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Original Article

Randomized testing of taste discrimination in a case of congenital aglossia

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ABSTRACT

Aims: Isolated Congenital Aglossia (ICA) is a rare syndrome where an individual is born without a tongue. A few anecdotal reports have identified taste as a sensation experienced by the person with congenital aglossia (PWCA). To date no systematic investigations have been reported. This study aimed to systematically determine gustatory function in a PWCA.

Methods: The current study utilized a randomized, double-blinded, controlled trial that tested the five basic tastes: sweet (sucrose), sour (acetic acid), salty (sodium chloride), bitter (caffeine), and umami (monosodium glutamate, MSG) in a 44 year old female PWCA. Five concentration levels (three for salty) were tested in triplicate for each stimulus. A nose clip was used to exclude contribution by olfactory detection. Contingency tables were constructed to determine relationships between identification accuracy and stimulus or concentration level.

Results: The sweet (17.1 g/L), salty (0.58 g/L), and bitter (0.02 g/L) stimuli were detected at comparable concentrations to those reported in non-randomized trials, while sour (0.02 g/L) was detected at a lower concentration. The most common substitution was salty for umami ($n = 7$). Identification accuracy was significantly associated with taste stimuli $\chi^2 = 12.634$, $p = 0.013$. Concentration level was significantly associated with identification accuracy only for salty, $\chi^2 = 9.000$, $p = 0.011$.

Conclusion: This study has demonstrated the perception of different tastes in a PWCA. This is the first known report of umami being identified as a unique taste in a PWCA. Variations in threshold taste concentrations compared to normal individuals indicate certain gustatory dysfunction.

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The aim of this current study was to extend previous research by scientifically examining gustatory function in a case of isolated congenital aglossia (ICA). This rare syndrome, in which an individual is born without a tongue, was first reported by de Jussieu in 1718. Since then, eleven reports of congenital aglossia (CA) have appeared in the literature without the presence of other syndromes or symptoms.[1–13] Co-anatomical morbidities that have been associated with ICA are generally limited to micrognathia (abnormally small lower jaw), which is a common finding among hypoglossia and aglossia, as it is unlikely that normal mandibular formation would occur without the development of the tongue.

It is well known that the tongue's primary biological functions are in the areas of taste, mastication and deglutition. In the normal population, the tongue plays a major role in the sensory discrimination of taste, through receptors which are located on the tongue surface.[14] This sensation would logically be disrupted without the structure, yet there are reports in the literature[2,4–9,12,13,15–18] mentioning or describing taste awareness in persons with congenital aglossia (PWCA). These reports describe the ability to taste but do not go into any particular detail with the exception of three authors. Eskew and Shepherd indicated taste could be distinguished only on the surface of the soft palate following application by small brush in various regions of the oral cavity at threshold levels of 3% cane sugar, 1% sodium chloride, 0.1% sulfuric acid, and 0.01% quinine.[2] Goto reported testing of taste with resulting thresholds of 5% cane sugar, 10% sodium chloride, 0.1% quinine, and 1% acetic acid.[4] The solutions were applied with a brush to various areas of mucus membrane in the oral cavity. Results indicated taste was distinguished in the sublingual area and anterior faucial pillers. In 2008, Salles et al included a description of taste testing in a 14 year old female PWCA who was reported to eat her family's regular diet without difficulty.[13] Results indicated recognition of acid taste from the lowest concentration (sulfuric acid, 0.015 g/L), bitter was reported as a new taste at the third concentration (quinine, 0.012 g/L) and was perceived as sweet upto the last concentration; sweet was perceived as a new taste at the fifth concentration (sucrose, 8 g/L) and perceived as bitter at the last concentration (16 g/L); salty was perceived as a new taste at the forth concentration (sodium chloride, 0.75 g/L) and identified as salty from the fifth concentration on (1.5 g/L). Salles' results suggested inverted sweet and bitter tastes could be considered a difficulty in perception; however, there appeared to be a greater sensitivity to salty and acid substances. Salles and Costa-Bezerra tested the taste sensitivity of the 44 year old PWCA from the current study at the University of Brasilia in 2013[19] (personal unpublished report to Dr. McMicken). The identical flavors from Salles et al,[19] as in 2008,[13] were presented in random order with doses of increasing concentrations. Water was given between each presentation. Each flavor had 5 concentrations. When the PWCA identified a flavor, no further same substance concentrations were offered. A nose clip was not used. Threshold results were: acid (citric), 0.015 g/L lowest concentration; bitter (caffeine) 0.003 g/L, lowest concentration; salt (sodium chloride) 0.09 g/L, lowest concentration and sweet (sucrose), 0.2.0 g/L, 3rd concentration. These researchers conclude the PWCA appeared to have greater sensitivity than their normal control group.

Limitations of these four previous reports include: no details of the experimental methodology, the number of trials, the total substance dilution amounts, complete taste substitution details, and in most cases, whether a nose clip was used to lessen the chance of a possible olfactory component.

1. Background on taste in the normal population

In the normal population, taste buds are in groups of upto 100 neuroepithelial cells embedded in the epithelium of the oral cavity. In humans, there are approximately 5000 taste buds in the oral cavity. They are located on the surface of the tongue, the palate, and the epiglottis.[20,21] It is the taste buds or sensory receptors on the surface of the tongue, activated by saliva, which are responsible for perception and taste discrimination of sweet, sour, salty, bitter, and umami (the taste of glutamate and other L-amino acids).[22] According to Kinnamon, taste buds on the anterior two thirds of the tongue, located within papillae, are innervated by the cranial nerve VII, the facial nerve. Taste buds on the posterior tongue are housed in a different group of papillae, and are innervated primarily by cranial nerve IX, the glossopharyngeal nerve.[20] Taste buds on the soft palate are innervated by a branch of the lingual nerve, while the epiglottis and larynx by the superior laryngeal nerve, which is a branch of the cranial nerve X (vagus nerve). Despite the various locations of taste buds in different sections of the oral cavity, and supposed differences in regional sensitivity on the tongue, the concept of a tongue map with zones for sweet, bitter, salty, and sour has been proven to not be a valid or reliable concept.[23] Gustatory function and cell specificity is reportedly maintained regardless of location. Kinnamon stated that despite the often-related tongue diagram of taste, it is well known that there is no credible map of taste sensitivity on the tongue, although variations in thresholds are present in the many different oral cavity locations.[20]

The olfactory and gustatory senses are closely linked in their chemo-sensitivity.[24] Testing for taste with or without the presence of a tongue must be accomplished without olfactory influence. As noted by Steele et al, an additional complication when testing for taste is the olfactory receptors possibly being responsive to the odor.[25] To avoid this possibility, it is recommended an odorless stimulus be used in research as well as the implementation of a nose clip.

The present research explores the gustatory threshold levels in the PWCA described in prior research.[9–11] The following questions guided the current research:

1) What is the taste sensitivity response of the PWCA in a randomized, multi sample, double-blind controlled study using sweet, sour, salty, bitter, and umami substances as taste stimuli.
2) How do the thresholds from current study in 2014 compare with previous reported thresholds obtained on this PWCA at the University of Brasilia[19]?
3) What are the confusion substitutions in taste testing with this PWCA?

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