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Research Report

Vocal and gestural productions of 24-month-old children with sex chromosome trisomies

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Abstract

Background: Children with sex chromosome trisomies (SCT) frequently show problems in language development. However, a clear description of the communicative patterns of these children is still lacking.
Aims: To describe the first stages of language development in children with SCT in comparison with those in typically developing (TD) children. The purpose was to verify the existence of possible differences in communicative skills (in both vocal and gestural modality) and identify the presence of possible early predictors (i.e., low vocabulary size and low gesture production) of later language impairment in children with SCT.
Methods & Procedures: Fifteen 24-month-old children with SCT (eight males with Klinefelter syndrome (KS) and seven females with triple X syndrome (TX)) and fifteen 24-month-old TD children (eight males and seven females) participated in the study. Their spontaneous communicative productions were assessed during a semi-structured play session in interaction with a parent. In addition, their vocabulary size was assessed using a parental report (the Italian version of the MacArthur Communicative Development Inventories).
Outcomes & Results: With regards to their vocabulary size, 60% of children with SCT (75% of children with KS and 43% of children with TX) were at risk for language impairments (i.e., they had a vocabulary size smaller than 50 words). In addition, TD children showed better lexical and syntactic skills than children with SCT in their spontaneous communicative productions. However, the production of communicative gestures was higher in children with SCT than in TD children. Boys with KS appeared to differ from TD males in more aspects of communication than girls with TX differed from TD females.
Conclusions & Implications: The study showed the importance of early detection of language risk factors in children with SCT, while also considering the use of compensatory strategies (e.g., the use of communicative gestures).

Keywords: sex chromosome trisomies, language development, gestures, late-talking children, Klinefelter syndrome, triple X syndrome.

What this paper adds

What is already known on the subject

Children with SCT usually exhibit impairments in language development. However, a clear description of the early communicative competencies of these children is still lacking.

What this paper adds to existing knowledge

The study shows that a high percentage of children with SCT are at risk for language impairments. However, the presence of compensatory strategies (i.e., the use of communicative gestures) is noted. Different communicative patterns are found for males with KS and females with TX: having KS appeared to be a higher risk factor for language impairment than having TX.

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What are the potential or actual clinical implications of this work?
The early detection of language risk factors in this population allows intervention during the early stages of children's communicative development and appropriate intervention when needed.

Introduction

Sex chromosome trisomies (SCT) are genetic syndromes characterized by the presence of three sex chromosomes rather than the normal two. Three different conditions have been identified as follows: females with 47,XXX karyotype (i.e., triple X syndrome—TX) and males with 47,XXY (i.e., Klinefelter syndrome—KS) and 47,XYY (47,XYY syndrome) karyotype. The trisomy of sex chromosomes compared with that of autosomes (i.e., trisomy 21, 18 or 13) has a relatively mild impact on children's development. This is because, first, most genes on the Y chromosome are involved only in sex-specific characteristics and do not affect cognition, and second, only one of the X chromosomes is fully functional due to X chromosome inactivation (i.e., a process by which one of the copies of the X chromosome present in female mammals is inactivated) (Willard 2000).

The prevalence of SCT is not well known, especially because many cases likely go undetected due to their mild phenotype. Boyd *et al.* (2011) found an overall prevalence of SCT of 1.88 per 10,000 births in Europe. Recent data suggest an incidence of approximately 1 in 1000 female births for TX (Messer *et al.* 2013), approximately 1 in 1000 male births for 47,XYY syndrome (Ross *et al.* 2009), and between 1 in 426 and 1 in 1000 males for KS (Bojesen *et al.* 2003). However, it has to be noted that KS is more easily and frequently detected due to the impact on individuals' sexual development (i.e., frequent problems with secondary sexual characteristics and fertility issues). A study conducted in Australia (Herlihy *et al.* 2011) identified KS as the most common chromosomal aberration in men, with an incidence of up to 1 in 450 men.

The first studies on SCT (e.g., Johnston *et al.* 1974) showed increased rates of intellectual disability and other mental health problems in these individuals. However, these studies were biased because the only participants were individuals with the most severe phenotypes who were identified due to their significant neuropsychological problems. In fact, prospective studies based on individuals identified by prenatal or newborn screenings have shown that the cognitive outcomes of these children are much better than the original literature suggested (e.g., Boada *et al.* 2009).

Although there are phenotypic differences between the three SCT, people with these genetic conditions usually share a tendency to have problems with language development. In fact, as Bishop *et al.* (2011) noted, the rates of language and communication impairments are high in all the SCT. The principal similarity between these genetic syndromes is language impairment (Bishop *et al.* 2011), although motor impairment has also been found in these conditions (Salbenblatt *et al.* 1987, 1989).

Since an association between early language delay and later risk for persistent language impairment has been found in several prospective studies (e.g., Dale *et al.* 2003, Rescorla and Turner 2015, Rice *et al.* 2008), the present study focused on children's early communicative skills to detect the presence of possible delays or atypical patterns. In particular, we focused our attention on the production of words and communicative gestures in 24-month-old children with SCT, because this age has been widely considered a key time point in identifying late-talking children, that is, those children with a vocabulary size below 50 words at 24 months and at risk of developing a language impairment (Rescorla 1989, Rescorla *et al.* 2014). Although many factors, including early child characteristics, socio-economic status and maternal factors, have been found to be associated with language outcomes (Reilly *et al.* 2010), the present study focused on two recognized predictors of possible language delays or impairment: a low vocabulary size and a low gesture production. With regard to word production, there is a wide agreement in considering low vocabulary size as a possible predictor of later language impairment (e.g., Rescorla 1989, Paul *et al.* 1991). Some follow-up studies have found that approximately half the children who are identified as late talking at 24 months fail to catch up to their peers by 36 months (e.g., Paul *et al.* 1991), although other studies (e.g., Dale *et al.* 2003) found that some children whose language is delayed at 24 or 30 months catch up over the next few years. With regard to gesture production, many studies have shown that children's use of gestures is associated with their later vocabulary development (e.g., Iverson and Goldin-Meadow 2005, Suttora and Salerni 2012, Zampini and D'Odorico 2009), in particular Thal *et al.* (1991) found that truly delayed children were significantly poorer in gesture production than late-talking children who caught up.

In the present study, we focused our attention on language development in children with KS and TX. Children with 47,XYY were not included due to the small number of children with this diagnosis in our service at the moment of the study. In the following we

give a description of the characteristics of children with KS and TX.

Klinefelter syndrome (KS)

KS was first described by Klinefelter *et al.* (1942), who identified a group of male adults presenting with infertility, hypogonadism, gynaecomastia and increased gonadotropin levels. However, it was only in 1959 that its genetic cause, which consists of an extra X chromosome in males, was identified by Jacobs and Strong (1959). Individuals with KS usually show endocrine dysfunction that leads to fertility problems.

As shown by Bishop and Scerif (2011), the neuropsychological profile of children with KS has some characteristics in common with that of children with specific language impairment: their language development lags behind other aspects of development, and their IQ is in the normal range. Expressive language is typically more severely impaired than receptive language. Moreover, a high rate of academic difficulties and learning disabilities has been identified in these children at school age (Bishop *et al.* 2011, Rovet *et al.* 1996, Stewart *et al.* 1986).

A systematic review in patients with KS confirmed at birth (Bender *et al.* 1986) showed that the IQs of children with KS have a normal distribution but with the mean score shifted slightly to the left. Therefore, a greater percentage of individuals with KS than of the normal population scores in the low average range. In addition, Bender *et al.* (1990) showed that their overall mean intelligence scores on standardized tests were in the average range, but their mean verbal cognitive scores were approximately 10 points lower than their performance scores, although atypical motor development has been identified in these children (Ross *et al.* 2008, Simpson *et al.* 2003).

Attention deficit and hyperactivity disorder (ADHD) is frequently detected in children with KS (Lee *et al.* 2011, Tartaglia *et al.* 2012). Tartaglia *et al.* (2010), studying a group of 57 children and adolescents with KS, found that 34% of their participants showed predominantly inattentive symptoms, whereas only 2% showed combined inattentive and hyperactive symptoms. Moreover, the rates of autism spectrum disorder (ASD) in children with KS are above population levels, even when considering children identified through prenatal screenings, as found by Bishop *et al.* (2011), who reported that 11% of their participants with KS had diagnoses of ASDs (two out of 19 participants). The increased incidence of both ADHD and ASD have to be considered in preschool-age children due to the frequent comorbidity between these disorders and language impairment (e.g., Loucas *et al.* 2008, Redmond 2016).

However, it must be noted that there is significant variability between individuals with KS: many children and adolescents have minimal or no behavioural problems, whereas others show significant difficulties (Tartaglia *et al.* 2010).

Triple X syndrome (TX)

TX was first described by Jacobs *et al.* (1959), who identified this syndrome in a woman with normal intelligence and fertility problems. Similar to the case for KS, the first description of the TX phenotype was biased by the identification of patients with more severe clinical features. However, TX is usually diagnosed incidentally, mostly by prenatal screening, and the majority of women with TX are unaware of their condition because they are only mildly affected or asymptomatic (Lalatta *et al.* 2010).

Similar to boys with KS, girls with TX also have an increased risk of language delay, mild learning disability and poor motor coordination. In their literature review, Leggett *et al.* (2010) found a significant reduction in IQ of approximately 20 points in children with TX. However, there are some differences between the phenotypes of children with KS and TX. The cognitive profile is more unbalanced in children with KS because they tend to have higher nonverbal than verbal skills, whereas in girls with TX, both verbal and nonverbal abilities are frequently equally impaired (Leggett *et al.* 2010). However, these data are in contrast with a previous study that showed that deficits in verbal IQ were significantly greater than those in performance IQ in girls with TX (Netley 1986).

Language impairment is highly represented in the population of children with TX. In fact, Bishop *et al.* (2011) found that 24% of girls in their study (i.e., seven out of 30) had received speech and language therapy. In contrast, ASD was not found in any of the girls with TX in that study.

Aims

Although the presence of language impairment has been frequently reported in children with SCT, a clear description of the communicative patterns of these children is still lacking. Moreover, to the best of our knowledge, no observational data on the spontaneous communicative productions of young children with SCT have been reported in the literature.

The present study aimed to describe the first stages of language development in children with KS and TX, in comparison with those in typically developing (TD) children, to verify the existence of different communicative skills (in both vocal and gestural modality) and to identify the presence of possible early

predictors (i.e., low vocabulary size or lack of gesture production) of language impairment in children with SCT. As reported above, the study focused on the communicative productions of 24-month-old children because this age has been widely considered a key time point in identifying late-talking children (Rescorla 1989). The evidence of specific communicative patterns in these children could be useful for the early identification of those children who need support and to programme specific interventions to enhance their communicative abilities.

Methods

Participants

In total, 30 children participated in the study: 15 children with SCT (eight males and seven females) and 15 TD children (eight males and seven females). The children with SCT were recruited through the hospital Fondazione IRCCS Cà Granda Ospedale Maggiore Policlinico in Milan, Italy, where they were participating in a monitoring programme. Among the SCT children, eight had a diagnosis of KS (i.e., males with 47,XXY karyotype) and seven had a diagnosis of TX (i.e., females with 47,XXX karyotype). All the children were diagnosed before birth by amniocentesis. None had additional genetic and/or neurological conditions. They had normal hearing and no history of ear infections. Inclusion criteria for children with SCT were full-term birth and belonging to monolingual Italian-speaking families. The TD participants were selected from a sample of children who were participating in a monitoring programme by the Department of Psychology at the University of Milano-Bicocca. All the TD children were born at term, came from monolingual Italian-speaking families, and none was reported by parents as having any developmental or hearing problem, or history of ear infections. The methods used in the study were approved by the Ethic Committee of the University of Milano-Bicocca. Children's parents signed a written informed consent form prior to inclusion in the study.

The children's mean chronological age was 24 months (SCT group: mean = 24.33, SD = 0.88, range = 23–26. TD group: mean = 24.40, SD = 0.51, range = 24–25). The mean developmental quotient, as assessed by Griffiths Developmental Scales (Luiz *et al.* 2006) or Bayley Scales (Bayley 1993), was 102.53 (SD = 10.88, range = 93–124) in children with SCT and 107.36 (SD = 8.55, range = 92–120) in TD children. Therefore, all the children with SCT had appropriate psychomotor development. The two groups of children were not statistically different for chronological age ($U = 105$, $p = 0.775$) or developmental quotient ($U = 73.5$, $p = 0.172$).

Procedure

The children's vocabulary size was assessed by their parents' compilation of the Italian version of the MacArthur Communicative Development Inventories – words and sentences form (Il Primo Vocabolario del Bambino—PVB; Caselli and Casadio 1995, Caselli *et al.* 2007).

Moreover, to observe the children's spontaneous communicative productions, 15-min (mean = 14.62, SD = 1.30, range = 9.5–15.5) semi-structured play sessions in which children interacted with a parent were video recorded. All the children participated in the observation sessions with their mothers, with the exception of two children with SCT who participated with their fathers. Parents were instructed to play with their children as usual using the following set of toys: a farm, some children's books and a doll with a play food set. Every 5 min a new toy was introduced, but the children were allowed to play with all the items presented at will.

Both children's vocal and gestural productions were considered. All the children's utterances produced during the observation sessions were transcribed in CHAT format (Codes for the Human Analysis of Transcripts; MacWhinney 2000). An utterance was defined as a vocal production in a unique conversational turn, separated from other productions by a pause longer than 1 s (D'Odorico and Jacob 2006).

Coding and measures

The children's spontaneous utterances (i.e., those were not produced in immediate repetition of a parent's previous utterance) were classified as either preverbal or verbal (the coding scheme is adapted from Zampini and D'Odorico 2011).

The preverbal communicative productions were classified as follows:

- Preverbal level 1, i.e., communicative grunts (e.g., '*mh*') and vowel sounds (e.g., '*oh*').
- Preverbal level 2, i.e., babbling (e.g., '*da*', '*bababa*'), non-words (e.g., '*taca*', which has the structure of a word but is not a real word) and onomatopoeias (e.g., '*bau bau*' /woof woof/).

The verbal productions were classified according to the following hierarchical coding scheme:

- Single words, i.e., productions composed of a single significant element (e.g., '*casa*' /home/).
- Transitional forms, i.e., utterances composed of two or more vocal elements, of which at least one is a word, but which cannot be considered a real-word combination. Formulas (e.g., '*chi è?*' /who's it?/), horizontal repetitions (e.g., '*canta*

canta' /sing sing/), non-word combinations (e.g., '*bimbo taca*' /baby taca/, in which '*taca*' is a non-word) and chains (e.g., '*macchina casa*' /car house/, in which the two words are not connected by a semantic link), as well as utterances composed of a single word produced in association with a closed-class word (e.g., '*la palla*' /the ball/) were coded as transitional forms. (For a complete list and detailed description of these utterances, see Zampini and D'Odorico 2011.)
- Word combinations, i.e., two or more words joined by a semantic link and uttered in close temporal succession (D'Odorico and Carubbi 2003) (e.g., '*bimbo bello*' /nice baby/; '*ballo qui*' /dance here/).

For each child, the total number of utterances per min and the number of utterances in each category per min were computed. In addition, the proportion of preverbal and verbal utterances from the total number of vocal productions was computed.

Moreover, the following measures were computed to assess children's vocabulary skills:

- Word tokens, i.e., the total number of words per min uttered during the observation session.
- Word types, i.e., the number of different word roots per min uttered during the observation session (e.g., '*mangi-o*' – '*mangi-a*' /eat – eat-s/ were considered a single word type).

In addition, the mean length of utterance (MLU) was computed on children's verbal utterances, considering the ratio between the total number of words uttered and the total number of verbal utterances (i.e., word tokens/number of verbal utterances).

The children's gestures produced during the observation sessions were classified according to the following coding scheme:

- Pointing, i.e., extending the index finger in the direction of an object, a person or an event.
- Showing, i.e., holding up an object in the listener's line of sight.
- Conventional, i.e., gestures that have a culturally defined meaning and form (e.g., nodding and waving bye-bye).
- Iconic, i.e., gestures that refer to objects, persons or events, reproducing a physical or functional characteristic (e.g., flapping arms to refer to an airplane).

For each child, the total number of gestures per min and the number of gestures in each category per min were computed.

Reliability

The intercoder reliability of two independent coders (blinded to group status) was assessed in 20% of the sessions (i.e., three sessions of children with SCT and three sessions of TD children). The mean occurrence agreement percentages (i.e., the percentage of events identified by both the observers) were 84% for the vocal productions and 80% for the gestures. The Cohen's K coefficient was calculated to assess the accuracy of the coding (i.e., the attribution of a specific code to an event): the K computed for the vocal productions was 0.79 (disagreements were solved by the intervention of a third coder), whereas no disagreement was found in the coding of gestures.

Data analyses

Due to the small number of participants, a non-parametric test (Mann–Whitney non-parametric test for independent samples) was used to compare both vocal and gestural productions in the two groups of children (i.e., children with SCT versus TD children). Moreover, to assess the presence of possible differences in the communicative profiles of children with KS and TX, the vocal and gestural productions of these children were compared. A male–female comparison in the TD group was performed to assess if gender differences potentially contributed to explaining the children's communicative profiles. In addition, the productions of children with KS were compared with those of TD males, and the productions of children with TX were compared with those of TD females. By convention, the p-value of 0.05 or less was considered statistically significant, but to be more conservative, a Bonferroni correction was applied for multiple comparisons. Therefore, for the subgroup comparisons the adjusted p-value of 0.01 or less was considered statistically significant.

Results

Children's vocabulary size

In relation to the number of words spontaneously produced by children, as assessed by parents with the PVB, the difference between children with SCT (mean = 92.80, SD = 98.31, range = 10–316) and TD children (mean = 326.20, SD = 115.07, range = 135–523) appeared to be statistically significant ($U = 211$, $p < 0.001$). As reported in figure 1, the vocabulary size of the TD children was consistent with normative data (Caselli *et al.* 2007) (i.e., the majority of TD children had a vocabulary size ranging from the 25th to the 75th percentile), whereas nine out of 15 children (six males and three females) in the SCT group had

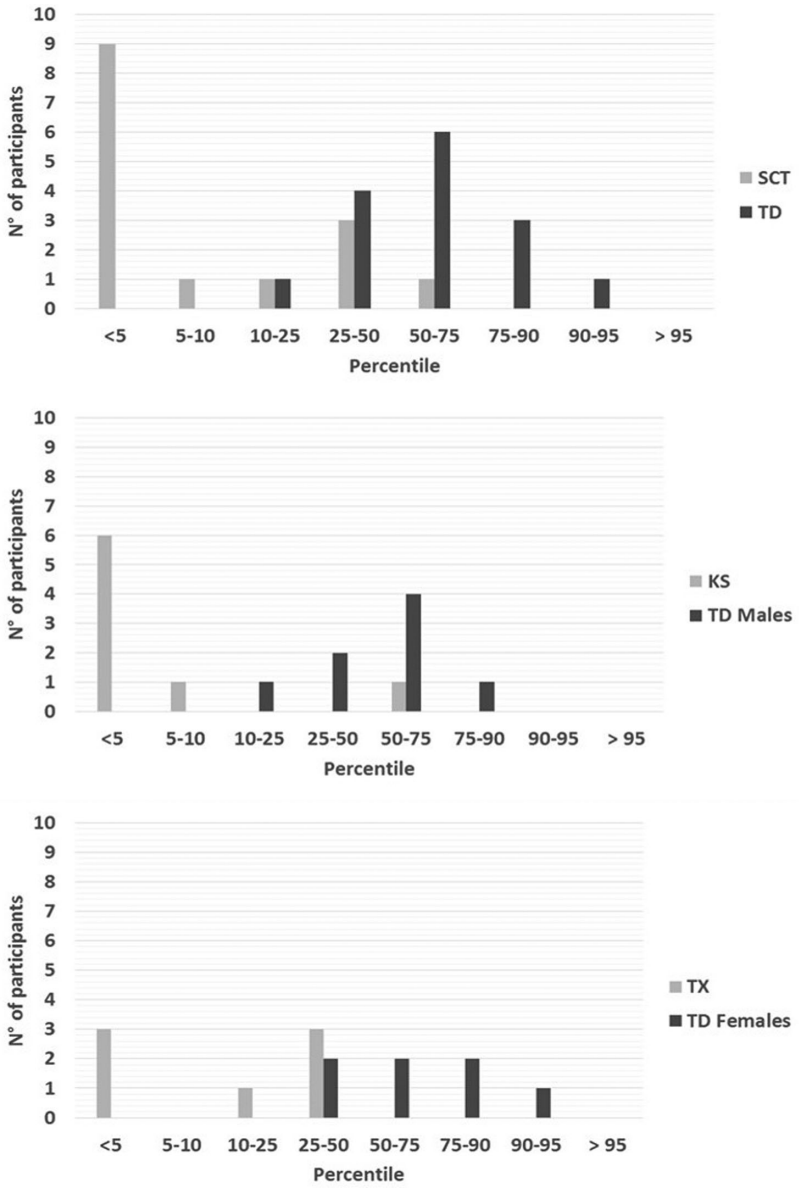

Figure 1. Number of participants in each PVB's percentile of word production

a vocabulary size below the 5th percentile. Therefore, 60% of children with SCT were at risk for language impairments.

Vocal and gestural production of children with SCT and TD children

Data on the proportion of preverbal and verbal utterances produced by the children in the two groups during the observation session are reported in figure 2. The proportion of verbal utterances was significantly lower in children with SCT than in TD children ($U = 17$, $p < 0.001$). The number of different types of utterances per min, the number of word types and tokens per min, and the MLU of the participants in the two groups are reported in table 1. The Mann–Whitney test showed a significantly higher production of single words, transitional forms and word combinations in TD children than in children with SCT, whereas a significantly higher number of preverbal level 1 productions (i.e., the simplest vocal productions) was found in children with SCT. Furthermore, in relation to lexical production and MLU, TD children showed higher competence, producing a significantly greater number of word types and tokens and longer utterances.

With regard to gestural production, the data reported in table 2 show a statistically significant difference only in the number of pointing gestures, which

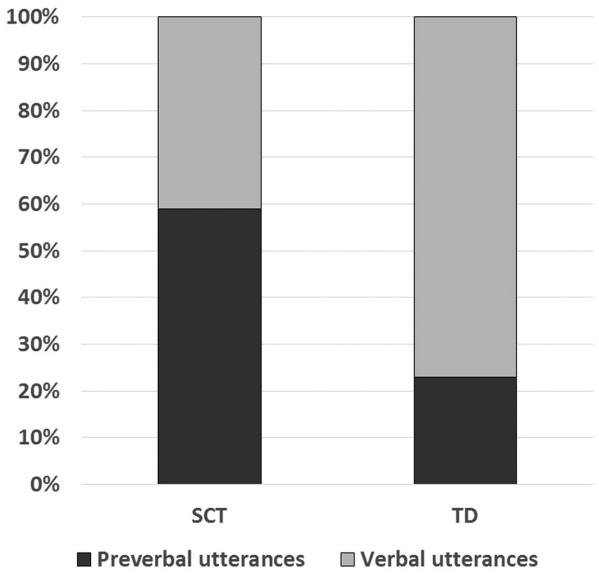

Figure 2. Percentage of preverbal and verbal utterances produced during the observation session

were produced more frequently by children with SCT than TD children.

Communicative profiles of children with KS and TX

To verify the existence of different communicative profiles in children with KS and TX, a comparison between males' and females' vocal and gestural productions was performed in each group. Figure 3 and 4, respectively, show the children's vocal and gestural productions per min. No statistically significant differences were found between boys with KS and girls with TX in either vocal (all p-values > 0.01) or gestural productions (all p-values > 0.01). The same result was found when comparing vocal (all p-values > 0.01) and gestural (all p-values > 0.01) productions of TD males and TD females. However, note that boys with KS appeared to be less competent than girls with TX in each vocal production category (i.e., they produced a higher number of preverbal utterances and a lower number of verbal utterances), whereas the differences between TD males and females were less marked. Additionally, with regard to gestural production, the differences between boys with KS and girls with TX appeared to be more marked than the differences between TD males and females. In particular, a higher production of gestures (total, pointing and conventional) was found in boys with KS, though this was not statistically significant.

Finally, to assess better the existence of specific communicative patterns in children with different SCT, the communicative productions of boys with KS were compared with those of TD males, and the communicative productions of girls with TX were compared with those of TD females (table 3). Children with KS showed a statistically significant ($p < 0.01$) higher number of preverbal level 1 productions and a statistically significant lower number of single words, word types and word tokens than TD males. In addition, children with KS showed a significantly higher number of pointing gestures than the control group. With regard to females, the differences between children with TX and TD girls were less marked; the girls with TX showed a significantly lower number of word types than TD females; in addition, their MLU was significantly lower. No significant differences were found in gestural production.

Discussion

The study aimed to describe the spontaneous communicative skills of 24-month-old children with SCT, in particular children with KS and TX. Both vocal and gestural productions were considered in order to assess their communicative skills. Note that, when considering the indirect data from parents' compilation of the PVB inventory, the majority of children with SCT are late-talking children. Therefore, they could be considered at risk for developing language impairment (Rescorla 1989, Rescorla *et al.* 2014). In particular, 75% of boys with KS and 43% of girls with TX had vocabulary sizes

Table 1. Vocal productions of the children in the two groups during the observation session

	SCT			TD				
	Mean	SD	Range	Mean	SD	Range	U	p
Preverbal level 1/min	**1.96**	1.43	0.60–5.53	**0.57**	0.51	0–1.42	**29.0**	**< 0.001**
Preverbal level 2/min	1.07	0.79	0–2.67	1.58	1.00	0.33–3.93	145.5	0.174
Single words/min	**1.73**	1.68	0–5.67	**4.02**	2.18	0.73–8.90	**187.0**	**0.001**
Transitional forms/min	**0.59**	0.61	0–2.00	**1.68**	0.88	0.20–3.24	**193.5**	**< 0.001**
Word combinations/min	**0.45**	0.76	0–2.33	**1.71**	1.64	0–5.93	**182.0**	**0.003**
Word types/min	**1.15**	1.18	0–3.60	**4.07**	1.60	1.03–7.80	**210.5**	**< 0.001**
Word tokens/min	**3.74**	4.04	8–11.60	**13.33**	6.57	3.38–26.07	**205.0**	**< 0.001**
MLU	**1.25**	0.20	1–1.62	**1.77**	0.48	1.12–2.70	**186.0**	**< 0.001**

Note: MLU, mean length of utterance.

Table 2. Gestural productions of the children in the two groups during the observation session

	SCT			TD				
	Mean	SD	Range	Mean	SD	Range	*U*	*p*
Total gestures/min	1.82	1.34	0.47–4.47	1.02	0.51	0.15–2.20	77.5	0.148
Pointing/min	**0.87**	0.60	0.07–2.60	**0.40**	0.24	0–0.76	**40.0**	**0.002**
Showing/min	0.15	0.16	0–.60	0.25	0.21	0–0.60	146.5	0.161
Conventional/min	0.78	1.05	0–3.27	0.33	0.42	0–1.47	88.5	0.325
Iconic/min	0.01	0.04	0–0.13	0.04	0.05	0–0.13	149.5	0.126

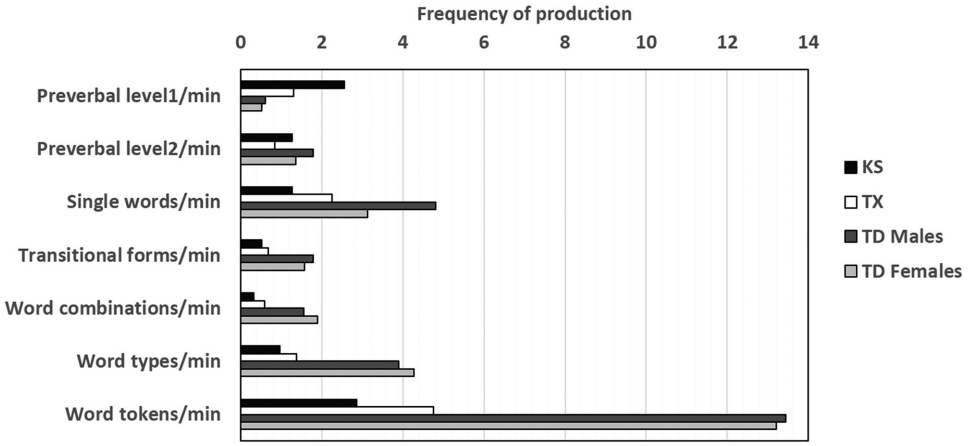

Figure 3. Vocal productions per minute of males and females in each group

lower than the 5th percentile of the normative Italian population. It must be underlined that being a late-talking child is not a diagnosis and does not necessarily imply that these children will develop a language impairment. In fact, there are different studies which found that many of these children catch up their initial delay (e.g., Dale *et al.* 2003) or found how other factors (e.g., socio-economic status and maternal factors) have a major role than early vocabulary size in explaining later language impairment (e.g., Reilly *et al.* 2010).

However, the identification of this risk factor for language impairment in children with SCT emphasizes the importance of directly observing and analysing their spontaneous communicative productions in their natural environment, such as during parent–child interactions. With regards to vocal communicative productions, the children with SCT showed a lower percentage of verbal utterances (i.e., utterances containing at least one word) than TD children matched for chronological age. In addition, single words, transitional forms and word combinations were produced with a significantly lower frequency and a significantly lower variety (e.g., word types) and complexity (e.g., MLU) by children with SCT.

However, note that children with SCT attempted to compensate for their verbal difficulties using a higher number of simple vocal productions (i.e., preverbal level 1) and a higher number of pointing gestures. In particular, the high number of communicative gestures can be considered a protective factor, since children's use of gestures appeared to be associated with their later vocabulary development in different populations (e.g., Thal *et al.* 1991 in late-talking children; Suttora and Salerni 2012 in preterm born children; and Zampini and D'Odorico 2009 in children with Down syndrome). Therefore, despite their language difficulties, children with SCT were able to communicate with their partner and did not show problems in using preverbal intentional communicative behaviours. The presence or absence of compensatory strategies to communicate, such as preverbal productions or communicative gestures, is a fundamental factor in predicting possible interaction problems, especially considering that children with KS are at risk not only for language impairment but also for ASD (Bishop *et al.* 2011). An early analysis of children's communicative skills, which considers not only parental reports but also direct observations of children's non-verbal behaviours, could help distinguish between different developmental trajectories (i.e., children with a specific language delay compared with children in which a language delay is a symptom of ASD).

With regards to the specific communicative profiles of children with KS and TX, boys with KS appeared to be less competent than girls with TX in their linguistic skills. Although no statistically significant differences emerged between the spontaneous communicative

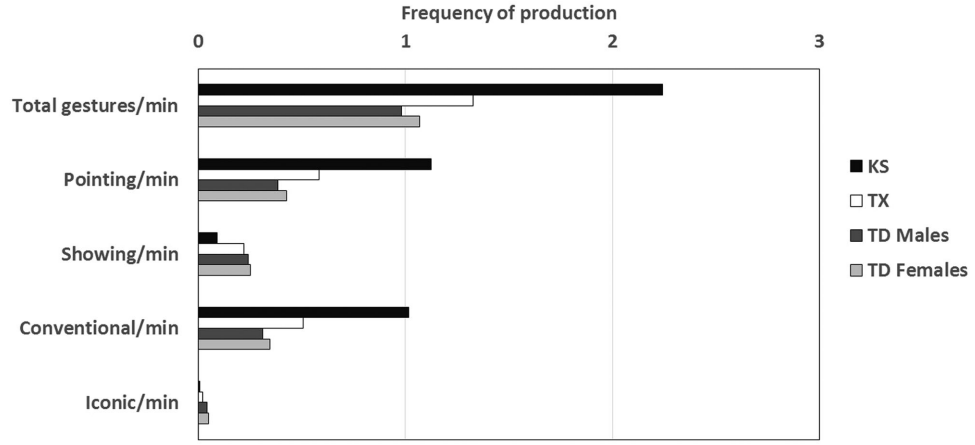

Figure 4. Gestural productions per minute of males and females in each group

Table 3. Comparison of vocal and gestural productions between males and females in the SCT and TD groups

	Boys with KS versus TD males		Girls with TX versus TD females	
	U	*p*	*U*	*p*
Preverbal level 1/min	**5**$^{(KS>TD)}$	**0.003**	7	0.026
Preverbal level 2/min	40.5	0.328	34	0.259
Single words/min	**57**$^{(KS<TD)}$	**0.007**	35	0.209
Transitional forms/min	55	0.015	41	0.038
Word combinations/min	52.5	0.028	39	0.073
Word types/min	**59**$^{(KS<TD)}$	**0.003**	**48**$^{(TX<TD)}$	**0.001**
Word tokens/min	**60**$^{(KS<TD)}$	**0.002**	43	0.017
MLU	45	0.054	**48**$^{(TX<TD)}$	**0.001**
Total gestures/min	18.5	0.161	20	0.620
Pointing/min	**5.5**$^{(KS>TD)}$	**0.003**	14	0.209
Showing/min	43.5	0.234	28.5	0.620
Conventional/min	27	0.645	16.5	0.318
Iconic/min	41	0.382	33.5	0.259

Note: MLU, mean length of utterance.

productions of these two groups of children, the boys with KS appeared to differ from TD males in more aspects of communication than the girls with TX differed from TD females. In particular, males with KS differed from TD males in both vocal production, showing lower verbal competence, and gestural production, showing greater use of gestures to compensate for their linguistic problems. With regards to the girls' spontaneous communicative productions, the children with TX appeared to be less competent than TD girls in some vocal aspects (i.e., they showed a lower number of word types and a shorter MLU), but they did not differ in the frequency of production of single words and word combinations. In addition, no significant differences were found in the communicative gestural productions of the girls in the two groups, which was likely because the girls with TX did not need to compensate for their verbal problems as much as the boys with KS did.

The present study showed the existence of a delay in the first communicative skills of children with SCT in terms of vocabulary size and production of verbal utterances. The importance of early detection of language risk factors (e.g., low vocabulary size or lack in gesture production) lies in the possibility of early intervention and careful and continuous monitoring of children's communicative development. As found in studies on late-talking children, providing early intervention, especially through parent training, is important to help them become successful communicators (e.g., Girolametto *et al.* 1996, Mahoney *et al.* 1999). For instance, empowering parents to become their children's language facilitator by using more effective linguistic input can maximize opportunities for children's communicative development in everyday situations (e.g., the programme 'It Takes Two to Talk'; Girolametto, Weitzman, McCauley and Fey 2006).

Limits and future directions

The direct observation of children's spontaneous communicative competence in a population rarely studied with observation techniques is a strength of the study. However, the small number of participants is an important limitation. The number of children involved in the study, in addition to high individual variability in language development during the second year of life, do not allow for the generalization of the results to the entire population of children with SCT.

Future studies should analyse other possible early predictors of language impairment, for instance language comprehension or joint attention skills, to understand which factors allow to early identify children at risk for language impairment. In addition, future

studies should longitudinally analyse the communicative development of children with SCT to identify the percentage of children who actually develop language impairment and verify if those children who use communicative compensatory strategies (e.g., gestural production) have reduced risk of language impairment. In addition, future studies should involve younger children with SCT to identify predictive indexes in an earlier stage of development and maximize the effects of early intervention.

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