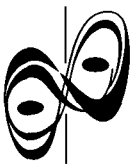

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Conjoined Twins in Hungary, 1970-1986

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Abstract. The total prevalence of conjoined twins (birth + prenatally diagnosed) was 1:68,000 in the study of 1970-1986. Symmetrical cases (the so-called siamese twins) have an obvious predominance (92.3%). Associated major malformations occurred in 80% of conjoined twins and more than 1/5 were discordant. The surviving time of liveborn conjoined twin sets was not more than two days except in two surgically successfully separated pairs. The family study did not indicate a higher recurrence risk. The case group was compared to two control groups and it appeared that the periconceptional use of oral contraception and ovulation induction were mentioned more frequently in pregnancies resulting in conjoined twins.

Key words: Conjoined twins, Population-based study, Prevalence, Epidemiology, Etiopathology

INTRODUCTION

The birth prevalence of conjoined twins shows a wide range in different countries (between 1:4,242 and 1:210,000, Table 1) due to objective (eg, racial) and technical (eg, ascertainment) differences. Similar differences have been published by the International Clearinghouse for Birth Defects System [15] (Table 2). Based on the mean birth prevalence of European countries, one to three conjoined twin births per year were expected in Hungary. However, four cases in 1983 and 1986, and five in 1974, 1976, 1977, 1982, were notified to the Hungarian Congenital Malformation Registry (HCMR). Furthermore, six conjoined twin sets were recorded in 1978. These higher figures prompted us to evaluate again the occurrence of conjoined twins in Hungary, 1970-1986, based on the HCMR. The data for the years 1970-1977 have been previously published [18].

Table 1 - Birth prevalence of conjoined twins

Reference	Location	Study period	Birth prevalence
Zake [30]	Uganda	1971-80	1:4,242
Mudalier [20]	India	1930[a]	1:6,250
Emanuel et al [13]	Taiwan	1965-68	1:6,500
Bland and Hammer [4]	Rhodesia	1950-62	1:14,000
Ryden [25]	Sweden	1934[a]	1:20,000
Center for Disease Control [7]	Atlanta, Georgia	1968-78	1:30,000
Rudolph et al [24]		1967[a]	1:31,000
Beischer and Fortune [1]	Australia	1936-66	1:40,000
Feldman [14]	England	1937[a]	1:50,000
Robertson [23]	World's Literature	1953[a]	1:50,000
Potter [22]	Chicago, Illinois	1931-61	1:50,000
Bhettey et al [3]	South Africa	1974-75	1:60,000
Källen and Rybo [16]	Sweden	1965-74	1:75,000
Edmonds and Layde [12]	USA	1970-77	1:97,000
Milham [19]	New York State	1945-65	1:166,000
Bender [2]	USA	1962-65	1:200,000
Stevenson et al [26]	24 countries	1961-64	1:210,000

[a]Year of report.

Table 2 - Number, rate per 100,000 births and birth prevalence of conjoined twins in the system of the International Clearinghouse

Program	Years	Number of conjoined twins			Number of births	Rate per 100,000 births	Birth prevalence[b]
		SA[a]	TP[a]	Births			
Denmark	83-87		1	4	240,750	1.66	1:60,000
New Zealand	82-86			4	252,560	0.58	1:63,000
Italy: IPIMC[c]	78-86			14	971,549	1.44	1:70,000
Czechoslovakia	61-85			54	3,903,936	1.38	1:72,000
Hungary	70-86		3	36	2,642,710	1.36	1:73,000
South America[d]	67-86			23	1,714,952	1.34	1:75,000
Italy: IMER[e]	78-86			2	150,168	1.33	1:75,000
Australia	82-86			16	1,198,756	1.33	1:75,000
Spain	78-87			7	665,856	1.05	1:95,000
France: Paris	83-85		5	1	120,749	0.83	1:120,000
Norway	67-85			9	1,097,650	0.82	1:120,000
England-Wales	78-85			36	5,107,543	0.70	1:140,000
Sweden	73-86	2	5	7	1,377,299	0.51	1:200,000
France: RAA[f]	76-86			3	777,912	0.39	1:260,000

[a]SA = spontaneous abortions; TP = terminated pregnancies.

[b]Refers only to conjoined twins born.

[c]IPIMC = Italian multicenter monitoring program, based on report from 147 hospitals.

[d]Data from hospitals in Argentina, Bolivia, Brazil, Chile, Columbia, Ecuador, Paraguay, Peru, Uruguay and Venezuela.

[e]IMER = Emilia Romagna, region of Italy.

[f]RAA = Rhone-Alps-Auverge, region of France.

The study period of 1970-1986 comprised 54 recorded cases of conjoined twins out of 2,642,710 total births. As far as we know, this is one of the largest population-based series of conjoined twins reported so far. The HCMR is based on the compulsory notification of malformed index patients diagnosed from the birth till the age of one year [5]. (Since 1985, the prenatally diagnosed and selectively terminated malformed fetuses have also been recorded). Notification is exclusively the task of obstetricians and neonatologists (in Hungary nearly all deliveries take place in hospital and the birth attendants are physicians), pediatricians (who treat malformed babies) and pathologists (autopsy is obligatory at infant age but not in stillbirths and abortions). The multiple sources of notification result in a considerable overlap, but duplications are eliminated through the use of personal record cards. Notification is practically complete: recently, the prevalence of all malformed index patients has approached 5% of total births and the ascertainment is nearly complete in major congenital abnormalities including conjoined twins. However, misdiagnoses show a wide range for the different types of congenital abnormalities. Another purpose of this study was to check-up the diagnoses of conjoined twins.

MATERIALS AND METHODS

The cases coded as 759.4 according to the International Classification of Diseases (ICD) were evaluated in the material of the HCMR, 1970-1986. The evaluation was completed by two special approaches. First, the detailed pathological and obstetrical documentations were requested officially from obstetrical in-patient clinics and pathology institutions. Photographs were available in 11/54 cases. Second, a case-control epidemiological study was organized for the evaluation of etiological factors.

Three types of controls were planned to match the 39 cases verified according to sex and birth date (year and month) of conjoined twins and residence of parents.

1) *Healthy singleton* controls were ascertained randomly from the control material of the Hungarian Case-Control Surveillance of Congenital Anomalies, 1980-1986 [8].
2) Unidentified *multimalformed singleton* controls were selected from the material of the HCMR, 1970-1986 [9].
3) *Healthy twin* controls were ascertained randomly from the monochorial monozygotic twin material of the Budapest Twin Registry, 1970-1986.

Data of multimalformed singleton controls were obtained by mailed questionnaires. Differences between conjoined twins and multimalformed singletons could be attributed to different methods of data collection. However, as compared to conjoined twins, about 60% of healthy singletons differed in birth year and more than 2/3 of healthy twins in the residence of parents. Each case of conjoined twins had three healthy and multimalformed singletons, and three healthy twin pairs. However, in two cases of conjoined twins the parents had moved to new unknown places, while in one case the family refused to cooperate. Thus, the data of 324 control mothers were compared with the data of 36 cases of conjoined twins. The

parents of all conjoined twins and their matched singleton and twin controls were visited at home and personal, family and pregnancy data were obtained by the questionnaire.

RESULTS

Of 54 recorded cases of conjoined twins, the notified diagnoses were confirmed only in 39 cases on the basis of obstetrical and pathology documentation. Of 15 notified cases, 4 were acardius amorphus. In 4 more cases, one twin had serious multiple malformations, while the other ones were macerated stillborns. "Multimalformation" or "monster" without specification were the notified diagnoses in both stillborn twins in 3 cases. Two unspecified "conjoined twins" occurred in a male-female and a dichorial stillborn twin pair, respectively. One twin pair was notified as being severely macerated without mention of abnormalities. Finally, a twin pair with feto-fetal transfusion was notified as conjoined twins. The proportion of misdiagnoses was 27.8%. After the exclusion of the 4 cases of acardius, wrongly coded into 759.4 instead of 759.8, the rate of misdiagnoses is 22.0% (11/50).

Eventually, therefore, 39 cases of conjoined twins were verified in Hungary, 1970-1986. Three cases were prenatally diagnosed at the 15th (triplets), 18th and 19th week of gestation, respectively, and they were terminated. Thus, the *birth* prevalence of the 36 cases of conjoined twins was 1:73,400 while the *total* (birth plus fetal) prevalence was 1:67,700. This figure was 1:55,200 in the years 1976-1983.

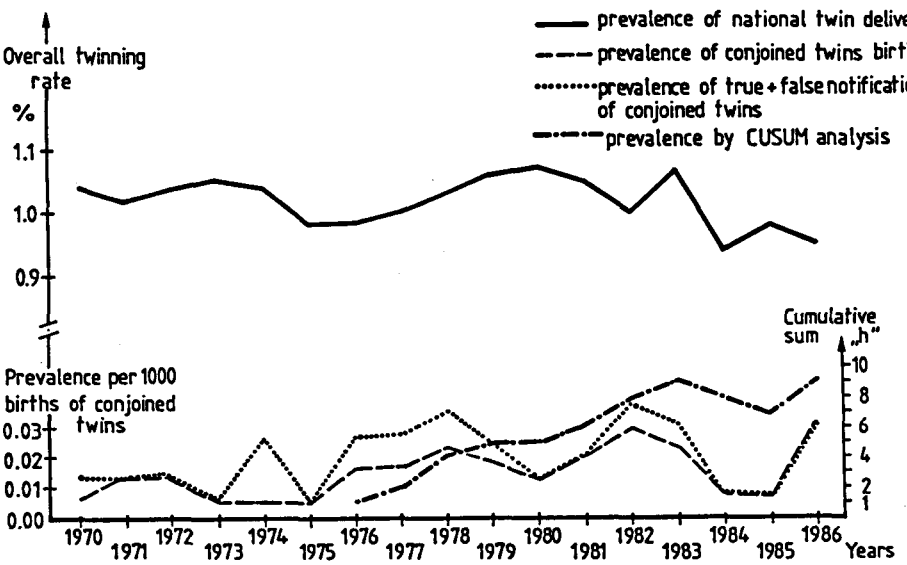

Fig. 1. Yearly distribution of conjoined twins and the overall twinning rate, 1970-1986.

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The *annual birth prevalence* of conjoined twins in relation to national twinning rate in 1970-86 is shown in Fig. 1. There was no correlation between the two frequency distributions ($r = -0.02$). The total prevalence of conjoined twins in this period was 14.76 per million births. However, it cannot be excluded that the completeness of the HCMR was lower in the early seventies. Nevertheless, the annual variation was not statistically significant ($\chi^2_{16} = 9.40$; $P > 0.05$). In the second step, the prevalence of conjoined twins was evaluated by CUSUM analysis. The period 1970-1975 was considered as a baseline figure. The cumulative sums show a significant rise from 1981 in contrast to the relatively small rise in mean prevalence.

Hungary is divided into 20 administrative units: 19 counties and Budapest, the capital. The *territorial distribution* of conjoined twins shows no significant differences within Hungary ($\chi^2_{19} = 1.636$; $P > 0.05$).

Table 3 - Distribution of conjoined twin types in Hungary

Types		No. of cases		%
I.	*Terata catadidymus* duplication in anterior or joined in posterior portion of body	7		17.9
	A. *Dicephalus* two distinct heads, one body		6	15.4
	B. *Pygopagus* joined by lateral and posterior surface of coccyx and sacrum-back-to-back		1	2.6
II.	*Terata anadidymus* duplication in posterior or joined by anterior portion of body	4		10.3
	A. *Syncephalus* single head, two bodies separate from umbilicus		3	7.7
	B. *Craniopagus* joined at cranial vault, two bodies separate except at head		1	2.6
III.	*Terata anacatadidymus* duplication joined at midpoint	23		59.0
	A. *Thoracopagus* joined at thoracic wall		14	35.9
	B. *Omphalopagus* cartilage		2	5.1
	C. *Xyphopagus* joined at xiphoid process		4	10.3
	D. *Thoracoomphalopagus* joined at thorax and abdomen		3	7.7
IV.	*Other types*	5		12.8
	A. *Parasite*		3	7.7
	B. *Combination*		2	5.1
	C. *Not stated*			
Total		39		100.0

Table 4 - Data of conjoined twins evaluated in Hungary, 1970-1986

Case no.	Type of conjoined twins	Year of birth	Sex		Congenital anomalies	Pregnancy outcome	Birth weight (A + B)	Gestation week
1.	Thoracopagus	1970	F	A + B	three arms, two hands on the common arm, common thumb with separate 4-4 fingers	SB	4800	40.
2.	Thoracopagus	1971	F	A	cleft lip and palate	SB	6000	40.
				B	—			
3.	Craniopagus	1971	M	A	absence of right hand	LB-ID	1200	24.
				B	absence of left forearm, absence of third finger of right hand, "sirenomelia"			
4.	Dicephalus	1972	M	A + B	anal atresia	LB-ID	3260	38.
5.	Syncephalus	1972	M	A	Eisenmenger complex in the heart	SB	2800	32.
				B	—			
6.	Thoracopagus	1973	F	A	double vagina	SB	3250	32.
				B	esophageal atresia with T-E fistula, duodenal atresia, doubling of uterus, polycystic kidney			
7.	Xyphopagus	1974	F	A	—	SB	3800	39.
				B	—			
8.	Parasite	1975	M	A	*exomphalos* ("schisis"), encephalocele, cleft lip and palate	SB	4300	40.
				B	—			
9.	Dicephalus	1976	F	A	cor biloculare (accessory pancreas)	LB-ID	3270	37.
				B	—			
10.	Syncephalus	1976	M	A	—	LB-ID	2100	28.
				B	—			
11.	Syncephalus	1976	M	A	—	SB	1000	32.
				B	—			
12.	Combination	1977	M	A	*exomphalos*, ventricular septal defect, polycystic kidney, left	SB	2750	28.
				B	hydrocephalus, polycistic kidney (hydrothorax, hydrops fetalis)			

(continued)

Table 4 - *Continued*

Case no.	Type of conjoined twins	Year of birth	Sex		Congenital anomalies	Pregnancy outcome	Birth weight (A + B)	Gestation week
13.	Xyphopagus	1977	F	A	cerv. and sacr. spina bifida occulta, pes varus (mallobulation of spleens)	SB	2800	33.
				B	—			
14.	Pygopagus	1977	M	A	atrial and ventricular septal defect, hermaphroditism	LB-ID	4650	38.
				B	cor uniloculare, anorectal stenosis, hermaphroditism (displaced kidneys)			
15.	Dicephalus (incomplete)	1978	F	A + B	polycystic kidneys	LB-ID	3500	
16.	Thoracopagus	1978	F	A + B	*cor commune triloculare biventriculare,* (intestinal eventeration)	LB-ID	3180	38.
17.	Parasite	1978	M	A	—	SB	1000	28.
				B	—			
18.	Thoracopagus	1978	F	A	diaphragmatic defect, pharyngeal and lingual agenesis, esophageal atresia, atrial and ventricular septal defect	LB-ID	1850	32.
				B	esophageal atresia, duodenal and intestinal agenesis			
19.	Dicephalus	1979	F	A + B	*cor commune, partial*	LB-ID	3230	37.
20.	Thoracopagus	1979	F	A + B	*common liver*	SB	2500	34.
21.	Thoracopagus	1979	F	A + B	*cor commune, partial; common liver, common thymus*	SB	5400	38.
22.	Parasite	1980	M	A	*exomphalos*, thoracal scoliosis, deformation of thorax, right	SB	2500	36.
				B	*exomphalos*, ventricular septal defect, hypoplasia of lungs, amelia, esophageal atresia			
				A + B	*common artery trunk*			

(continued)

Table 4 - *Continued*

Case no.	Type of conjoined twins	Year of birth	Sex		Congenital anomalies	Pregnancy outcome	Birth weight (A + B)	Gestation week
23.	Omphalopagus	1980	F	A + B	(six blood-vessels in the umbilical cord)	SB	6500	38.
24.	Combination	1981	M	A + B	heart and aortic defects (monoomphal type of umbilical cord), *common thymus, common intestinal system, common spleen, common scrotum*, pes equinovarus bilateral	LB-ID	4100	38.
25.	Thoracopagus	1981	M	A	four fingers on the right hand, rudimentary right ear, *common* (horseshoe) *kidney*, esophageal atresia, complex heart defect	SB	1725	30.
				B	*exomphalos*, three spleens			
26.	Xyphopagus	1981	F	A + B	*cor communis* and *complex heart defect duodenum commune, partial*	SB	2800	31.
27.	Thoraco-omphalopagus	1982	F	A + B	complex heart defect, intestinal atresia, cleft lip and palate	LB-ID	3600	39.
28.	Thoraco-omphalopagus	1982	F	A + B	anomalies of digestive system, musculoscetal malformations	LB-ID	1300	29.
29.	Thoracopagus	1982	F	A + B	cleft lip and palate, *common ventricule*, syndactyly and reduction anomalies of hands	LB-ID	5100	39.
30.	Dicephalus (dipus tetra-brachius)	1982	M	A	—	SB	1900	28.
				B	—			
31.	Xyphopagus	1983	M	A + B	Intestinal atresia, genito-urinary malformations (*common cloaca, 1 penis* with epispadias, 2 scrotums with 1-1 testicle)	LB-SS	5550	40.
32.	Omphalopagus	1983	F	A	*exomphalos*, displasia of hip	LB-SS	4050	33.
				B	*exomphalos*, displasia of hip, clubfoot, (cranial deformation)			

(continued)

Table 4 - *Continued*

Case no.	Type of conjoined twins	Year of birth	Sex		Congenital anomalies	Pregnancy outcome	Birth weight (A + B)	Gestation week
33.	Thoracopagus	1983	F	A B	— —	IA	190	20.
34.	Thoracopagus	1984	F	A + B	cor triloculare biventriculare, *common truncus*, agenesis of lung lobe, *hepar commune*	SB	3150	40.
35.	Thoraco-omphalopagus	1985	F	A + B	*agenesia of sternum, common liver* and *common intestinal system*	IA	570	22.
36.	Dicephalus	1986	F	A +B	cor triloculare biventriculare with ventricular septal defect	SB	4100	40.
37.	Thoracopagus (triplet pregnancy)	1986	F	B + C	cor triloculare biventriculare with ventricular septal defect, pulmonary stenosis, malformation of vena cava inferior and vena portae, (umbilical hernia)	SB	950	25.
38.	Thoracopagus	1986	M	A B	— —	SB	3500	38.
39.	Thoracopagus (triplet pregnancy)	1986	F	A B	— —	IA	90	16.

M = male; F = female; SB = stillbirth; LB = livebirth; ID = infant death; SS = separated by surgery; IA = induced abortion: terminated.

The *types of conjoined twins* are presented i n Table 3. Symmetrical cases (the so-called siamese twins) have an obvious predominance (92.3%). More than 1/3 of all cases (14/39) are thoracopagus twins. The following most common types were dicephalus (6 cases) and xiphopagus (4 cases). Two sets had various combinations of the other categories. Three asymmetrical (parasite) cases were recorded (birth prevalence, 1:850,000).

The *distribution of associated congenital anomalies* is shown in Table 4. Of the 39 cases of conjoined twins, 31 had major congenital anomalies (79.5%). This occurrence is much higher than usual (5%), though a high proportion of these anomalies is or may be connected with conjoined twinning. However, it is not easy to say whether these anomalies were associated or not with site of conjoining. That was not the case in exomphalos, common liver, common thymus, common heart and great vessels, common intestinal system. These anomalies, without other defects, occurred in 5 cases (italics in Table 4). The above-mentioned anomalies and minor anomalies (in brackets) were excluded from the analysis of observed vs expected rates (Table 5). One conjoined twin pair (No. 23) was affected only by a minor anomaly. Only one conjoined twin was affected by one congenital anomaly in 5 pairs. Both twins were multimalformed (two or more anomalies) in 9 pairs, while one twin was multimalformed in 6 pairs. The observed figures of intestinal atresias (27.0 ×), esophageal atresia (22.7 ×), anal atresia (13.7 ×), limb reduction deficiency (11.6 ×), anomalies of respiratory system (8.2 ×), cleft lip and palate (3.2 ×) and congenital cardiovascular malformations (1.8 ×), exceed significantly the expected figures based on the national figures [10].

Table 5 - The observed and expected proportion of congenital anomalies (CAs) in conjoined twins

Congenital anomalies (CAs)		Observed		Expected
		N	%	%
740-742.	CAs of nervous system	2	2.7	3.6
743.	CAs of eye	0	0.0	0.5
744.	CAs of ear, face and neck	1	1.3	0.8
745-747.	CAs of heart and vessels	18	24.3	13.2
748.	CAs of respiratory system	3	4.1	0.5
749.	Cleft palate and cleft lip	6	8.1	2.5
750-751.	CAs of digestive system			
	Anal atresia	3	4.1	0.3
	Esophageal atresia	5	6.8	0.3
	Intestinal atresia	6	8.1	0.3
	Other	3	4.1	3.8
752.	CAs of genital organs	6	8.1	12.5
753.	CAs of urinary system	5	6.8	2.7
754-756.	CAs of musculoskeletal system	7	9.5	51.5
755.2-4	Limb deficiency	8	10.7	0.7
757-759.	Other CAs	1	1.3	6.8
Total		74	100.0	100.0

A reverse trend was found in musculoskeletal system and urinary system due to the rare diagnosis of congenital dislocation of the hip and undescended testis in conjoined twins.

In the majority of cases the anomalies were concordant or half concordant (ie, partly common occurrence of congenital anomalies in twins) but the discordance was obvious in 7 cases (7/31 = 22.6%).

The *sex ratio* shows a female predominance 15:24 = 0.385, but the difference is not significant ($\chi_1^2 = 2.57$; $P > 0.05$). That means that the occurrence of conjoined twins was 18.69 per million births for females vs 11.05 per million births for males.

Of 36 births, 21 were *stillborn* (59.3%). The surviving time of liveborn sets was not more than two days, except in two surgically successfully separated pairs (Fig. 2).

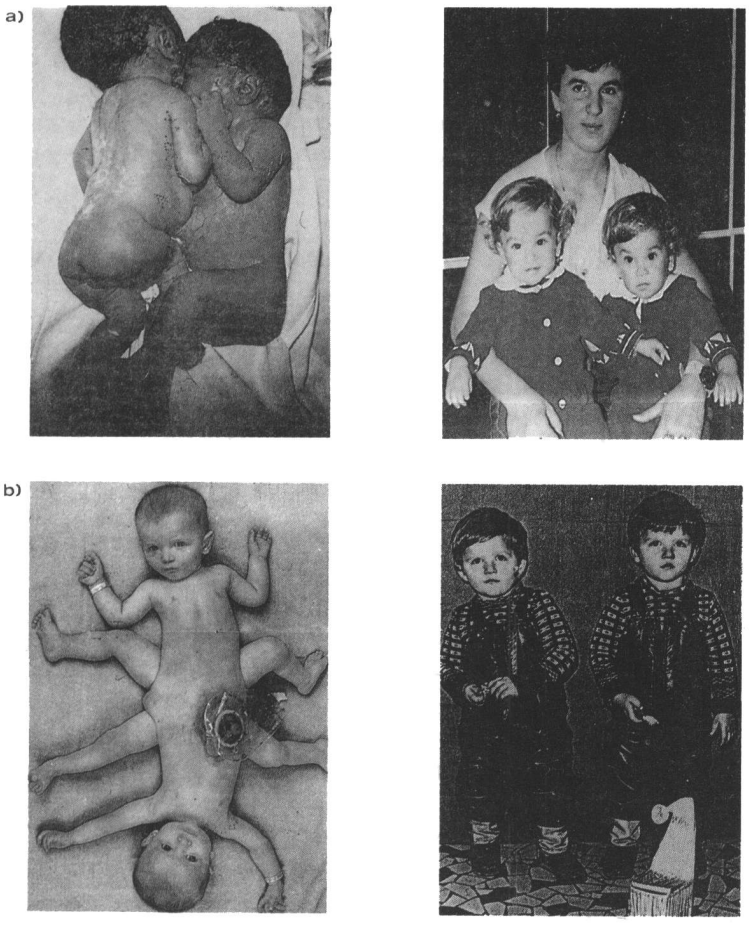

Fig. 2. Successfully separated omphalopagus (Case 32, a) and xyphopagus (Case 31, b) conjoined twins before and after operation.

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The average *birth weight* of conjoined twin pairs was 3260 g (without terminated cases). Their separated mean weight was smaller than that of twins (2085 g). The mean gestational time was also significantly shorter (34.7 wk) in comparison with twins (37.0 wk).

Of the 36 cases of conjoined twins, 11 (30.6%) were delivered by *cesarean section*. The means and distributions of maternal and paternal age, birth and pregnancy orders, did not differ significantly from the values of three matched control groups.

The *family study* did not detect other conjoined twins in these families. The occurrence of twins and congenital anomalies among sibs was not higher than expected. The occurrence of fetal death was higher in the previous pregnancies of conjoined twins' mothers (26.7%) than in those of healthy singleton controls (15.6%) and of healthy twin controls (16.5%). However, the differences were not significant. The highest rate of fetal deaths was found in multimalformed singleton controls (27.6%, Table 6).

Table 6 - Previous pregnancy outcomes of mothers of conjoined twins and controls

Pregnancy outcome	Conjoined twins (N=36)		Healthy twins (N=108)		Healthy singletons (N=108)		Malformed singletons (N=108)	
	N	%	N	%	N	%	N	%
Livebirth	21	46.4	68	51.1	78	60.5	89	51.1
Induced abortion	12	26.7	41	30.8	30	23.2	28	16.1
Miscarriage	12	26.7	22	16.5	20	15.5	48	27.6
Stillbirth			2	1.5	1	0.8	7	4.0
Ectopic							2	1.2
Total	45	100.0	133	100.0	129	100.0	174	100.0

The higher rates of *pregnancy complications* are obvious in the study material but probably these are consequences, and not the causes, of conjoined twinning.

Table 7 - Data of family planning

Family planning	Conjoined twins (N=36)		Healthy twins (N=108)		Healthy singletons (N=108)		Malformed singletons (N=108)	
	N	%	N	%	N	%	N	%
Extramarital	2	5.6	5	4.6	4	3.7	6	5.6
Unplanned	3	8.3	7	6.5	3	2.8	4	3.7
Inadvertent conceptions in pill-users	6	16.7[a]	4	3.7	3	2.8	8	7.4
Ovulation induction	2	5.6[b]	1	0.9			1	0.9

[a] $\chi_1^2 = 6.98$, $P < 0.05$; [b] $\chi_1^2 = 6.77$ (after Yates correction), $P < 0.05$.

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Six cases of conjoined twins were conceived immediately after or during the use of *contraceptive pills* (16.7%) and two mothers were treated by ovulation inducing drugs (Table 7). These rates exceed significantly the figures of control groups. The rate of extramarital and unplanned pregnancies did not show a significant surplus in the study material.

Among *maternal diseases* immediately prior to conception or during pregnancy, hepatitis occurred in 5 cases and nephritis in 4 cases. These numbers exceeded figures of matched control samples. Hypothyroidism was diagnosed in two cases. Furthermore heart disease, diabetes and serious alcoholism was mentioned in one case.

The possible *occupational hazards* were mentioned more frequently in the study material (16.6%) than in malformed singletons (6.5%), healthy singletons (0.9%) and healthy twins (1.9%). However, this difference can be explained by a recall bias. Nevertheless, it is worthwhile noting that the origin of conjoined twinning was explained by clinicians in terms of occupational exposition in two cases. In Case 6 the mother worked in a fur factory where she inhaled the vapours of dimethyl ketone, denaturated alcohol and petroleum during the first trimester of her pregnancy. The mother of Case 35 worked as an electroplater in chromium industry. Also, three mothers mentioned a severe occupational poisoning one or two years before conception. In Case 7, the mother was a typesetter in a printing plant and she suffered from a lead pollution. In Case 12, the mother worked as a laboratory assistant and she had ammonia-poisoning. In Case 16, the mother was a tinner and she was exposed to cupric sulphate (blue vitriol).

DISCUSSION

The observed total prevalence of conjoined twinning was 1:68,000 in Hungary. This rate is similar to the values estimated by nationwide Swedish data [16], by Castilla et al in Latin America [6] and by some other countries of the International Clearinghouse (eg, Czechoslovakia, Denmark, Italy, New Zealand, Australia) (Table 2). The differences in birth prevalence of conjoined twinning in the various countries can be attributed to several causes. Conjoined twinning occurs more frequently in the African and Asian populations [4,13,20,30]. This is surprising because the occurrence of monozygotic twinning is nearly the same in all ethnic groups and variations are known only for dizygotic twinning. The low number of cases and the heterogeneous evaluation of stillbirths and spontaneous abortions may also play a role. In some studies only symmetrical cases have been evaluated.

No temporal or geographical clustering of conjoined twinning were found in Hungary, 1970-1986. However, it was remarkable that the total prevalence was 7.99 per million in the period 1970-75 vs 19.94 per million in 1976-83 ($\chi_1^2 = 5.44$; $P < 0.05$). There was a significant increase after 1981 with the use of the CUSUM method, too. The ascertainment was the same during the study period. The highest occurrence (4 cases, 31.01 per million births) was recorded in 1986. Additionally, of these four cases, two were triplets. The expected occurrence of triplets among

conjoined twins is extremely rare: 1 in 70 millions. These two exceptional events occurred in the same year, with a difference of only a few weeks, in two women living 100 km apart from each other.

A cluster in time and/or space of conjoined twins was reported several times. In the State of New York, 22 cases were found from 1945 to 1965 [19]. Twelve of these twin sets were born in the period 1955-1959 (6 expected) and 6 of these were born in 1959. Five of the latter were born in the same area of western New York, 50 miles from each other. In South Africa, 12 sets of conjoined twins were born just over twelve months in 1974-75 [3]. In a Swedish hospital (Skövde) 3 sets were born among approximately 2,300 annual births [16]. Recently, a cluster of conjoined twins was observed in Latin America [6].

The distribution of conjoined twin types is similar in large series of the literature [6,12,29]. There is a considerable excess of thoracopagus cases. The proportion of dicephalus is somewhat higher while the occurrence of pygopagus is lower in the Hungarian material than in other studies. The high rate and distribution of associated anomalies is interesting from the pathogenetic aspect.

The higher proportion of females among conjoined twins is a general caracteristic [19,24]. Male conjoined fetuses could have a higher risk for early fetal death. The rate of miscarriages in the previous pregnancies of conjoined twins' mothers was higher than expected based on Hungarian population figure: 26.7% vs 13.1% in the study period [11]. This double rate may indicate prenatal selection in previous pregnancies, too. The miscarriage rate in the previous pregnancies of mothers of conjoined twins was also higher than either control group. Such a finding is unlikely to be due to recall bias [17,27]. However, in a study of women "sensitized" to their reproductive functions (by virtue of their participation in a long-term study of menstrual cycles), overall only three quarters of recorded miscarriages were recalled. Recall was influenced by length of gestation and the interval between miscarriage and interview [28].

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