

Titre de l'étude

Etude de tolérance d'administrations intranasales répétées d'ocytocine selon 3 schémas d'administration chez des bébés présentant un SPW

Auteurs/PI/Institution/Labo

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Contexte/Objectif de l'étude

Etudier la tolérance d'administrations intranasales répétées d'OT pendant 7 jours qui suivent la première dose selon 3 schémas d'administration chez des nourrissons présentant un SPW âgés de moins de 5 mois.

Prestation du Plateau Technique

Séquences :

T1W-IR : 1*1*1.5 mm, 10mn50

rsfMRI : 3*3*3mm, TR/TE= 2800/33ms, 200dyn

Matériel :

- Antenne 32 CX

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The Use of Oxytocin to Improve Feeding and Social Skills in Infants With Prader-Willi Syndrome

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abstract

BACKGROUND AND OBJECTIVES: Patients with Prader-Willi syndrome (PWS) display poor feeding and social skills as infants and fewer hypothalamic oxytocin (OXT)-producing neurons were documented in adults. Animal data demonstrated that early treatment with OXT restores sucking after birth. Our aim is to reproduce these data in infants with PWS.

METHODS: We conducted a phase 2 escalating dose study of a short course (7 days) of intranasal OXT administration. We enrolled 18 infants with PWS under 6 months old (6 infants in each step) who received 4 IU of OXT either every other day, daily, or twice daily. We investigated the tolerance and the effects on feeding and social skills and changes in circulating ghrelin and brain connectivity by functional MRI.

RESULTS: No adverse events were reported. No dose effect was observed. Sucking assessed by the Neonatal Oral-Motor Scale was abnormal in all infants at baseline and normalized in 88% after treatment. The scores of Neonatal Oral-Motor Scale and videofluoroscopy of swallowing significantly decreased from 16 to 9 ($P < .001$) and from 18 to 12.5 ($P < .001$), respectively. Significant improvements in Clinical Global Impression scale scores, social withdrawal behavior, and mother–infant interactions were observed. We documented a significant increase in acylated ghrelin and connectivity of the right superior orbitofrontal network that correlated with changes in sucking and behavior.

CONCLUSIONS: OXT is well tolerated in infants with PWS and improves feeding and social skills. These results open perspectives for early treatment in neurodevelopment diseases with feeding problems.

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WHAT'S KNOWN ON THIS SUBJECT: In a Prader-Willi syndrome mouse model early oxytocin administration can strongly modify the course of the disease with short- and long-term effects on feeding and social skills. There are no data of oxytocin effects in human infants.

WHAT THIS STUDY ADDS: We report that 7-day intranasal oxytocin administration in infants with Prader-Willi syndrome is well tolerated and improves sucking/swallowing, social skills, and mother–infant interactions. Changes in brain connectivity of superior orbitofrontal cortex correlate with clinical improvements.

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