

Epidemics of poliomyelitis in the Maltese island of Gozo: genetic susceptibility

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Abstract

There were fewer epidemics of polio in Gozo than in the much bigger island of Malta, but over many years the proportion of cases was similar. Within Gozo, the attack rate was greater in some villages and this was caused by some extended, related families with genetic susceptibility to polio. In these families, there was considerable consanguinity. Two thirds of the polio cases were related. To understand polio, one must study the disease over many years, not just isolated statistics from unconnected epidemics.

Key words

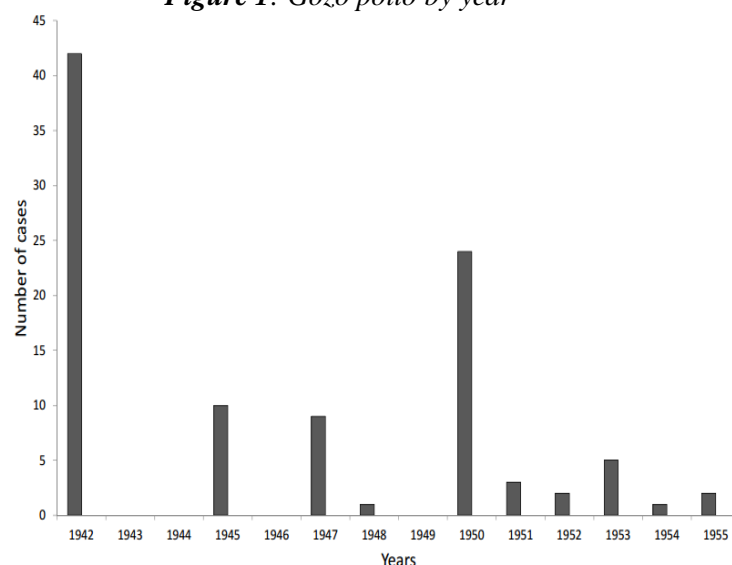
Consanguinity, genetic susceptibility, Malta, poliomyelitis

Introduction

The smaller island of Gozo lies north-west of Malta (for a map, see¹) with an area of 67 square kilometres and about one tenth the population. The polio cases on Gozo and their controls form the basis of this study: many of the parents and grand-parents of polios and controls on the island of Malta had migrated from Gozo and have been included. Polio seasons typically began after September and lasted over Christmas to spring with very few cases in May to August. Therefore the annual incidence (Fig. 1) is given for each July to the next June, emphasising the extent of the epidemics. In most years when there were no cases on Gozo, there were some on the bigger island of Malta. The last cases on Malta and Gozo occurred in 1964, after which the Sabin oral polio vaccine (OPV) was used to immunise the population.

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Figure 1: Gozo polio by year



Materials

With the permission and support of the Chief Government Medical Officer, I was able, in 1982-1987, to examine the records of the Infectious Diseases Hospital 1926-1964 (which were later trashed by unknown vandals) and the Physiotherapy notes at Saint Lukes Hospital. With the permission and support of the Archbishop of Malta, the Bishop of Gozo and the Director of the Public Registry (a lawyer) I was able to trace the polios and their controls in the records of baptisms and marriages which, in the parishes, also gave dispensations for consanguinity.

I found details of 1 072 polios in Malta and Gozo and these were traced to their great grandparents. For each polio, a control child (prefixed 'C') was found either five baptisms before or after in the same parish and traced in the same way. The marriages of the great grand-parents were traced in three parishes although this involved more than doubling the work. A register of polio cases in the Gozo Health Department gave details of cases plus another 2 whose parents came from Gozo, but who were paralysed on Malta. I found cases from 1909 to 1940 in the Gozo hospital records when they later received treatment as adults. One teenager, born in Detroit, but paralysed on holiday in Gozo, was traced through his uncommon surname. The father of one polio

child was a UK soldier and 3 fathers of controls were born in the UK.

Results

There were 39 cases with paralysis in the Maltese islands prior to the 1942-1943 epidemic and of these, seven were from Gozo. Although the numbers are small, one might have expected only four from Gozo as the population was about one tenth that of Malta. In the years after 1943, Gozo had fewer outbreaks than Malta suggesting that the virus travelled less frequently to Gozo. Nevertheless, the cases from Gozo were similar to the ratio of the populations of the two islands. Of the 110 polio children born on Gozo, 67% were related. Eight were sibs, 15 were first cousins, (17 were first and second cousins) and 64 were second cousins.

The 1942 epidemic

The first case occurred on 3 December in Victoria and was followed the next day by two sibs in Zebbug and others in Nadur, Victoria and Sannat (part of Victoria). These cases were not at first diagnosed as polio by the local pediatrician (personal interview). For these cases to have occurred almost simultaneously all over the island suggests a common source infecting a number of adults who travelled to the different villages. There were no soldiers on the island and little petrol for transport. Probably a small boat had returned from Marsa or Valletta on the main island of Malta after carrying produce to market – the siege had been lifted, but food was still scarce. There were 35 cases until January, with two unrecognised at the time. The last cases in Zebbug on 12 December and Victoria on 21 December left the virus circulating in Xaghra (11 January) and Nadur (18 January), reappearing in Gheinseilem on 16 April and Xaghra again on 1 May 1943.

The majority of cases, 70%, were under 3 years old when paralysed suggesting that there had been previous silent waves of virus circulating before the isolation of Malta after June 1940 by the Italian navy – there had been single cases in 1937 and 1939. Not everyone had been infected in these earlier waves – the 1937 case was 24 yr old and there was a 7 yr old in Gharb in 1942.

Later epidemics

In later years a few older children were paralysed – a 10 yr old and a 14 yr old in Victoria in 1946 and 1961 respectively and a 9 yr old in Xewkija in 1954, suggesting that although each wave of viruses circulated very widely, there were a very few children who escaped infection.

In 1945-1946 there were 5 cases in Kercem (part of Victoria) starting on 17 December (the only cases ever in Kercem) with 3 other cases in Victoria and 1 each in Xewkija and Xaghra. An even smaller outbreak started

in July 1947 with 2 cases in each of Xewkija, Victoria and Nadur and 1 case in Xaghra with the last case in October. Two cases in June 1948 in Xaghra and June 1949 in Zebbug may represent continuing presence of a virtually avirulent strain. In August 1950 another wave of cases continued until July 1951 with 23 cases in all the major villages, including 8 children in Nadur.

The incidence was far from uniform (Table 1) with the highest in Zebbug the smallest and most remote village, and the lowest in the cosmopolitan Victoria and two small remote hamlets. However, there were several cases where a polio in one parish was related to several in another parish e.g. two polios living in Gheinseilem were closely related to at least four, and possibly six others in Xaghra.

Table 1: The incidence of polio in Gozo 1942 – 1964.

Town/village	No. of children No. of polio cases % cases/children < 4 yr 1931 census ²	No. of polio cases 1942-1943	No. of polio cases 1945 +	% cases/ children
Zebbug	89	4	4	9
Gheinsielem	145	7	4	7.6
Nadur	296	8	12	6.6
Qala	122	1	6	5.7
Xaghra (Caccia)	297	10	5	5
Xewkija	240	1	10	4.6
Gharb & Ghasri	142	3	3	4.5
Victoria (Rabat)*	673	7	18	3.7

*I have included San Lawrenz, Kercem and Sannat with Victoria (see ¹). There was no census after 1931 because of the war.

Genetic susceptibility in the villages

Although a US doctor had published many papers citing families with multiple cases, in the 1900's³, genetic susceptibility had been ignored after 1935, in favour of research for vaccines. The first page of the medical notes of the polio children from 1942-1943 and those from earlier and later years, is blank with no genetic or family information. However, the genealogical data of the grand-parents and great grand-parents has shown many relationships.

In the village of Nadur, I traced further ancestors because the complicated consanguinities suggested further relationships. In the 1948 census the village had a population of more than 3,000, most of whom had been born in the parish. There were 29 cases of polio in

the parish, of which 23 were related in a convoluted, tightly knit group of families linked by many consanguinities (Table 1), with two cases now living in other villages. Of the 39 cases in Malta and Gozo prior to the 1942-1943 epidemic, two were from this group from Nadur (cases #20 and #32 in 1909 and 1921). There were five cases and one in another parish in the 1942-1943 epidemic and others in 1947, 1952, 1956 and

1958 with nine in 1950-1951 and two, with one in another parish, in 1953.

The 23 polios were related with 33 links to each other and to 12 controls in the same family complex. There was one pair of polio sibs, and another mixed pair of a polio and a control. There were more consanguinous marriages among the polio ancestors than among those of the controls (Table 2).

Table 2: *Consanguinity among parents and grand-parents on Gozo: marriages of Parents ranged from 1908 to 1957 and grand-parents from 1871 to 1922. (Details of dispensations from the parish registers)*

	Parents						Grand-parents					
	No.	II	II/III	III	III/IV	IV	No.	II	II/III	III	III/IV	IV
Polis	103	6	2	9	0	0	249	4	7	15	14	33
Sibs										1		2
Shared										1	5	6
Controls	111	5	0	8	0	1	251	7	1	10	7	22

Three pairs of mixed sibs, each with one polio and one control shared grand-parent marriages: these and other shared grand-parents have been included in both totals. Dispensations for IV have not been required since 1917.

The small, straggling village of Zebbug is isolated in the North West corner of the island and had eight polios, seven of whom were closely related with many consanguinities (Table 3). The seven polios were related with ten links to each other and to a control in the same family complex. There were two cases and a pair of sibs in the 1942-1943 epidemic. The eighth case, #1243, was related to case #154 at another village through a great grand-parent. The father of case #443 from a smaller parish nearby was married to a lady from the village, whose sister was married to case #459 from another village. The mother of case #494 from Nadur was from here: her parents were II/III plus IV and her maternal grand-parents were related to case #865. In the 1948 census⁴ there were more than 1,000 inhabitants most of whom had been born in the village, although many left for other villages. There were many more children since the previous census.

The large parish of Xaghra lies between Zebbug to the north-east and Nadur to the east with four 1942 polios related in a family group with two others living in Gheinseilem and a third who was living in Germany. Two cousins (one was 8 yr old) had unusual surnames and were almost certainly related to three of the family group. A pair of sibs had a paternal grand-parent IV and another polio had one who was III/IV.

Xewkija lies just south of the main road from the port of Mgarr (Gozo) to Victoria and suffered ten polios

in the last years of the epidemics - one of whom was 9 yr old. There were three pairs of polios who were second cousins with several consanguinities and a pair of cousins, one of whom was brother to a possible polio who had a limp.

Gharb is an isolated parish to the east of the island where a 24 yr old with parents III suffered polio in the inter war years. There were two pairs of second cousins and one polio a cousin to two second cousins in St. Julian and Cospicua on Malta. One polio whose parents were III, had maternal grand-parents who were II/III.

Gharb is an isolated parish to the east of the island where a 24 yr old with parents III suffered polio in the inter war years. There were two pairs of second cousins and one polio a cousin to two second cousins in St. Julian and Cospicua on Malta. One polio whose parents were III, had maternal grand-parents who were II/III. The large town of Victoria had the smallest proportion of cases, of which 15 and a pair of sibs were not related to others. The 13 yr old from Detroit, was a second cousin to a case in Mellieha on Malta. One polio was a second cousin to another polio and to two step-brothers. A 10 yr old polio whose parents were cousins, II, was a second cousin to a 14 yr old whose grand-parents were II/III. Four second cousins were from Sannat (parents II/III), Victoria (grand-parents IV) and two (second-cousins) from Xaghra (one with grand-parents IV, the other with parents III and grand-parents IV). One polio

with grand-parents IV was cousin/second cousin to a polio in Zurrieq (Malta) with parents III and grand-parents IV.

Isonymy

There was little difference in the number of isonymous marriages between the polios and the controls. The number of isonymous marriages was highest in Nadur and was low in Victoria, similar to

consanguinity. Five of the 12 parent and 13 of the 26 grand-parent isonymous marriages were consanguineous.

Surnames of polios

Some uncommon surnames were associated with polios and their great grand-parents also showed that they were less common among the controls (Table 4).

Table 3: Consanguineous marriages in villages in Gozo. (Details from the parish registers)

Village	<u>Polios</u>						<u>Control</u>					
	Parents			Grand Parents			Parents			Grand Parents		
	Consanguineous						Consanguineous					
	No	Percentage		No	Percentage		No	Percentage		No	Percentage	
Nadur	28	4		53	19 ^a		23	10		61	14 ^b	
Xaghra	14	1		39	16		14	1		37	7	
Xewkija	11	1		24	7 ^a		13	0		32	7 ^a	
Gheinsielem	6	1		21	3		11	1		16	4	
Qala	6	2		16	7		7	0		12	4	
Gharb	8	2		17	4		6	0		16	4	
Zebbug	7	2		13	8		9	1		19	4	
Total	80	13	16%	183	63	34%	83	13	16%	193	44	23%
Nadur	23	3	13%	65	4	6%	28	1	3%	58	3	5%

Notes: ^a of which one was a double consanguinity:

^b of which four were double, eg II + IV).

Table 4: Surnames of great grand-parents associated with polios or controls

Surname	Parish	No.		Totals on Gozo	
		Polios	Controls	Polios	Controls
X	Gharb	17	0	31	14
Y	Victoria	10	2	35	15
Z	Xaghra	10	2	11	7
U	Victoria	8	3	11	4
Q	Victoria	7	1		
	Gharb	7	2	16	3
O	Xaghra	41	29	57	41

Sibs

There were eight pairs of sibs in my sample – three with both polio sibs, four where only one had polio, and one pair of both controls. Of those with both polios, two had grand-parents III/IV and IV and there were also two pairs of polio sibs on Malta for each of which one grand-parent came from Gozo. Of the four with mixed polio and control, two had only one Gozitan parent and another pair were step-children (a control Gozitan and a polio child on Malta): another had IV grand-parents. A mixed pair in Cospicua (Malta) had one grand-parent from Gozo. The parents of the pair of control sibs were isonymous with one pair of III/IV grand-parents and grand-parents of a polio.

Discussion

At the beginning of this study there was no suggestion among the published papers and the archives that the distribution of polios was other than random in the various villages. The unexpected concentration of polios in some villages has meant that the controls are also more frequent in those villages and are not a reflection of the entire population. I chose the baptism of a control four distant from the polio to avoid baptisms of twins and cousins: I did not know that the priest in those times would hurry to baptise a new-born infant in case it died or was still born.

In general, there were more consanguineous marriages among the parents of polios and many more among the grand-parents (Table 2). The large village of Nadur had the greatest proportion of consanguineous marriages and, surprisingly, Zebbug had few (Table 3). There have been fewer consanguineous marriages in recent years so there are far fewer among parents than among the grand-parents. As in Malta, some families showed multiple consanguinities in both parent and grand-parent marriages (Table 5). Consanguineous marriages over several generations were common in a few families, probably due to the limitations on meeting other suitable teenagers. Family events, marriages, fiesta celebrations and birthdays provided the unsupervised meeting of teenagers. Within a village, there would be a hierarchy of respectability. At the top would be someone wealthy, the president of the church laity, president of the band club and the leading political party. His family would not normally marry beneath them. During epidemics, meetings between children would have been avoided.

It was said of Zebbug that ‘intra-village marriages up to the war [1940] were 80%’ and ‘new households set up in the village as close to the core as possible’.⁵ Vella⁶ reporting a high incidence of thalassemia minor, said ‘Zebbug was described by a prominent educated villager as one large family’. Vassallo⁷ had reported on consanguinity in Gozo. In small communities, counts of isonymous marriages from marriage registers may

include some which are related. However, although an isonymous marriage with a rare surname may well indicate common sharing of genes, this may not apply with more common surnames. There may well be many non-intermarrying groups for instance among e.g. Buttigieg, which may include both rich and poor. The Maltese surnames of Catania and Messina were possibly given to immigrants from those towns in Sicily, but immigrants with these same Maltese surnames may be unrelated and came at different times. Marriages from two villages may be between a couple from adjacent farms in the two parishes.

Conclusion

There is much evidence of genetic susceptibility to poliomyelitis.

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Table 5: Multiple consanguinities in Gozitan villages

Village	Polio	Family of		
		Parent	Grand-parents respectively	Great Grand-parents
Zebbug	483	III	III + IV	
Gheinsielem	1241	II	IV	
Nadur	1220	III	II/IV & II/III + III	III + IV & II + III
	1222		II + IV & II/III + III	III + IV & II + III
	1227		III & IV	IV & III
	1228	II/III	III + IV & IV	III + IV
	1223		III/IV	III
Qala	1248	II	III	III
Xaghra	1204	III	IV	
	1203		IV	IV
Gharb, Ghasri	1252	III		II/III

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