heart rate. In a separate study, an acute loud noise was found to induce increases in blood pressure and heart rate, likely due to sympathetic activation [5].

When in a relaxed state, an individual's respiration rate and amplitude maintain a steady-state pattern; these features change when the individual experiences stress [26]. Generally, respiratory responses tend to vary with the levels of valence and arousal of affective stimuli [27]. Furthermore, Jonghwa and Andre [28] found that features in both electrocardiographic and respiratory signals were useful for emotional differentiation.

Case description

The client, whom we will refer to as Sean, was a 15-year-old male with spastic quadriplegic cerebral palsy, visual impairment, and global developmental delay, secondary to birth asphyxia. He had elevated or fluctuating tone throughout his extremities, and reduced or fluctuating tone in his neck and trunk. Sean's personalized equipment included bilateral wrist splints and ankle–foot orthoses and a Tilt-in-Space manual wheelchair. Sean lived at home with his family and attended a special education school. He had a history of infantile seizures, ongoing respiratory and swallowing difficulties, gastroesophageal reflux secondary to hiatus hernia and oesophageal stricture, and recurrent bronchiolitis and pneumonia. He also had a sleep disorder characterized by significant sleep apnea due to airway obstruction and poor onset of sleep. Sean depended on trained caregivers for all activities of daily living and was enterally fed via a gastrostomy tube. His primary caregiver was his mother, who reported an ability to discern preference based on facial expression, body language, and an idiosyncratic yes/no response (i.e. opening his mouth for yes, and pursing his lips for no). This response was not consistent, and usually required the interpretation of a familiar caregiver. Over the years, Sean had tried numerous assistive devices to facilitate communication, including a Big Mac switch mounted above Sean's left arm, a chin switch and lever switch. As well, different communication strategies had been entertained, including auditory scanning, a picture communication book and various switch-activated software programs. At the time of the study, these aforementioned devices and strategies had all been abandoned, and Sean's preferences and responses were primarily interpreted by the attending caregiver.

Instrumentation

The aforementioned peripheral ANS signals were non-invasively acquired using a ProComp Infiniti multi-modality encoder from Thought Technology Ltd. and a laptop computer. EDA was measured via two Ag–AgCl electrodes (SA9309M, Thought Technology Ltd.) placed on the palmar surface of the second and third finger of Sean's left hand. Heart rate was recorded using a photoplethysmography sensor (SA9308M, Thought Technology Ltd.) fastened to the tip of his fourth finger. Thoracic respiration was measured using a stretch sensitive belt (SA9311M, Thought Technology Ltd.) positioned around the upper aspect of the trunk. Skin temperature was detected with a 0.125 in. bead thermistor

Figure 1. Instrumentation set-up for acquiring the peripheral ANS signals in the present study (EDA; BVP = blood volume pulse).
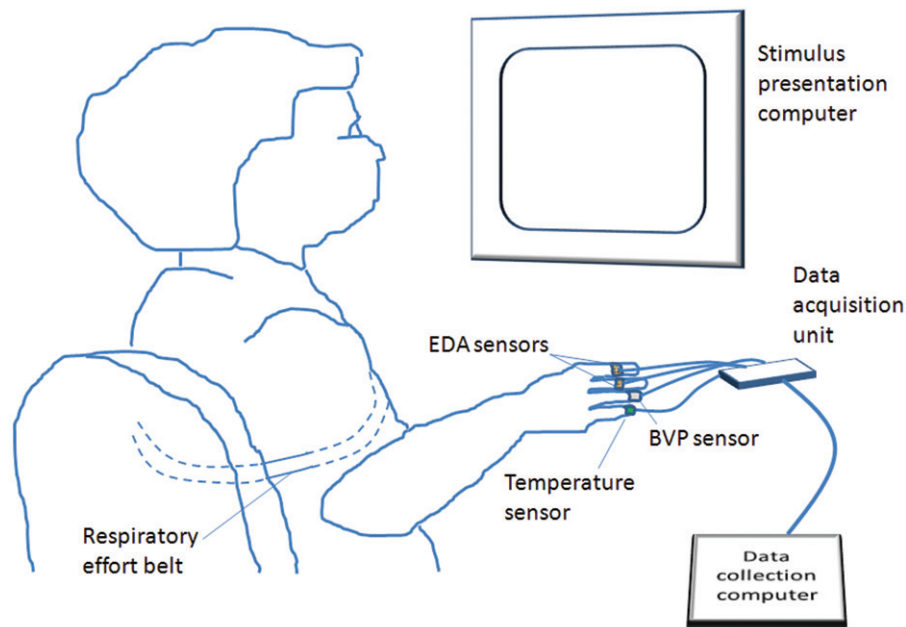
(SA9310M, Thought Technology Ltd.) securely fastened to the fifth finger of Sean's left hand. Figure 1 summarizes the instrumentation set-up.

Study protocol

Three data collection sessions were conducted over three different days within a one month period, in a controlled, indoor hospital environment with minimal distraction. Any unexpected events such as alerts from the Physical Address (PA) system, client coughing, sneezing, or extraneous movement were recorded by the researchers to help filter the data during the subsequent offline analysis. In each session, Sean was seated in his wheelchair, facing a computer screen. At the beginning of each session, a baseline recording of at least 300 s was obtained, wherein no stimulus was presented. This was followed by several stimulus presentations, as described in Table I. We consulted Sean's primary caregiver (his mother) to identify these candidate stimuli. The stimuli were presented on a laptop computer, which was connected to two speakers (for presenting the audio stimuli). Over the three sessions, Sean completed each stimulus test at least twice. No prior information about the stimuli was given to him. Since the purpose of this study was to examine Sean's natural reactions as manifested in his physiological signals, the researchers and his parent refrained from talking to him during the presentation of stimuli. Also, in order to prevent distraction during the tests, the researchers and the parent stayed out of Sean's sight, such that only the laptop, which presented the stimuli was in his field of view. The protocol was approved by the research ethics board of the paediatric rehabilitation hospital and affiliated university. Sean's mother provided written consent for participation in this study.

Data analysis

The recorded physiological data were analysed offline. Initially, each physiological signal was visually inspected for potential reactions, i.e. significant deviations from baseline values or patterns. To quantitatively detect these reactions, the following algorithms were employed.

EDRs, indicators of potential sympathetic nervous responses, were detected using the algorithm presented by Blain et al. [13], which tracks the centroid of the first derivative of the EDA signal. The total number of EDRs per trial was tabulated.

Significant changes in the slope of the temperature in either a positive or a negative direction were tracked. A least-squares line was fit to the first 5 s of data. The cumulative mean square error from this line was then computed at 1-s intervals. A significant slope change was identified when the cumulative mean square error exceeded an empirically determined threshold. At this point, a new least-squares line was estimated with the next 5 s of data and the above process iterates. The total number of significant changes in slope was tabulated for each trial.

The respiration rate and amplitude were calculated over 5-s windows for the baseline trial of each session with consecutive windows overlapping by 1 s. In the two-dimensional feature space defined by respiration rate and amplitude, we identified the centre of the cluster (mean vector) and assumed a radially symmetric distribution. The contour which enveloped 95% of the baseline data was defined as the boundary of the baseline state. For each of the subsequent trials recorded during that session, respiration rate and amplitude of each second of data recording were extracted, forming a trajectory in the two-dimensional feature space. Points that fell outside the boundary of the baseline state were indicative of a change in respiration. The total number of such changes was summed over the duration of each trial.

The inter-beat interval (IBI) of Sean's heart was calculated from the raw blood volume pulse data collected during each trial. Taking the inverse of the IBI signal yielded an estimate of Sean's heart rate. For the duration of the trial, changes in heart rate were automatically scored, such that an increase in heart rate (i.e. acceleration) yielded a score of $+1$, and a decrease in heart rate (i.e. deceleration) yielded a score of -1.

Table I. Stimuli investigated in reaction tests with the client.

Factor	Variations	Detail
(1) Presentation modality	(a) Visual	A random sequence of two types of visual stimuli: (1) images of the planets in the solar system and (2) picture of persons very familiar to the client (his mother and sister). Duration: 100 s
	(b) Audiovisual	A random sequence of two types of visual stimuli: (1) fireworks with sound and (2) grey static screen without sound. Duration: 80 s
(2) Familiarity with stimulus	(a) Content	A random audio sequence of words unfamiliar to the client (name of different planets in the solar system) randomly interrupted with one familiar word (client's name). Duration 100 s
	(b) Tone	Repetition of experiment 2a with words spoken in familiar (maternal) and unfamiliar voices. Duration: 100 s
(3) Colour	Red, blue, green, yellow, and black	Picture of a toy car moving in random directions on the display. Every 5 s, the car picture colour changed. Duration: 100 s
(4) Startle	(a) Random sounds-paper crumple	A random sequence of dull repetitious sounds (e.g. hammering and knocking) and paper crumple (startle stimulus identified by client's parent). Duration: 100 s
	(b) Waves and glass breaking	A random sound sequence of a shoreline waves (soft and relaxing sound) and glass breaking (sharp and startling sound). Duration: 80 s
(5) Anticipation	(a) Drum roll-ta-da	An auditory pattern consisting of a drum roll followed immediately by a ta-da alert. After several repetitions the drum roll was followed by silence. Duration: 55 s
	(b) Knock-chime	An auditory pattern consisting of knocking followed immediately by a high pitch chime. After several repetitions, the knocking was followed by silence; duration 55 s
(6) Association	(a) Talk-clap	An auditory pattern consisting of a lady's voice saying 'one, two, three, clap!' followed immediately by the sound of applause. After several repetitions, the speech was followed by silence; duration 58 s
	(b) Talk-music	An auditory pattern consisting of a man's voice saying '[*Client's name*], let's play music!' followed immediately by music. After several repetitions, the speech was followed by silence; duration 85 s

The net change in heart rate (sum of the indicator sequence) and the local maximum heart rate were estimated over consecutive 5-s windows where neighbouring windows overlapped by 1 s. The resulting points populated a two-dimensional feature space defined by the net change in heart rate and the local maximum heart rate. The points generated during baseline were assumed to have originated from the same physiologic state; the centroid of all points was calculated and a radially symmetric joint distribution was assumed. The contour which encompassed 95% of the points was defined as the boundary of the baseline state. The same two features (i.e. net change in heart rate and local maximum heart rate) were extracted for each second of each subsequent trial. Those that fell outside of the boundaries of the baseline cluster were flagged as changes in cardiac activity. The total number of such changes was summed over each trial.

Since all of the above analyses proceeded in one segment increments, there was at most one reaction per second per signal modality. For each trial, we counted the number of reactions, N, among the four physiological signals. Equivalently, this quantity could be interpreted as the total time in seconds where Sean was responsive. For each condition in Table I, the tally, N, of reactions was compared with the total number of physiological reactions during baseline via a χ^2 test of homogeneity of proportions [29] at a 5% significance level.

Results

Figure 2 shows an example of Sean's baseline EDA, heart rate, temperature, and respiration signals. Note the monotonic behaviour of the EDA and temperature recordings, and the periodic pattern of respiration and blood volume pulse signals. The dip in respiration amplitude at about 60 s is likely due to a shift in Sean's trunk position, which in turn induced a sudden change in the average tension of the respiration band.

Table II summarizes the number of reactions during the stimulus trials and their comparison with baseline reactions. For this comparison, we included all the baseline recordings from Sean, resulting in a total of 2331 s of recording time and 520 reactions. The cells marked with asterisks represent trials where the total number of physiological reactions significantly differed from that observed in the corresponding sessional baseline (χ^2 test, $p < 0.05$).

Table III presents the distribution of detected reactions across the four physiological signals within each type of trial. In all trials except one (anticipated aural pattern), Sean's temperature signal was his most labile physiological modality, implying that he is a temperature responder. Between the four physiological signals, respiration amplitude demonstrated the smallest proportion of reactions. Given that Sean had ongoing respiratory difficulties and a history of pneumonia, respiratory lability may have been compromised.

Discussion

The stimuli used in this study were selected at the discretion of Sean's mother, who was his primary caregiver. Our results include instances of agreement between caregiver predictions and reactions in Sean's physiological signals. Figure 3 depicts sample recordings from the presentation modality test
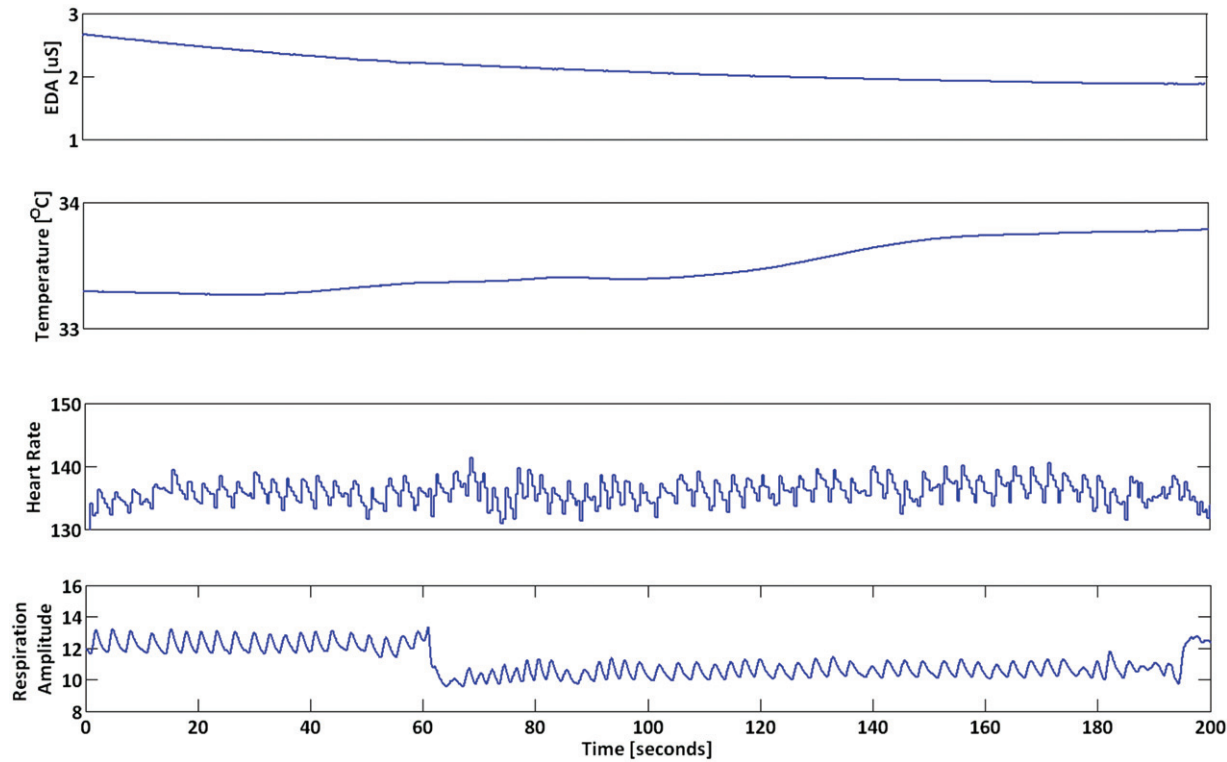
Figure 2. Client's physiological signals in relaxed state (baseline); from top to bottom: EDA, fingertip temperature, heart rate, and respiration amplitude. Heart rate and respiration amplitude are dimensionless quantities [30].

Table II. Trials with a significantly different, i.e. either significantly higher or significantly lower.

Factor	Trial	Session	Number of physiological reactions
Presentation modality	Visual	1	5*
		2	17
	Audiovisual	1	24*
		2	13
Familiarity with stimulus	All content in familiar tone	1	21
		2	6*
	All content in unfamiliar tone	1	24
		2	22
	Familiar content in familiar tone	1	23
		2	26
	Familiar content in unfamiliar tone	1	24
		2	35*
Colour	Red–blue–green–yellow–black	1	22
		2	23
		3	29*
Startle	Startling sound	1	3*
		2	22
		3	16
Anticipation	Anticipated aural pattern	1	17
		2	18*
Association	Associative aural pattern	1	19
		2	6

*Number of physiological reactions from that observed in the baseline condition, using χ^2, $p < 0.05$.

(audiovisual trial, session 1 in Table II), for which the total number of physiological reactions was significantly different from baseline ($\chi^2 = 3.93$, $p = 0.0476$). This finding echoed the perspective of the parent, who predicted considerable reaction to the audiovisual stimulus.

There were also cases where the number of reactions derived from the physiological signals contradicted the parent's prediction. Of particular interest are the factors for

which Sean's mother expected intense reactions, but in fact elicited no significant change in Sean's physiological signals. One example is the case of test 2(b) in Table I. In this trial, Sean listened to a random audio sequence consisting of words that were very unfamiliar to him (i.e. names of different planets in the solar system), and one familiar word (i.e. his name). The unfamiliar words were announced in a stranger's voice, while the familiar word was presented in his mother's

Table III. Proportion of reactions in each physiological signal. Each row shows the proportion of the total number of reactions in the given trial occurring in each physiological modality.

| Factor	Trial	Reactions			
		EDA (%)	Temperature (%)	Respiration (%)	Heart rate (%)
Presentation modality	Visual	23	31	19	27
	Audiovisual	19	29	15	37
Familiarity with stimulus	All content in familiar tone	32	40	14	14
	All content in unfamiliar tone	27	29	18	26
	Familiar content in familiar tone	27	31	16	26
	Familiar content in unfamiliar tone	30	31	14	25
Colour	Red–blue–green–yellow–black	27	27	22	24
Startle	Startling sound	23	54	5	18
Anticipation	Anticipated aural pattern	37	26	8	29
Association	Associative aural pattern	27	31	19	23
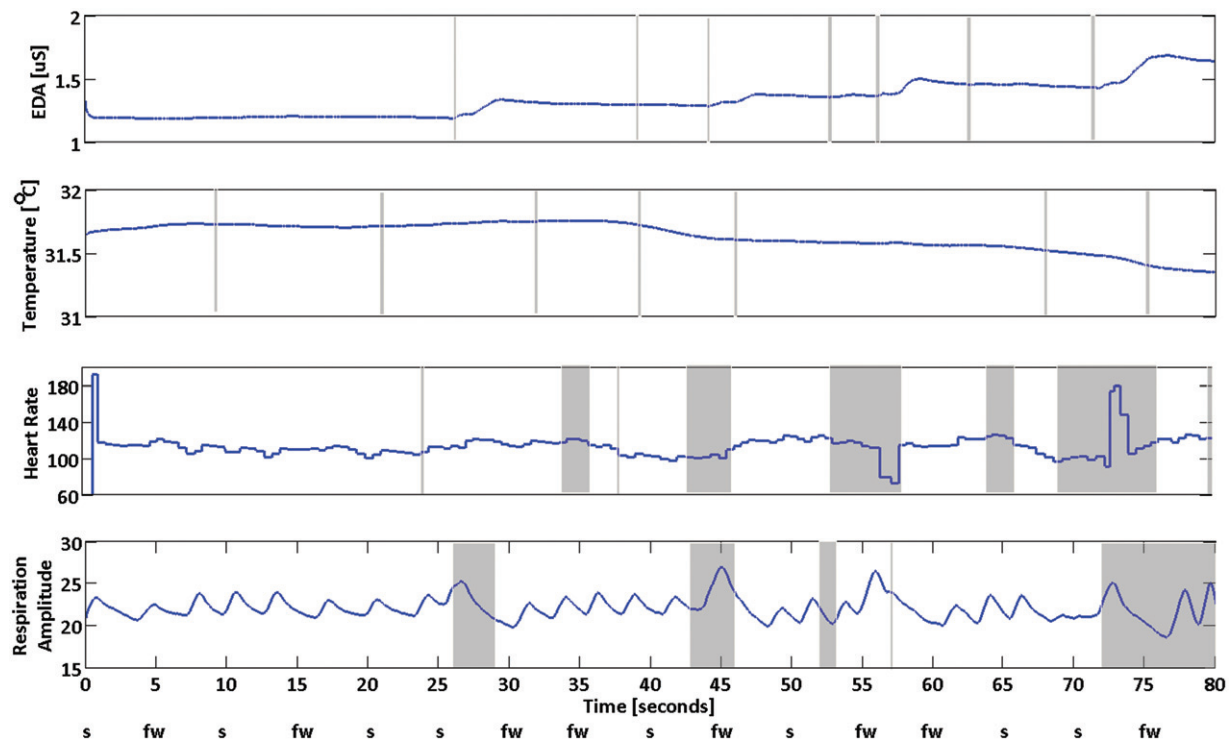
Figure 3. Client's physiological reactions to audiovisual stimulus as detected by the algorithm. The sequence of letters at the bottom of the graph identifies the type of stimulus: 's' denotes a static gray image, and 'fw' denotes a fireworks video. Vertical lines denote reactions in the EDA and skin temperature plots, while shaded blocks denote reactions in the respiration and heart rate signals, as detected by the algorithm. In this trial, the total number of reactions in the client's physiological signals $N = 24$ was significantly different from the corresponding sessional baseline ($\chi^2 = 3.93$, $p = 0.0476$).

voice. Sean's mother strongly believed that her son's reaction would be most significant in the latter case, as both the content and the tone of the keyword were very familiar to him. However, the physiological results proved the opposite. While there were scattered reactions to the familiar stimulus, no consistent reaction to the familiar stimulus was detected. We repeated this test in another session, and observed similar disagreement between the parent's prediction and physiological findings. This can be seen from Table II, under the third trial of the second factor (i.e. factor: familiarity with stimulus, trial: familiar content in familiar tone). For this type of trial, the total number of physiological reactions showed no significant change from baseline in both sessions.

Another case of strong disagreement between the parent's prediction and physiological reactions is seen in the paper crumple sound startle test (test 4(a) in Table I). While Sean's

mother predicted a definite reaction to the paper crumple sound, Sean's physiological signals exhibited very relaxed patterns, which resembled those of baseline. This is reflected in Table II. The total number of reactions to the startling sound was significantly lower than baseline in the first session, and showed no significant difference from baseline in the second and third sessions.

These results suggest that the caregiver's understanding of the client's reactions to sensory and perceptual stimuli may be incomplete. Physiological data may complement caregiver acumen in deciphering the reactions of non-verbal clients with severe and multiple disabilities.

In the long run, if a client is trained sufficiently to realize that his feelings or choices can be conveyed through his body signals, then he/she may perceive a sense of control over the outcome of different situations. Furthermore, drawing on

evidence from biofeedback studies [31, 32], the client can potentially be trained to voluntarily control his/her peripheral autonomic signals and thus attain an independent voice. This may avert the cognitive, motivational, and affective deficits associated with learned helplessness [33, 34].

Limitations

A physiological signal may change as a result of any number of internal or external stimuli. For example, EDRs may arise from spontaneous increases in EDA, internal stimulation such as mental activity, and external stimulation such as startling noises [35–37]. The task of separating out the source of these changes is non-trivial and has yet to be successfully addressed. In this study, we implicitly assumed that all physiological changes were due to unconscious reactions to the presented stimuli, running the risk of reporting inflated reaction counts. To mitigate this risk, we have compared reaction counts to each stimulus against baseline activity, assuming that the non-stimuli induced reactions occur equally in all recordings.

While our results suggest potential value in physiological signals, reaction count analysis alone is unlikely to tell the complete story about Sean's feelings and preferences. Pattern-based analyses, a broader range of affective stimuli, additional physiological signals, a greater number of test sessions and replication with a larger population are required to gain a fuller appreciation of the client preferences. Furthermore, the method in this study did not give us any information about valence, only arousal.

Conclusion

We explored the potential of identifying arousing stimuli for a non-verbal client through analysis of his physiological signals. We followed an exploratory approach through which the effect of six different groups of stimuli on the client's electrodermal, thermal, respiration, and heart rate reactions was investigated. We found that the client was more aroused by audiovisual stimuli, aural stimuli articulated in unfamiliar voice, and aural stimuli containing anticipatory patterns. These findings did not agree with the prediction of client's mother about her son's reaction to these stimuli, implying that physiological data may complement caregiver acumen in deciphering the reactions of non-verbal clients with severe and multiple disabilities. We also found that temperature was the most labile physiological signal of the client in this case study.

Declaration of interest

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