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Cognitive functions in Arabic-speaking children with velopharyngeal insufficiency and their impact on speech intelligibility

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Abstract

Background Patients with orofacial cleft are at high risk for neurobehavioral problems including learning disability, impaired language function, psychosocial adjustment issues, and persistently reduced academic achievement. All these factors may be related to decrease intellectual abilities of those patients. The presence of velopharyngeal insufficiency (VPI) leads to affection of speech intelligibility due to atypical consonant productions, abnormal nasal resonance, nasal air emission, compensatory articulatory mechanism, and facial grimace.

Objective This study aimed at assessing the cognitive functions of patients with (VPI) and their effect on speech intelligibility.

Methodology Fifty patients with (VPI) were selected from the Outpatient Clinic of the Phoniatic Unit in Assiut University Hospital. All patients were evaluated by protocol of nasality assessment including auditory perceptual assessment of speech, assessment of overall intelligibility of speech, nasoendoscopy, and psychometric evaluation.

Results The mean intelligence quotient (IQ) of patients with VPI was 75.2 ± 14.5 with a range between 41 and 107. The main defect was present in quantitative potential and then verbal ability followed by visual ability with memory having the highest mean. Patients with repaired cleft palate had the highest score (86.53 ± 9.96), while the least score was reported among those with velopharyngeal disproportion (72.50 ± 9.59). There was a nonsignificant negative correlation between IQ degree and speech unintelligibility ($p=0.82$).

Conclusion About half of the patients with (VPI) have below average mentality. Patients with repaired cleft palate had the highest (IQ) score. Increased (IQ) score was accompanied by decreased speech unintelligibility, although it does not reach the level of significance.

Keywords VPI, Cleft palate, Speech intelligibility, Cognitive dysfunction

Background

Velopharyngeal insufficiency (VPI) refers to structural defects which result in a gap in the velopharyngeal valve. This is often because the velum is short relative to the position of the posterior pharyngeal wall.

Velopharyngeal insufficiency has many causes, including cleft palate, submucous cleft, velopharyngeal disproportion, and acquired defects following various surgical procedures (after adenoidectomy, oral and pharyngeal tumors removal) [1]. VPI implies the presence of hypernasality, inappropriate nasal escape, and decreased air pressure during the production of oral speech sounds with decreased speech intelligibility [2]. The term intelligibility refers to “speech clarity” or the proportion of a speaker’s output that a listener can readily understand. Reduced speech intelligibility leads to misunderstanding,

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frustration, and loss of interest by communication partners. As a result, communication decreases or remains at a low level [3]. Patients with mild and moderate intellectual disabilities showed distinct difficulties in their speech production that affect both the quality and intelligibility of their verbal output. Their speech is characterized by an overall high error rate and the occurrence of both typical and atypical phonological processes [4].

Children with cleft palate performed more poorly on cognitive-intellectual measures than their peers [4]. A number of studies suggest that the cognitive deficits may be secondary to linguistic deficits. They found that children with cleft palate have lower scores on verbal IQ measures than on performance measures [5–7].

Some studies have tried to identify specific patterns of cognitive difficulty in children with clefts. They found problems with visual perceptual skills [7–9], while others have shown no deficit in these skills [10, 11]. This study aimed to evaluate the cognitive functions of Arabic-speaking children with velopharyngeal insufficiency and their impact on speech intelligibility.

Methods

Fifty patients aged 3–27 years presenting with velopharyngeal insufficiency due to repaired/unrepaired cleft palate or cleft lip and palate or due to velopharyngeal disproportion were recruited from the Outpatient Clinic of the Phoniatric Unit at Assiut University Hospital during the period from April 2019 to April 2020. They were free from syndromic cleft palate, hearing impairment (sensorineural hearing loss), neurological diseases, attention-deficit hyperactivity disorder (ADHD), delayed language development, and no history of speech therapy. All patients were evaluated by the following protocol of nasality assessment:

- A. *Patient interview*: Data collected from the parents were parent's education and job, consanguinity, age of the child, analysis of complaint, developmental milestones, illness of early childhood, operative intervention and its effect on regurgitation and speech, previous speech therapy, and subjective impression of the following: hearing, swallowing, mental ability, and scholastic achievement.
- B. *Auditory perceptual assessment (APA) of speech*: It was done by three expert phoniatricians after recording the participant's speech. Speech sample includes reading a standardized text or counting to assess the type of nasality (open, closed or mixed), degree of nasality (mild, moderate, severe), consonant precision, the compensatory articulatory mechanisms (glottal articulation and pharyngealization of fricatives), facial grimace, audible nasal air escape, and

overall intelligibility of speech. All these parameters were graded along a 5-point scale in which (0) = normal and (4) = severe affection. Speech intelligibility was assessed according to Subtenly et al. [12]. The rating scale was composed of 5 degrees: (0) = normal for age and sex, (1) = mild difficulty in understanding-repetition not required, (2) = moderate difficulty-repetition required infrequently, (3) = marked difficulty-repetition required frequently, and (4) = unintelligible with repetition.

- C. *Visual assessment of the vocal tract*: This includes examining lips, dentition, bite, alveolus, hard and soft palate (if clefted, fistula, scar, and palatal length), and lateral and posterior pharyngeal walls, size of tonsils, size and movement of the tongue, and also examination of nose, ear, and larynx.
- D. *Simple clinical tests*: Gutzman's [a/i] test [13] and Czermak's [cold mirror] test [14] were performed.
- E. *Flexible nasoendoscopy*: (Storz Tele pack X LED-TP100) to assess the movement of the velum, lateral and posterior pharyngeal walls, the movement of each component is given a score of (0–4): (0) = the resting (breathing) position, (2) = half the distance to the corresponding wall, and (4) = the maximum movement reaching and touching the opposite wall. Also, the pattern of closure of the velopharyngeal port, whether coronal, sagittal, circular, or other, the velopharyngeal gap and its size, the presence of adenoid and its size, and the presence of Passavant's ridge were assessed.
- F. *Language evaluation*: By Arabic language test [15] and articulation test [16].
- G. *Psychometric evaluation*: By Stanford Binet intelligence quotient 4th edition with its four subtests assessing (verbal ability, visual ability, quantitative potential, and memory) [17].
- H. *Audiologic assessment*
- I. *Nasometry*: Nasometer 6200 (Kay Elemetrics/PEN-TAX) was used to measure nasal resonance.

Statistical analysis

Data analysis has been performed using SPSS model 20 IBM SPSS (IBM Corp., Armonk, New York, USA). Categorical data have been offered such as number and percent. Quantitative data with normal distribution are expressed as mean \pm standard deviation and range. Student *t*-test was used to compare quantitative data of two groups, while in the case of more than two groups, ANOVA was used. Quantitative data with abnormal distribution expressed as median (minimum–maximum) and compared by Mann–Whitney *U*-test was used. The correlation tests were conducted using Spearman's

correlation coefficient to correlate between abnormally distributed data and different parameters. The statistical differences were considered significant when *P* was lower than 0.05.

Results

Demographic data of studied patients (*n* = 50)

The mean age (\pm SD) of enrolled patients was 11.50 ± 4.95 years with a range between 3 and 27 years. Thirty (60%) patients were females, and twenty (40%) were males. In terms of education, seven patients (14%) were preschoolers, 21 (42%) were primary school students, 13 (26%) were preparatory school students, 3 (6%) were secondary school students, 2 (4%) were postsecondary school students, and 4 (8%) were illiterate. Consanguinity was present in 33 patients (66%), and 17 patients (34%) had no consanguinity (Table 1).

Distribution of the patients according to the cause of VPI

As regards the cause of VPI, cleft palate only (CPO) was the most frequent type in 35 patients (70%), followed by velopharyngeal disproportion 7 (14%), bilateral cleft lip and palate (BCLP) were 5 (10%), and unilateral cleft lip and palate (UCLP) were 3 (6%) patients (Table 2).

Auditory perceptual assessment (APA) of patient's speech

Three patients had mixed nasality, while the other 47 (94%) patients had open nasality. As regards the degree of nasality, it was found that 28 (56%), 19 (38%), and 3 (6%) patients had slight, mild, and moderate degree, respectively. It was found that 14 (28%) patients had no consonant imprecision, while slight, mild, and moderate

Table 1 Demographic data of studied patients

Item	<i>N</i> = 50
Age (years)	11.50 ± 4.95
Range	3–27
Sex	
Male <i>N</i> (%)	20 (40%)
Female <i>N</i> (%)	30 (60%)
Educational level (<i>N</i> %)	
Preschool	7 (14%)
Primary school	21 (42%)
Preparatory school	13 (26%)
Secondary school	3 (6%)
Postsecondary school	2 (4%)
Illiterate	4 (8%)
Consanguinity (<i>N</i> %)	
None	17 (34%)
Present	33 (66%)

Data expressed as frequency (percentage). Test of significance: chi-square test

Table 2 Distribution of the patients according to the cause of VPI

VPI cause	<i>N</i> = 50	%
Velopharyngeal disproportion	7	14.0
Cleft palate only (CPO)	35	70.0
Unilateral cleft palate (UCLP)	3	6.0
Bilateral cleft lip and palate (BCLP)	5	10.0

Data expressed as frequency (percentage). Test of significance: chi-square test.
VPI velopharyngeal insufficiency

consonant imprecision was present in 15 (30%), 14 (28%), and 7 (14%) patients, respectively. Ten (20%) patients had glottal compensatory articulation, and 7 (14%) patients had pharyngeal articulation, while 9 (18%) patients had both types of articulation. Audible nasal air emission was absent in only 6 (12%) patients, while it was present in 44 (88%) patients. Facial grimace was detected in 19 (38%) patients. Regarding overall unintelligibility, 21 (42%), 10 (20%), 11 (22%), and 1 (2%) patients had slight, mild, moderate and severe unintelligibility, respectively (Table 3).

Degree of intelligence quotient and its subclasses among studied patients (*n* = 50)

The mean IQ (\pm SD) of all patients was 75.2 ± 14.5 with a range between 41 and 107. As regards IQ subclasses, it was found that 11 (22%), 26 (52%), 11 (22%), and 2 (4%) patients had average, below average, mild MR, and moderate MR, respectively (Fig. 1).

Subtypes of intelligence quotient among studied patients

The main defect was present in quantitative potential (76.04 ± 13.01) and then verbal ability (79.62 ± 12.11) followed by visual ability (79.88 ± 14.3) with memory having the highest mean (82.02 ± 12.73) (Table 4).

Distribution of subtypes of IQ, nasality degree, speech unintelligibility, and different causes of VPI

There was no significant difference between different causes of VPI and IQ degree. There was no significant difference between different causes of VPI and nasality degree. There was a moderate significant difference between causes of VPI and speech unintelligibility, as patients with BCLP had the highest grade of speech unintelligibility while patients with velopharyngeal disproportion had the least grade of speech unintelligibility (Table 5).

IQ degree among repaired and unrepaired cases of VPI

It was found that different causes of VPI had highly significant effect on IQ degree ($p < 0.001$). In general,

Table 3 Auditory perceptual assessment (APA) of patient’s speech

APA	N= 50
Nasality type	
Open	47 (94%)
Mixed	3 (6%)
Nasality degree	
Slight	28 (56%)
Mild	19 (38%)
Moderate	3 (6%)
Consonant imprecision	
None	14 (28%)
Slight	15 (30%)
Mild	14 (28%)
Moderate	7 (14%)
Compensatory articulation	
No	24 (48%)
Glottal	10 (20%)
Pharyngeal	7 (14%)
Both	9 (18%)
Audible nasal air escape	
Absent	6 (12%)
Present	44 (88%)
Facial grimace	
Present	19 (38%)
Absent	31 (62%)
Speech unintelligibility	
Normal	7 (14%)
Slight	21 (42%)
Mild	10 (20%)
Moderate	11 (22%)
Severe	1 (2%)

Data expressed as frequency (percentage). Test of significance: chi-square test

patients with repaired cleft palate had the highest score (86.53 ± 9.96), followed by unrepaired cleft palate, while the least score was reported among those with velopharyngeal disproportion (72.50 ± 9.59) (Fig. 2).

Distribution of IQ degree and speech unintelligibility

It was found that different grades of speech unintelligibility had no significant differences with IQ degree ($p=0.82$). But in general, patients with no speech unintelligibility had the highest IQ (86.19 ± 8.89), while the least value was reported among those with moderate unintelligibility (67.10 ± 9.54) (Table 6).

Correlation matrix between IQ degree, nasality degree, and speech unintelligibility

There was a significant positive correlation between the degree of nasality and speech unintelligibility ($p=0.04$). Increased nasality degree is accompanied by increased speech unintelligibility ($r=0.492$). There was a nonsignificant negative correlation ($p=0.82$) between IQ degree and speech unintelligibility. Also, there was a nonsignificant negative correlation between IQ and nasality degree ($p=0.845$) (Table 7).

Discussion

Cognitive dysfunction in children with clefts of the lip and palate has been documented for decades [18]. Some studies reported that children with non-syndromic clefts had lower IQs [19, 20] and lower scholastic achievement [20] than that of the general population. In this study, the mean IQ was (75.2 ± 14.5) with a range between 41 and 107. So, there is obvious downgrading in the cognitive function of patients with VPI. This agreed with Persson et al. [21] who found that the group with cleft palate alone had a significantly lower

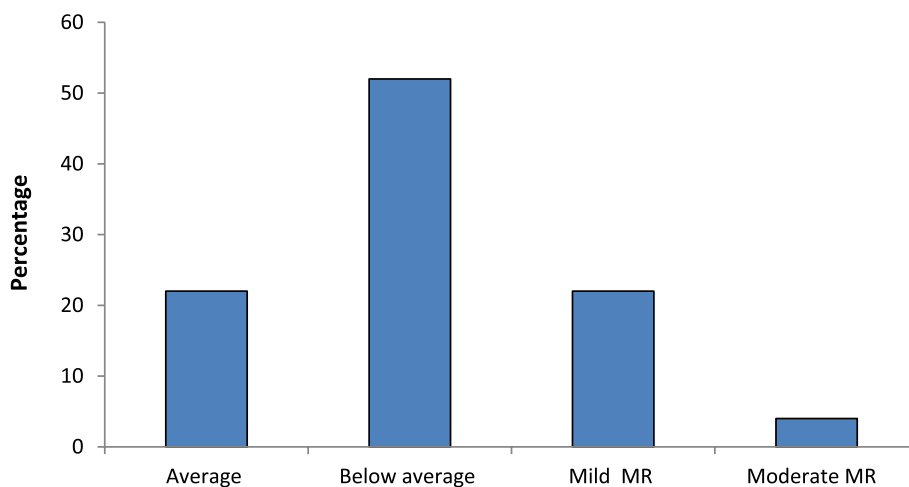


Fig. 1 Degree of total IQ and its subclasses among studied patients

Table 4 Subtypes of intelligence quotient among studied patients

	Total (n = 50) Mean ± SD	IQ subclass			
		Average (n = 11) Mean ± SD	Below average (n = 26) Mean ± SD	Mild MR (n = 11) Mean ± SD	Moderate MR (n = 2) Mean ± SD
		IQ degree	75.2 ± 14.5	93.18 ± 5.65	77.31 ± 5.65
Verbal ability	79.62 ± 12.11	94.55 ± 6.82	80.81 ± 5.92	65.45 ± 6.07	60 ± 2.83
Visual ability	79.88 ± 14.3	98 ± 8.91	80.46 ± 6.94	66.36 ± 4.65	47 ± 4.24
Quantitative potential	76.04 ± 13.01	90.73 ± 7.39	77.62 ± 8.33	62.18 ± 4.42	51 ± 4.24
Memory	82.02 ± 12.73	97 ± 7.28	83.15 ± 7.55	68.73 ± 6.21	58 ± 0

Data expressed as mean (SD). IQ intelligence quotient. Test of significance: chi-square test

Table 5 Distribution of subtypes of IQ, nasality degree, speech unintelligibility, and different causes of VPI

	Total (n = 50)	VPI causes				p-value
		Velopharyngeal disproportion (n = 7)	CPO (n = 35)	UCLP (n = 3)	BCLP (n = 5)	
IQ degree	75.2 ± 14.5	72.43 ± 8.75	75.74 ± 14.86	70.67 ± 25.93	78 ± 14.3	0.861
Verbal ability	79.62 ± 12.11	78.43 ± 6.78	79.26 ± 12.64	79.33 ± 18.9	84 ± 12.73	0.868
Visual ability	79.88 ± 14.3	78 ± 9.93	80.03 ± 14.89	74.67 ± 21.39	84.6 ± 13.74	0.795
Quantitative potential	76.04 ± 13.01	72 ± 9.17	76.17 ± 13.21	76.67 ± 20.03	80.4 ± 14.59	0.754
Memory	82.02 ± 12.73	83.14 ± 8.23	82.2 ± 13.7	77.33 ± 17.01	82 ± 11.14	0.931
Nasality degree						
Slight	27 (54%)	4 (57.1%)	18 (51.4%)	2 (66.7%)	3 (60%)	0.937
Mild	20 (40%)	2 (28.6%)	15 (42.9%)	1 (33.3%)	2 (40%)	
Moderate	3 (6%)	1 (14.3%)	2 (5.7%)	0 (0%)	0 (0%)	
Speech unintelligibility						
Normal	6 (12%)	1 (14.3%)	5 (14.3%)	0 (0%)	0 (0%)	0.005**
Slight	22 (44%)	4 (57.1)	16 (45.7%)	2 (66.7%)	0 (0%)	
Mild	10 (20%)	1 (14.3%)	8 (22.9%)	1 (33.3%)	0 (0%)	
Moderate	11 (22%)	0 (0%)	6 (17.1%)	0 (0%)	5 (100%)	
Severe	1 (2%)	1 (14.3%)	0 (0%)	0 (0%)	0 (0%)	

Test of significance: chi-square test, ** moderate significance

VPI velopharyngeal insufficiency, IQ intelligence quotient, CPO cleft palate only, UCLP unilateral cleft lip and palate, BCLP bilateral cleft lip and palate

score on the general intellectual capacity test than the control group. This may be explained by some research that found children and adults with cleft lip and palate (CLP) have abnormal brain structure and function. They have smaller brain volumes, with the frontal lobes and certain subcortical nuclei (caudate, putamen, and globus pallidus) being most affected [22]. The brain of adults with isolated cleft palate (ICP) showed normal cerebral volumes, but an abnormality in tissue distribution in which the frontal and parietal lobes were substantially increased in volume compared with normal, and the temporal and occipital lobes were significantly decreased in volume. The cerebellum was also decreased in volume [23]. These differences in brain

volume and structure may be related to the cognitive problems in people with orofacial clefts [24].

In this study, patients with UCLP had the least IQ score (70.67 ± 25.93) followed by patients with velopharyngeal disproportion (72.43 ± 8.75) and then patients with CPO (75.74 ± 14.86), while patients with BCLP had the highest IQ score (78 ± 14.3). This disagrees with Nopoulos et al. [25] who found a relationship between severity of clefting and severity of cognitive deficit in which subjects with bilateral CLP (most extensive clefting) were the most severely affected cognitively, while subjects with CPO (least extensive clefting) were the least affected. However, this may be due to the small number of subjects in each clefting group in this study.

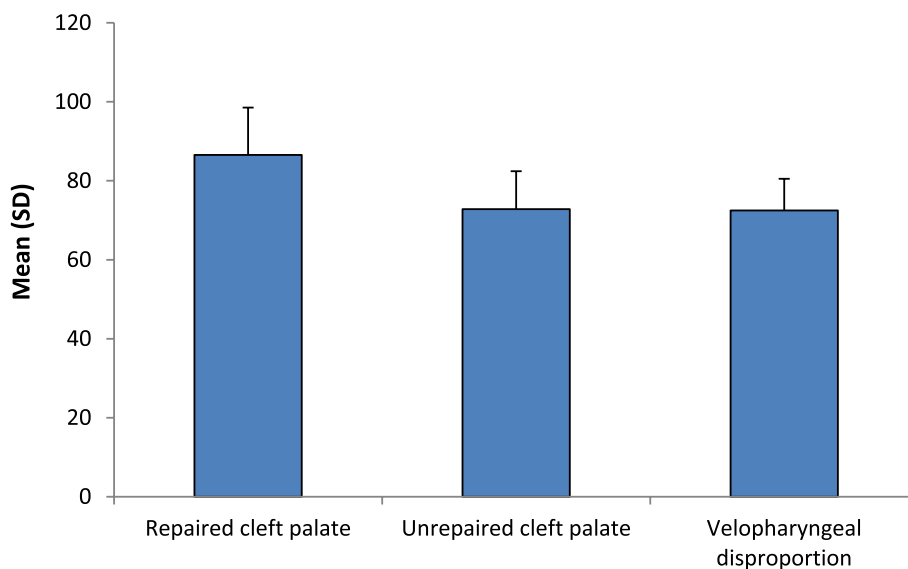


Fig. 2 IQ degree among repaired and unrepaired types of VPI

Table 6 Distribution of IQ degree and speech unintelligibility

Speech unintelligibility ^a	N	IQ mean ± SD
Normal N (%)	6 (12%)	86.19 ± 8.89
Slight N (%)	22 (44%)	74.45 ± 11.45
Mild N (%)	10 (20%)	73.57 ± 8.65
Moderate N (%)	11 (22%)	67.10 ± 9.54
p-value		0.82

Data expressed as mean (SD). p-value was significant if < 0.05

IQ intelligence quotient

^a Severe grade was not included where only one patient had severe unintelligibility. Test of significance: ANOVA test

Also, Nopoulos et al. [26] showed that adult males with non-syndromic cleft lip and/or palate have a specific pattern of cognitive deficits. Subjects with clefts were found to have general IQ scores below that of their matched controls. Subjects with clefts had specific and significant abnormalities in verbal abilities. On

the other hand, motor skills, verbal memory, executive function, and performance on a visuospatial task were not different from their matched control group.

Richman and Eliason stated that overall intellectual functioning is within the average range. However, there were specific cognitive deficits or delays in children with clefts. These deficits appear to affect the verbal abilities and visual-motor function, but the nonverbal and visual-perceptual functions are generally intact [27].

We found that patients with repaired cleft lip and palate had the highest IQ score. This may be attributed to better psychological consequences of early vs. later repair. That is in line with Murray et al. [28] who found lowered cognitive scores in infants having late cleft lip and palatal repair. As the disfigurement caused by unrepaired clefts not only makes these children less appealing to look at but also makes it difficult for parents to interpret infant expressions, early repair helps better face-to-face play and influences the quality of life of infants

Table 7 Correlation matrix between IQ degree, nasality degree, and speech unintelligibility

		IQ degree	Nasality degree	Speech unintelligibility
IQ degree	r-value	1.000		
	p	-		
Nasality degree	r-value	-0.134	1.000	
	p	0.845	-	
Speech unintelligibility	r-value	-0.053	0.492**	1.000
	p	0.829	0.04	-

p-value was significant if < 0.05. Test of significance: Pearson correlation, ** moderate significant

IQ intelligence quotient

as they might be accepted better by their families. Also, parents of infants having early cleft lip repair may find it easier to respond to infant social cues [28].

In our study, the different grades of speech unintelligibility had no significant differences with IQ degree. However, patients with no speech unintelligibility had the highest IQ, while the least value was reported among those with moderate unintelligibility. This may be explained by Coppens-Hofman et al. [4] who showed a strong association between the severity of intellectual dysfunction and speech intelligibility. Patients with intellectual disabilities have difficulties in their speech production that affect both the quality and intelligibility of their verbal output. Their speech is characterized by multiple phonological processes. As short-term and long-term verbal memories are both highly involved in speech production, the two systems are impaired in people with intellectual dysfunction. An additional factor to consider as a potential cause of reduced intelligibility is poor auditory feedback due to deficient auditory processing.

There was a significant positive correlation between nasality degree and speech unintelligibility. This may be explained by the fact that hypernasality affects vowel production and causes modification of the spectrum of F1 and F2 such as weakening of formants, decrease in the strength and enhanced bandwidth of F1 and F2, lower in the amplitude of F1 and F2, introduction of pole/zero pairs in the vicinity of F1, and shifts in the formant frequencies. These spectral modifications in the hypernasal speech will have an impact on the articulatory dynamics while producing vowels resulting in vowel centralization and in turn affecting speech intelligibility [29]. This finding agrees with Særvold et al. [30] who found that the presence of hypernasality and reduced intelligibility were clearly associated with speech in cleft palate patients. Children with speech difficulties appear to have higher risk of delayed phonological awareness development, associated literacy problems, and delays in the acquisition of reading skills [31].

Conclusion

Patients with VPI show mild overall cognitive deficit with particular deficit in quantitative potential. These deficits may be due to the same factors that underline the facial cleft-abnormal development. Speech intelligibility is affected by the degree of hypernasality, the degree of cognitive deficit, and the type of VPI. Early intervention for patients with cleft lip and palate prevents the deterioration of speech problem and helps better personality self-confidence and scholastic achievement.

Abbreviations

VPI	Velopharyngeal insufficiency
IQ	Intelligence quotient
ADHD	Attention-deficit hyperactivity disorder
APA	Auditory perceptual assessment
CPO	Cleft palate only
UCLP	Unilateral cleft lip and palate
BCLP	Bilateral cleft lip and palate
ICP	Isolated cleft palate

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Authors' contributions

ZA collects the data of all patients with cleft lip and palate regarding the history, clinical examination, and auditory perceptual assessment and also perform the clinical examination and nasofiberoptic examination. HA analyzed and interpreted the data regarding the auditory perceptual assessment and nasofiberoptic examination. EM interpreted all data of patients and performed revision of all section of the research. ZA was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Approval of the Ethics Committee of the Faculty of Medicine, Assiut University was obtained before initiating the study (IRB number 17100741). Consent to participate: Informed written consent to participate in this study was provided by all participants in the cases above 16 years old and parents /or legal guardians in the cases of children under 16 years old.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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