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A Prospective Search for Urinary Tract Abnormalities in Infants With Isolated Preauricular Tags

David Kohelet, MD, and Eliana Arbel, MD

ABSTRACT. *Objective.* To determine whether isolated preauricular tags are associated with urinary tract abnormalities.

Methodology. Seventy consecutive infants with isolated preauricular tags were examined by ultrasonography for urinary tract abnormalities on day 3 or 4 of life between January 1993 and August 1999, after parental consent and ethics approval. Karyotype analysis was conducted in all infants with urinary tract abnormalities. The study group was compared with a control group of 69 infants without preauricular tags hospitalized during the same period. The control group consisted of infants who underwent urinary tract ultrasonography as part of an investigation for persistent regurgitation and/or vomiting associated with cyanotic spells.

Results. Urinary tract abnormalities were detected in 6 infants with isolated preauricular tags (6/70; 8.6%). Types of anomalies were as follows: hydronephrosis in 5 cases and horseshoe kidney in 1 case. The causes of hydronephrosis were ureteropelvic junction obstruction in 3 cases and vesicoureteric reflux in 2 cases. None of the infants in the control group had such abnormalities. All infants with urinary tract abnormalities had normal chromosomes. No statistically significant differences existed between groups concerning birth weight, gestational age, intrauterine growth, and male-to-female ratio.

Conclusions. This study suggests a significant prevalence of urinary tract abnormalities in infants with preauricular tags. We recommend, therefore, that urinary tract ultrasonography be conducted in the routine assessment of infants with isolated preauricular tags. *Pediatrics* 2000;105(5). URL: <http://www.pediatrics.org/cgi/content/full/105/5/e61>; infant, preauricular tags, urinary tract anomalies, ultrasonography.

The association between ear malformations and renal anomalies was recognized as early as 1957.¹

Preauricular tags, protuberant fleshy papules located anterior to the tragus or just anterior to the crus of the helix, are commonly found in the neonatal period and are considered to be of minor clinical importance.² However, several studies have demonstrated that preauricular tags and pits may be associated with a higher incidence of conductive and/or sensorineural hearing impairment^{3,4} and urinary tract abnormalities.^{3,5,6} The cause of such associa-

tions, whether hereditary or not, has not been determined. The association between isolated preauricular tags and urinary tract abnormalities has not been thoroughly investigated.

We conducted a cross-sectional study designed to assess whether infants with isolated preauricular tags may be at risk for such abnormalities.

METHODS

All infants with preauricular tags born between January 1993 and August 1999 at Edith Wolfson Medical Center in Holon, Israel, were screened. An attending neonatologist examined all screened infants for additional congenital anomalies. Infants were excluded from the study if 1 or more of the following were present: 1) associated ear anomalies or other congenital anomalies; 2) a family history of renal anomalies in parents or siblings; and 3) parental refusal of informed consent. The investigational review board at our institution approved the study, and signed parental consent was obtained.

Ultrasonography of the urinary tract was performed on all eligible infants on day 3 or 4 of life. The same operator, who was unaware of the clinical course of each infant, examined all infants. When required, other imaging modalities (radionuclide scintigraphy and voiding cystography) were used subsequently at the pediatric follow-up clinic to provide accurate diagnosis and functional information. Voiding cystography was conducted in all infants to rule out vesicoureteric reflux, and radionuclide scintigraphy was performed to diagnose ureteropelvic junction obstruction and assess renal function.

Karyotype analysis was conducted in all infants with urinary tract abnormalities. The study group of 70 infants with isolated preauricular tags was compared with a control group of infants without preauricular tags. The control group consisted of 69 infants who underwent ultrasonography of the urinary tract after day 5 of life as part of an investigation for persistent regurgitation and/or vomiting associated with cyanotic spells. Infants from both groups were hospitalized at the same time period. Demographic and clinical data were collected for each infant in both groups (Table 1 and Table 2). Hydronephrosis was defined as pyelocaliectasis on renal ultrasonography. The degree of pyelocaliectasis was graded as 0 to 4 according to the classification of the Society for Fetal Urology.⁷

Comparisons between variables were conducted using the *t* test, for independent samples. A level of significance was set at $P < .05$. The χ^2 test was used to compare proportions between groups. Data are presented as the mean \pm the standard deviation.

RESULTS

During the study, 17 632 infants were born at our institution. Based on clinical indications and prenatally suspected findings, urinary tract abnormalities were detected by ultrasonography in 37 infants (37/17 632; .2%; 2/1000 births). Preauricular tags were detected in 83 infants. In 12 infants, preauricular tags were associated with ear anomalies as follows: atretic external canal ($n = 1$); accessory auricle ($n = 1$); dysplastic auricle ($n = 2$); and preauricular pits ($n = 12$). In 1 infant with an

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TABLE 1. Characteristics of Infants With Urinary Tract Abnormalities

Tag	Gender	Birth Weight (Grams)	Gestational Age (Weeks)	Ultrasonographic Findings	Final Diagnosis
1. Right	Female	3735	39	Bilateral grade 2 hydronephrosis	Bilateral grade 2 vesicoureteric reflux
2. Left	Male	2930	40	Grade 3 right hydronephrosis	Right ureteropelvic junction obstruction
3. Left	Female	3365	39	Grade 2 right hydronephrosis	Right ureteropelvic junction obstruction
4. Bilateral	Female	2640	39	Horseshoe kidney	Horseshoe kidney
5. Left	Male	3230	41	Grade 2 left hydronephrosis	Left ureteropelvic junction obstruction
6. Bilateral	Female	2805	38	Grade 3 right hydroureteronephrosis	Grade 4 right vesicoureteric reflux

TABLE 2. Comparisons Between Characteristics of Infants With Preauricular Tags and Infants of a Control Group

Variable	Study Group (n = 70)	Control Group (n = 69)
Birth weight (g)	3383 ± 394	3215 ± 565
Gestational age (wk)	39.1 ± 1.7	39.1 ± 1.5
Gender (n)		
Female	33	33
Male	37	36
Urinary tract abnormality (n)	6	0*

* $P < .02$.

isolated preauricular tag, there was a family history of polycystic kidney disease. The study group thus totaled 70 infants with isolated preauricular tags. None of the infants with isolated preauricular tags had family members with such malformations. The overall incidence of isolated preauricular tags in our population was 4.0/1000 (71/17 632). Thirty-six isolated preauricular tags were left-sided (51%), 23 were right-sided (33%), and 11 were bilateral (16%). Characteristics of infants with urinary tract abnormalities detected by ultrasonography are demonstrated in Table 1. Urinary tract ultrasonography and other imaging modalities disclosed that 6 of 70 infants had abnormalities, 1 infant had bilateral grade 2 hydronephrosis secondary to grade 2 bilateral vesicoureteric reflux, 2 infants had grade 2 right-sided hydronephrosis secondary to ureteropelvic junction obstruction, 1 infant had grade 3 right-sided hydroureteronephrosis secondary to grade 4 bilateral vesicoureteric reflux, 1 infant had a horseshoe kidney, and 1 infant had grade 2 left-sided hydronephrosis secondary to ureteropelvic junction obstruction. No relationship was found between urinary tract abnormality and the side of preauricular tags. At recent evaluations, none of the infants had reached a grade of obstruction that warranted surgical intervention. Our study documented abnormalities in the urinary tract of 6 (8.6%) of the 70 infants with isolated preauricular tags. None of the infants in the control group had urinary tract abnormalities. All infants with urinary tract abnormalities had normal chromosomes. None of the infants in both groups evidenced other associated congenital anomalies (on clinical examination). Characteristics of infants with isolated preauricular tags and infants of the control group are shown in Table 2. The mean birth weight of the study group was 3383 ± 394 g, and the mean gestational age was 39.1 ± 1.7 weeks. The intrauterine growth of all infants in the study group was appropriate

and the female-to-male ratio was 1:1.2. No statistically significant differences between the 2 groups concerning birth weight, gestational age, and male-to-female ratio were detected. However, the difference between the incidence of urinary tract abnormalities was statistically significant ($P < .02$).

DISCUSSION

The major finding of this study was that preauricular tags are associated with urinary tract abnormalities. The incidence of isolated preauricular tags detected in the study population (4/1000) is consistent with other reports.^{3,4} This study documents a prevalence of 8.6% of urinary tract abnormalities in infants with isolated preauricular tags. The prevalence of such abnormalities in this group is higher compared with the prevalence in a control group of infants ($P < .02$). Using ultrasonography as an initial diagnostic tool, we defined the prevalence of hydronephrosis in infants with preauricular tag. However, because ultrasonography is not a reliable tool for diagnosing vesicoureteric reflux in the neonatal period, we may assume that even more infants with preauricular tags might have had reflux if voiding cystography had been performed, given that 2 infants had vesicoureteric reflux.

The ratio between the number of infants with urinary tract abnormalities born at our institution and the total number of births during the study is similar to the incidence of .28% detected by routine prenatal ultrasonographic screening of 11 986 pregnant women in Sweden.⁸

Melnick et al⁹ described an association between ear malformations (gross ear deformities), branchial anomalies, and renal dysplasia and coined the term "branchio-oto-renal syndrome." It has been recently demonstrated that this syndrome is caused by mutations in the EYA1 gene.¹⁰

Although several investigators have reported an association between ear and urinary tract abnormalities, the ear abnormalities consisted of gross ear deformities.^{1,11} The association between isolated preauricular tags and urinary tract abnormalities has not been supported by other studies. Kugelman et al,⁴ who conducted ultrasonography in 23 infants with preauricular tags and pits, did not find associated abnormalities of the urinary tract. Conceivably, the discrepancies with our findings could be attributed to the small sample size of their population. A family history of urinary tract abnormalities could not account for such differences, because none of the infants in our study had a positive family history for urinary tract abnormalities.

The association between isolated preauricular tags and urinary tract abnormalities is difficult to explain based on localized concomitant disorders of blood flow and/or cell migration. Gorlin et al¹² proposed to lump together several conditions including facioauriculo-vertebral syndrome, hemifacial micrognathia, oto-mandibular dysostosis, Goldenhar syndrome, the first branchial arch anomalies, and the first and second branchial arches anomalies, proposing the term "oculo-auriculo-vertebral spectrum." The pathogenesis of the above spectrum is unknown. However, the wide variety of malformations in the above spectrum, including pulmonary and renal abnormalities would suggest a disorder of blastogenesis.¹³ Findings of this study support the hypothesis that the association between minor ear abnormalities (namely preauricular tags) and urinary tract abnormalities could be an additional condition of the oculo-auriculo-vertebral spectrum. Therefore, we recommend that urinary tract ultrasonography be conducted on all infants with isolated preauricular tags.

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