EUROCAT Joint Action Jan 2011 – Dec 2013

Evaluation Report

(April 2014)

EUROCAT Central Registry

Room 12L09
University of Ulster
Newtownabbey, Co Antrim
Northern Ireland, BT37 0QB
Tel: +44 (0)28 90366639
Fax: +44 (0)28 90368341
Email: eurocat@ulster.ac.uk
Web: www.eurocat.ulster.ac.uk

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WHO Collaborating Centre for the Surveillance of Congenital Anomalies

Summary

The EUROCAT Project Management Committee were responsible for the evaluation of the EUROCAT Joint Action Project and for the development of the Evaluation Plan. This included internally conducted Process and Effect Evaluation in addition to sub-contracting the services of an independent evaluator (one of the many methods employed to achieve effect evaluation). Process, output and outcome indicators, milestones and deliverables were monitored throughout and on the whole were met in a timely fashion in accordance with the project plan – enabling confirmation that the specific stated objectives of the project had been met.

Where possible EUROCAT Central Registry will endeavour to continue to conduct effect evaluation by way of monitoring relevant identified impact indicators, such as citations of Joint Action outputs, e.g. published scientific papers in peer-reviewed journals. In keeping with this strategy of monitoring impact indicators, EUROCAT has developed an impact case study relating to Antiepileptic Drug Safety in Pregnancy and tracked the citation profile of a number of published scientific papers in peer-reviewed journals (key outputs of the EUROCAT's last round of funding) in addition to any published scientific papers (in peer-reviewed journals) that arose during the current project. Such publications were widely cited, with some in particular having a notable impact as evidenced by particularly high citation indexes, and encouragingly new emerging outputs have already begun to accumulate a citation profile.

Over the period of the Joint Action (Jan 2011 to Dec 2013) there were 70,670 visitors to the EUROCAT website (from 182 different countries globally), 38,763 unique visitors and 207,221 page views. 54.2% of visitors were new to the website and 45.8% were returning visitors.

Since registration to obtain website prevalence data from the website was initiated in March 2012, 564 individuals from 54 different countries and diverse roles (spanning the entire range of EUROCAT's target audience) have registered to use EUROCAT's interactive website prevalence tables.

EUROCAT Central Registry continues to respond to a variety of external enquiries received by email from a diverse array of individuals spanning the full spectrum of EUROCAT's target audience i.e. Parents of affected individuals, patient organisations, pharmacists, clinicians, registry leaders, epidemiologists, pharma industry, students, government officials, public health officials, reporters etc. A special enquiry form has been developed to deal with increasing demand for information on medication exposure.

During the Joint Action EUROCAT Central Registry has added a "media interest" section to the website and has begun to populate that section of the website with reference to EUROCAT made within the media.

EUROCAT aimed to obtain constructive feedback (relating to the Joint Action) about its website and other outputs via a web-based evaluation survey tool. There were 133 respondents from 32 different countries globally. Interestingly 44% of the countries represented by respondents were non-EUROCAT member countries. Respondents visited the website for a diverse array of information but prevalence of congenital anomalies was the main topic of interest that individuals visited the website for. Respondents indicated that the risk factors that they had the most need for information on when visiting the website was medication exposure in pregnancy, folic acid and genetic factors. The vast majority of respondents described the interactive website prevalence tables as "very useful". Encouragingly 86% of respondents found the website easy to use. Some of those

who thought it was not easy to use provided constructive criticism that will be considered and acted upon where possible by the EUROCAT Website Dissemination Committee. For example, some of the responses given suggest that some individuals are unable to find existing information that is already on the website.

Both EUROCAT Symposia that took place within the timeframe of the EUROCAT Joint Action Project were very well received and well attended. 232 participants from 25 different countries attended the first EUROCAT symposium of the Joint Action on the prevention of congenital anomalies, and 186 participants from 26 countries attended the second.

A questionnaire (created by an external subcontractor and accessed via an online survey tool) was employed to focus on perception of "value" of the EUROCAT outputs/outcomes by targeted end-users. EUROCAT member registry leaders were asked to obtain relevant recipients that represented their nation in one of 4 categories – (i) Public Health Authority (ii) Clinician (iii) Department of Health (iv) Patient or patient organisation. 191 individuals received the EUROCAT Independent Evaluation Questionnaire Survey link. Of those 93 responded (creating the "Evaluation Panel") giving a 49% response rate. 22 respondents identified themselves as Registry Leaders. The results were analysed including and excluding Registry Leaders.

Of the non-registry leader respondents (excluded 22 respondents who identified themselves as a EUROCAT member registry leader), 94% would recommend that new registries (registering cases of congenital anomaly) become a member of the EUROCAT Network.

In corroboration with the results of the web-based evaluation survey, the provision of prevalence information was identified as the most important role of EUROCAT's ongoing congenital anomaly surveillance. Respondents found the website the most useful means to access EUROCAT information.

Of the non-registry leader respondents, 69% had accessed information or data provided by EUROCAT. This information or data was used for a wide variety of purposes, in particular to enable between country comparisons.

Worth noting is that 99% of respondents (excluding EUROCAT member registry leaders) believed that there was a continuing need for the work that EUROCAT does.

Constructive suggestions were received for how to improve the work of EUROCAT, for example increased engagement with the lay community including with patient organisations.

When asked what emerging areas EUROCAT should focus on in the future, responses included, epigenetic research, establishing links with WHO, environmental pollutants, and medication in pregnancy.

General comments emphasised the importance of EUROCAT (i) as a unique resource of large-scale quality epidemiological data on congenital anomalies (ii) to evaluate the possible risks of medicines taken by pregnant women.

Part A – Process and Effect Evaluation

An Evaluation plan for the EUROCAT Joint Action Project was agreed by the EUROCAT Project Management Committee and was first submitted to the EAHC as an annex to the second interim report of the EUROCAT Joint Action (Annex 1).

Process Evaluation

Process evaluation related to planning, organisation and assuring quality of implementation of the EUROCAT Joint Action project activities, identifying and overcoming obstacles and verifying that the stated objectives had been met. This included determining that the process/output indicators (detailed in WP3 report), the milestones and the deliverables (detailed in each respective WP report) had been met. The process indicators within the EUROCAT Joint Action project were designed to measure the progress of activities in the EUROCAT Joint Action and the way that these activities were carried out (e.g. annual data transmission by EUROCAT registries, by how many?, did they meet the deadline?).

The output indicators within the EUROCAT Joint Action project were designed to measure the quantity, quality and timeliness of the products of the EUROCAT Joint Action activity (e.g. update of website prevalence tables for all congenital anomaly subgroups, how many EUROCAT registries provided data to enable this?, was this achieved annually as planned?).

Process evaluation was internally conducted by the EUROCAT Project Management Committee and the Steering Committee.

Effect Evaluation

Effect evaluation related to evaluation of outcome and impact during the period of the EUROCAT Joint Action and for a five year period predating commencement of the EUROCAT Joint Action. Outcome indicators (detailed in WP3 report) were designed to measure the intermediate results generated by the EUROCAT Joint Action outputs (e.g. improvement in Data Quality Indicators in EUROCAT registries). Impact indicators measure the quality and quantity of long-term results generated by the EUROCAT Joint Action output (e.g. citations of EUROCAT prevalence data/published papers). EUROCAT will endeavour to monitor this in the years following the EUROCAT Joint Action Project.

Effect evaluation was internally conducted by the EUROCAT Central Registry team, in addition to sub-contracting an independent evaluation team to assist in surveying the value of EUROCAT (detailed in Section H of Part B)

To serve as a reminder…

The general objective of the EUROCAT Joint Action Project was:

The epidemiologic surveillance of congenital anomalies (CA) through the EUROCAT Network of population-based CA registries, and through doing so supporting the reduction of the public health burden of CA by promotion of health; reduction of teratogenic risks preconceptionally and in early pregnancy; high quality diagnostics, treatment and counselling prenatally and postnatally; and minimising inequalities in the experience of prevention and care.

The general objectives of the EUROCAT Network were stated as:

1. To provide essential epidemiologic information on CA in Europe
2. To facilitate early warning of new teratogenic exposures
3. To evaluate the effectiveness of primary prevention
4. To assess the impact of changes in prenatal screening
5. To act as an information and resource centre for the population, health professionals and policy makers regarding clusters, exposures and risk factors of concern
6. To provide a ready collaborative network and infrastructure for research related to causes and prevention of CA and treatment and care of affected children
7. To act as catalyst for the setting up of new registries in Europe collecting comparable, standardised data

The specific objectives of the EUROCAT Joint Action Project were:

1. Prevalence Information.

To provide comprehensive epidemiologic information on prevalence of CA in Europe. This was needed for public health planning and was essential background information for several other objectives, in particular assessment of the impact of policies for prevention, prenatal diagnosis and care of newborns with CA, and role of old and new (emerging) risk factors eg. swine flu.

Related indicators

Process Indicators	**Output Indicators**	**Outcome Indicators**
Successful annual data transmission to Central registry from all Associate Partner Registries.	Prevalence tables for 95 anomaly subgroups on the EUROCAT website, updated each year.	Citations of EUROCAT prevalence information.

Regarding the Process Indicator – the majority of Full and Associate Member registries successfully transmitted data to EUROCAT Central Registry each year of the EUROCAT Joint Action (see full detail in WP4 report). Most recently (in 2013 of the EUROCAT JA) prevalence data for 31 registries was updated to 2011 in EUROCAT Central Database and new prevalence tables uploaded to website - http://www.eurocat-network.eu/accessprevalencedata/prevalencetables. Data on the proportion of cases prenatally diagnosed was transmitted by 25 EUROCAT member registries.

Regarding the Output Indicator – Prevalence tables for the entire list of EUROCAT anomaly subgroups have been updated each year of the EUROCAT Joint Action (see full detail in WP4 report). EUROCAT allows web visitors to interactively interrogate anonymous aggregate prevalence data (updated biannually and made available globally within 6 weeks of receiving it), by registry, year and 89 CA subgroups of interest ie. Spina bifida, Down syndrome. In April 2014, data was updated to birth year 2012.

Regarding the outcome indicator – citations of EUROCAT prevalence information, collectively website registrations to use the prevalence tables available on the EUROCAT website (detailed in Section C of Part B), the results of the web-based evaluation survey (detailed in Section F of Part B), the log of external enquiries (detailed in Section D of Part B) and the results of the Independent Evaluation (detailed in Section H of Part B) all confirm that EUROCAT prevalence information is used widely. See also Section A of Part B for examples of citations of prevalence tables.

2. Network expansion and data quality improvement

To co-ordinate the establishment of new registries throughout Europe collecting comparable, standardised data. The integration of new registries and countries would

enable the sharing of knowledge and expertise and a widening contribution to public health planning. Data quality monitoring and improvement was an essential prerequisite for all other objectives within the EUROCAT Joint Action project that used EUROCAT data, and for effective comparison between countries.

Related indicators

Process Indicators	**Output Indicators**	**Outcome Indicators**
Integration of Latvia, Slovenia and Valencia as new Full Members of EUROCAT, with successful data transmission. Submission of revised ICD11 proposal to WHO.		Improvement in Data Quality Indicators in two thirds of Registries.

Regarding the first Process Indicator – Latvia and Slovenia have progressed to Affiliate membership level and Valencia has become a Full Member registry (see full detail in WP4 report), in addition to French West Indies and SW England.

Regarding the second Process Indicator – Revised ICD11 proposals were submitted to WHO (see full detail in WP5 report).

Regarding the Outcome Indicator – Data Quality Indicators were improved in two thirds of registries (see full detail in WP5 report).

3. Early warning

To co-ordinate the detection and response to trends and clusters and early warning of teratogenic exposures, to allow appropriate action to be taken regionally, nationally and at an EU level.

Related Indicators

Process Indicators	**Output Indicators**	**Outcome Indicators**
Improvement in number of Registries meeting earliest data transmission deadline.	Annual Statistical Monitoring Reports. Five in-depth investigations, including one by TEC and one of swine-flu impact. Environmental data linkage feasibility report.	Generation and preliminary investigation of clusters/trends in each registry.

Regarding the Process Indicator – There was in improvement in the number of registries meeting the earliest data transmission deadline (see full detail in WP4 report).

Regarding the Output Indicators – 3 annual statistical monitoring reports were been created (see full detail in WP4 and WP6 report). More than five in-depth investigations were conducted within WP6. The TEC investigation did not happen, alternatively resources were diverted to expand the in-depth investigation of the impact of swine-flu (see full detail in WP6 report). The environmental data linkage feasibility report was created as part of WP6 (see full detail in WP6 report).

Regarding the Outcome Indicator – Generation of clusters/trends in each participating registry, and subsequent preliminary investigation by affected registries, happened as part of each year's annual statistical monitoring process (see full detail in WP4 and WP6 report).

4. Primary prevention policy

To make recommendations for the inclusion of primary prevention of CA in national rare disease plans and to evaluate the effectiveness of existing primary prevention measures. A focus will be on folic acid but this objective will also assess other potential routes including management of chronic diseases, drugs in pregnancy, maternal infection and vaccination, environmental pollution, alcohol and smoking, and socioeconomic and migrant issues.

Related Indicators

Process Indicators	Output Indicators	Outcome Indicators
	Report on public health actions relevant to prevention of BD at EU MS level. Report on actions to prevent NTD by raising folic acid status at EU MS level. Report on potential consensus approach toward inclusion of primary prevention actions in national plans on RD.	

This output indicator was delayed. The three part report has been delayed until May 2014 (see explanation in WP7 report). However, two surveys were completed. The first on Policies for Primary Prevention of Neural Tube Defects with Folic Acid and Folate and the second on Public Health Actions on Primary Prevention of Congenital Anomalies. The EUROCAT/EUROPLAN Recommendations on policies to be considered for the primary prevention of congenital anomalies in National Plans and Strategies on Rare Diseases (developed as part of WP7 of the EUROCAT Joint Action) were compiled and published on the EUROCAT website and within the Public Health Genomics Journal. EUROCAT is already liaising with EUROPLAN to discuss measures to monitor the inclusion, implementation and impact (across Member States) of the EUROCAT/EUROPLAN Recommendations.

5. Prenatal screening information (such as for Down syndrome and genetic syndromes)

To assess the impact of developments in prenatal screening at a population level, with particular reference to Down Syndrome. Prenatal screening is continually evolving, and evolving differently in each country. EUROCAT will allow a common approach to monitoring of detection rates for individual anomalies and pregnancy outcomes in relation to policy and demographic characteristics eg. maternal age.

Related Indicators

Process Indicators	Output Indicators	Outcome Indicators
Integration of England and Wales NDSCR into central database.	Expansion of prenatal diagnosis tables on EUROCAT website.	Publication of 6 genetic syndrome papers.

Regarding the Process Indicator – Data were not incorporated into the EUROCAT Central Database as planned (see explanation in WP8 report). Alternatively England and Welsh National Down Syndrome Cytogenic Registry data were incorporated into the EUROCAT website as part of the prenatal diagnosis epidemiological tables.

Regarding the Output Indicator – Prenatal diagnosis tables were expanded on the EUROCAT website (see full detail in WP4 and WP5 report).

Regarding the Outcome Indicator – 3 papers have been published, one has been submitted (outcome pending) and 2 are to be submitted (see detail in WP8 report).

6. Postmarketing drug surveillance

To develop EUROmediCAT as an effective postmarketing surveillance tool in relation to medication use in pregnancy and risk of CA. Despite concerns since the thalidomide epidemic, there is still no effective postmarketing surveillance of drug use in pregnancy. Through the EUROCAT Joint Action EUROCAT aimed to continue to develop its role in this area by analysing a database of worldwide importance, and by exploiting new possibilities for linkage with prescription data.

Related Indicators

Process Indicators	**Output Indicators**	**Outcome Indicators**
3 Workshops on quality of medication exposure data. Joint meeting with ISPE and ENTIS.	3 scientific papers.	Protocol for early warning of drug-malformation associations.

Regarding the Process Indicators – workshops were held at each annual Registry Leaders' Meeting and a Joint meeting with ISPE and ENTIS took place (see full detail in WP9 report).

Regarding the Output Indicator – One report has been generated (to be converted to scientific paper in 2014), one paper has been published and one paper has been finalised for submission in 2014 (see full detail in WP9 report).

Regarding the Outcome Indicator – This protocol is now alternatively being developed as part of the FP7 funded EUROmediCAT project (see full detail in WP9 report).

Milestones and Deliverables

The EUROCAT Project Management Committee met frequently throughout the timeframe of the EUROCAT Joint Action to monitor progress and verify that milestones and deliverables were being met. On the whole the milestones and deliverables were achieved in a timely matter. A general overview per WP is provided below, but full detail is provided in each WP report.

WP1 – Coordination of the Joint Action
3 annual EUROCAT Registry Leaders' Meetings (milestones) were successfully achieved on time. Reports were slightly delayed due to the logistics of retrieving information from WP leaders who mostly preferred to wait until the entire reporting period was finished before sending completed information to the Project Manager. The Project Manager was also on maternity leave from M32-M36.

WP2 – Dissemination of the Joint Action
Each annual newsletter (3 milestones) and the Promotional Leaflet were delayed slightly mostly due to logistics – time taken to get the Joint Action project underway and to appoint the Project Manager and to retrieve relevant information from WP leaders and to

allow for design and editing. The minor delays did not impact on any task within the Joint Action Project.

2 EUROCAT Symposia were successfully achieved on time (both milestones and deliverables). See Section G in Part B for more information.

WP3 – Evaluation of the Joint Action
The PMC met frequently throughout the EUROCAT Joint Action Project and discussed evaluation (3 milestones). The Evaluation Report (the WP deliverable) has been completed to accompany the Final Report.

WP4 – Registration, Central Database, and Surveillance
The website tables were updated annually (milestones leading to Deliverable) to birth year 2011 (Deliverable), for EUROCAT's congenital anomaly subgroups, by registry, year and pregnancy outcome, on prevalence, perinatal mortality and prenatal diagnosis, with data quality indicators. 3 Annual Statistical Monitoring Reports on the detection of clusters and trends were produced (both milestones and Deliverable). Data transmission from new member registries (another milestone) was completed within the timeframe of the project.

WP5 – Coding and Classification, Data Quality
The coding and classification committee met 5 times as planned (milestones) and provided minutes. An Annual review of multiply malformed cases was also achieved with a very minor delay (milestone) as was implementation of EUROCAT Guide 1.4 (milestone). Publication of a revised set of data quality indicators on the website was also completed as planned. A scientific paper on epidemiology of multiple malformations was achieved much later than expected. There were very justifiable reasons given for the delay which did not impact on any other activity within the Joint Action Project.

WP6 – Investigation of Trends, Clusters and New Exposures
3 Annual Statistical Monitoring Reports were produced (2 annual milestones leading to a Deliverable). A milestone and a deliverable relating to the activity of the Taskforce for the Evaluation of Clusters were unable to be met. An explanation for this has been provided in the WP6 report and resources allocated to that activity were moved to other relevant activity within the WP. A set of scientific papers on epidemiology of selected anomalies and risk factors was created as planned within the timeframe of the project. A report on the feasibility of environmental data linkage and pilot study results was delayed slightly but has been completed. This did not impact on any other activity within the EUROCAT Joint Action project.

WP7 – Primary Prevention of Congenital Anomalies
The 2 milestones within the WP were met as planned and included achieving collection of public health actions relevant to prevention of birth defects and achieving collection of actions to prevent Neural Tube Defects by raising folic acid status. The deliverable associated with the WP - A 3-part report: I. Public Health Actions (Month 24). II. Prevention of Neural Tube Defects with Folic Acid (Month 24). III. National Plans (Month 36), has been delayed until May 2014. However, two surveys were completed. The first on Policies for Primary Prevention of Neural Tube Defects with Folic Acid and Folate and the second on Public Health Actions on Primary Prevention of Congenital Anomalies. The EUROCAT/EUROPLAN Recommendations on policies to be considered for the primary prevention of congenital anomalies in National Plans and Strategies on Rare Diseases (developed as part of WP7 of the EUROCAT Joint Action) were compiled and published on the EUROCAT website and within the Public Health Genomics Journal. EUROCAT is already liaising with EUROPLAN to discuss measures to monitor the inclusion, implementation and impact (across Member States) of the EUROCAT/EUROPLAN Recommendations.

WP8 – Prenatal Screening, Down Syndrome and Genetic Syndromes
This WP planned to produce 2 scientific papers on Down Syndrome and 4 scientific papers on genetic syndromes (collectively making up the WP deliverable). Milestones were set planning submission of the papers by particular Months within the Joint Action timeframe. 6 papers have been produced in accordance with the deliverable. 1 Down Syndrome Paper has been submitted and accepted for publication and one is about to be submitted. 2 genetic syndrome papers have been submitted and accepted for publication. Another has been submitted and the final one has been fully drafted and is about to be submitted. Throughout the milestones did not match the pace at which the papers were able to be created due to issues relating to data (see full detail in WP8 report).

WP9 – Medication During Pregnancy
The deliverable of this WP was to achieve 3 scientific papers on risks of specific medications and pharmacovigilance methodology. One report was created that will be converted to scientific paper post EUROCAT Joint Action. One paper was submitted and accepted for publication and another is about to be submitted.

The following milestones were met on time – a first annual workshop on ATC coding and a Joint symposium with ISPE and ENTIS. One milestone, an updated report on sources of data on maternal medication use used by registries contributing data to EUROCAT, was delayed substantially (in part due to maternity leave in the institution leading this WP) but has now been completed. The final milestone relating to data linkage was no longer considered part of the EUROCAT Joint Action as the related activity was moved to the FP7 funded EUROmediCAT project (Full detail in WP9 report). The start of EUROmediCAT three months after the start of the EUROCAT joint Action was a major step forward for pharmacovigilance in EUROCAT

Part B - Methods of Effect Evaluation

(a) **Citation Tracking**
Citation tracking (of peer-reviewed journal publications) using Scopus Citation Tracker (SciVerse) – conducted in April 2014.

Table: Citation record (excluding self-citations of all authors) of a selection of collaborative key peer-reviewed journal publications published as a result of activity undertaken by the EUROCAT Network during the last funding contract (2007-2010)

Year	Document Title	Authors	Journal Title	Vol	Iss	**2008**	**2009**	**2010**	**2011**	**2012**	**2013**	**2014**	**total**
						2	14	22	61	97	154	23	**373**
2013	Recent decrease in the prevalence of congenital heart defects in Europe	Khoshnood et al	Journal of Pediatrics	162	1	NA	NA	NA	NA	NA	1	0	**1**
2012	Epidemiology of small intestinal atresia in Europe: A register-based study	Best et al	Archives of Disease in Childhood: Fetal and Neonatal Edition	97	5	NA	NA	NA	NA	0	2	0	**2**
2012	Rare chromosome abnormalities, prevalence and prenatal diagnosis rates from population-based congenital anomaly registers in Europe	Wellesley et al	European Journal of Human Genetics	20	5	NA	NA	NA	NA	1	4	1	**6**
2012	Oesophageal atresia: Prevalence, prenatal diagnosis and associated anomalies in 23 European regions	Pedersen et al	Archives of Disease in Childhood	97	3	NA	NA	NA	NA	3	12	2	**17**
2012	Spectrum of congenital anomalies in pregnancies with pregestational diabetes	Garne et al	Birth Defects Research Part A - Clinical and Molecular Teratology	94	3	NA	NA	NA	NA	1	5	1	**7**
2011	EUROCAT Report 9: Surveillance of Congenital Anomalies in Europe 1980-2008	Boyd et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	0	1	0	0	**1**
2011	Paper 6: EUROCAT member registries: Organization and activities	Greenlees et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	0	1	3	0	**4**
2011	Congenital heart defects in Europe: Prevalence and perinatal mortality, 2000 to 2005	Dolk et al	Circulation	123	8	NA	NA	NA	5	18	31	5	**59**
2011	Paper 2: EUROCAT public health indicators for congenital anomalies in Europe	Khoshnood et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	0	7	8	0	**15**
2011	Paper 1: The EUROCAT network-organization and processes	Boyd et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	1	2	3	2	**8**

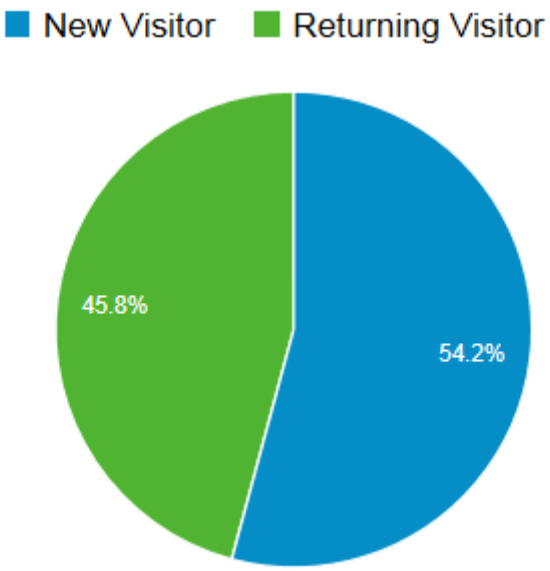
2011	Paper 5: Surveillance of multiple congenital anomalies: Implementation of a computer algorithm in European registers for classification of cases	Garne E et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	0	0	4	0	**4**
2011	Paper 3: EUROCAT data quality indicators for population-based registries of congenital anomalies	Loane et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	0	1	1	1	**3**
2011	Paper 4: EUROCAT statistical monitoring: Identification and investigation of ten year trends of congenital anomalies in Europe	Loane et al	Birth Defects Research Part A - Clinical and Molecular Teratology	91	1	NA	NA	NA	2	2	8	4	**16**
2011	Sex chromosome trisomies in Europe: Prevalence, prenatal detection and outcome of pregnancy	Boyd et al	European Journal of Human Genetics	19	2	NA	NA	NA	4	3	6	1	**14**
2010	Intrauterine exposure to carbamazepine and specific congenital malformations: Systematic review and case-control study	Jentink et al	BMJ (Online)	341	7785	NA	NA	1	6	19	16	1	**43**
2010	Valproic acid monotherapy in pregnancy and major congenital malformations	Jentink et al	New England Journal of Medicine	362	23	NA	0	4	25	23	36	3	**91**
2009	Maternal age-specific risk of non-chromosomal anomalies	Loane et al	BJOG: An International Journal of Obstetrics and Gynaecology	116	8	0	0	1	6	6	11	0	**24**
2008	Does lamotrigine use in pregnancy increase orofacial cleft risk relative to other malformations?	Dolk et al	Neurology	71	10	2	14	16	12	9	3	2	**58**

Table: Citation record (excluding self-citations of all authors) of collaborative peer-reviewed journal publications published as a result of activity undertaken by the EUROCAT Network during the EUROCAT Joint Action (Jan 2011- Dec 2013)

Year	Document Title	Authors	Journal Title	Vol	Iss	2012 5	2013 8	2014 3	total **16**
2014	Seasonality of congenital anomalies in Europe Birth Defects	Luteijn et al	Birth Defects Research Part A: Clinical and Molecular Teratology	In press		NA	NA	0	**0**
2014	Influenza and congenital anomalies: a systematic review and meta-analysis	Luteijn et al	Human Reproduction	In press		NA	NA	0	**0**
2014	Prevalence and risk of Down syndrome in monozygotic and dizygotic multiple pregnancies in Europe: Implications for prenatal screening	Boyle et al	BJOG: An International Journal of Obstetrics and Gynaecology	In press		NA	NA	0	**0**
2014	Prevalence, prenatal diagnosis and clinical features of oculo-auriculo-vertebral spectrum: a registry-based study in Europe	Barisic et al	European Journal of Human Genetics	In press		NA	NA	0	**0**
2013	Identifying associations between maternal medication use and birth defects using a case-population approach: An exploratory study on signal detection	De Jonge et al	Drug Safety	36	11	NA	0	0	**0**
2013	Atrioventricular septal defects among infants in Europe: A population-based study of prevalence, associated anomalies, and survival	Christensen et al	Cardiology in the Young	23	4	NA	0	0	**0**
2013	Trends in the prevalence, risk and pregnancy outcome of multiple births with congenital anomaly: A registry-based study in 14 European countries 1984-2007	Boyle et al	BJOG: An International Journal of Obstetrics and Gynaecology	120	6	NA	1	0	**1**
2013	Fraser Syndrome: Epidemiological Study in a European Population	Barisic et al	American Journal of Medical Genetics, Part A	161	5	NA	0	0	**0**
2013	Twenty-year trends in the prevalence of Down syndrome and other trisomies in Europe: Impact of maternal age and prenatal screening	Loane et al	European Journal of Human Genetics	21	1	NA	4	3	**7**
2013	Recent decrease in the prevalence of congenital heart defects in Europe	Khoshnood et al	Journal of Pediatrics	162	1	NA	1	0	**1**
2011	Differences in pandemic influenza vaccination policies for pregnant women in Europe	Luteijn et al	BMC Public Health	11		5	2	0	**7**

(b) **Google Analytics**

Over the period of the EUROCAT Joint Action (Jan 2011 to Dec 2013) there were 70,670 visitors to the EUROCAT website (from 182 different countries globally), 38,763 unique visitors and 207,221 page views. 54.2% of visitors were new to the EUROCAT website and 45.8% were returning visitors.



The top 10 countries visiting the website are detailed below and amount to 66% of total visits in the timeframe of the EUROCAT Joint Action.

1.	United Kingdom	**15,675** (22.18%)
2.	Netherlands	**5,391** (7.63%)
3.	France	**4,627** (6.55%)
4.	United States	**4,177** (5.91%)
5.	Italy	**3,894** (5.51%)
6.	Spain	**3,458** (4.89%)
7.	Germany	**3,025** (4.28%)
8.	Poland	**2,473** (3.50%)
9.	Belgium	**2,450** (3.47%)
10.	Ireland	**1,768** (2.50%)

(c) **Website Tables Registration**

EUROCAT Central Registry traces and profiles use of the website tables (since registration began in March 2012) by tracking the number of registrations by country and by type and the number of website table reports generated.

Since registration was initiated in March 2012, 564 individuals from 54 different countries have registered to use EUROCAT's interactive website prevalence tables. Of these individuals 316 requested to be added to the EUROCAT Newsletter mailing list (and provided their emails to enable this).

Table: Registration by Country

Country	Registrations	Country	Registrations
Australia	3	Luxembourg	3
Austria	8	Malta	4
Barbados	1	Moldova	1
Belgium	32	Morocco	1
Canada	6	Netherlands	28
Colombia	1	New Zealand	1
Croatia	5	Norway	4
Cyprus	1	Pakistan	3
Czech Republic	4	Poland	32
Denmark	16	Portugal	6
Egypt	2	Romania	1
Estonia	1	Russia	3
Finland	9	Saudi Arabia	2
France	49	Serbia	1
Germany	28	Serbia and Montenegro	1
Ghana	1	Slovakia	3
Greece	2	Slovenia	2
Greenland	1	Spain	35
Hong Kong SAR China	1	Sudan	1
Hungary	4	Sweden	1
India	9	Switzerland	11
Indonesia	1	Taiwan	1
Ireland	34	Ukraine	1
Israel	1	United Kingdom	135
Italy	32	Uruguay	2
JAPAN	1	USA	25
Lithuania	1	Unknown	2
Total	**564**		

Table: Registrations by Type

User type	Registrations
Affected person / parent of affected person	6
European Commission Official / WHO Official	1
Journalist	8
Medical Geneticist	40
Midwife	9
National Government Official with responsibility for Environment	1
National Government Official with responsibility for Public Health or Health Services	32
Nurse	8
Obstetrician	59
Paediatrician or surgeon	44
Patient Organisation - Abortion provider	1
Patient Organisation - ASBBF	1
Patient Organisation - Charity for British Nuclear Test Veterans	1
Patient Organisation - Down Syndrome	1
Patient Organisation - Down syndrome foundation	1
Patient Organisation - Fondation Jérôme Lejeune	1
Patient Organisation - Spina Bifida	1
Patient Organisation - Spina Bifida and Hydrocephalus	1
Patient Organisation - vice president of hydrocephalus and spina bifida Association Serbia	1
Pharmacovigilance Officer within industry	7
Public Health or Epidemiology Academic	115
Regional / Municipal Government of Health Authority Official	15
Student	111
Other - Actuarial	1
Other - Actuary	1
Other - adult congenital heart disease trainee	1
Other - ANESTESIOLOGIST	1
Other - cardiologist	1
Other - Clinical R&D chief officer	1
Other - Consultant	1
Other - consulting	1
Other - Doctor	1
Other - environmental activist	1
Other - Environmental Consultant	1
Other - Epidemiologist	1
Other - Epidemiologist in Industry	1
Other - Epidemiologist Industry	2
Other - European Parliament	1
Other - Healthcare professionals involved in Primary Prevention of malformations	1
Other - HTA	1
Other - Independent Epidemiology Researcher	1
Other - Industrial toxicologist	1
Other - information officer (EDMP data manager)	1
Other - Insurances	1

Other - Legal Research	1
Other - Local Government	1
Other - Medical communications	1
Other - Medical research	1
Other - NGO	1
Other - Pharmaceutical epidemiology	1
Other - Pharmaceutical industry	1
Other - Pharmaceutical physician	1
Other - Pharmacologist	1
Other - PhD student	2
Other - physician in prenatal medicine	1
Other - Physiotherapist	1
Other - Potential father	1
Other - private foundation for trisomy patients	1
Other - private health consulting firm	1
Other - Psychiatrist	1
Other - Regional Institute in Environment and Health	1
Other - Registered Charity	1
Other - rehabilitation specialist	1
Other - research	2
Other - Research Occupational Therapist	1
Other - Researcher	2
Other - retired academic	1
Other - Scientific consultant	1
Other - secretary of M.C.Addor	1
Other - services provider	1
Other - Social Policy charity Public Affairs Officer	1
Other - urologist and coordinator spina bifida centre of excellence	1
Other - Website programmer	1
Other - Website programmers	1
Other - Academic	44
Total	**564**

Table: Website Table Reports Generated by Month

Month	Reports Generated	
	Members	**Registrants**
Mar-12	0	55
Apr-12	0	97
May-12	0	23
Jun-12	163	185
Jul-12	258	34
Aug-12	27	154
Sep-12	68	162
Oct-12	119	322
Nov-12	67	266
Dec-12	14	251
Jan-13	74	376
Feb-13	94	187
Mar-13	108	214
Apr-13	101	297
May-13	207	221
Jun-13	157	186
Jul-13	64	150
Aug-13	62	166
Sep-13	139	329
Oct-13	130	231
Nov-13	54	245
Dec-13	24	73
Jan-14	54	192
Feb-14	75	242
Mar-14	14	73
Total	**2073**	**4731**

(d) **External Enquiries/Feedback**

- Approximately 70 external enquiries by email were received by EUROCAT Central Registry, since the EUROCAT Joint Action began

WHO?

- Parents of affected individuals, patient organisations, pharmacists, clinicians, registry leaders, epidemiologists, pharma industry, students, government officials, public health officials, reporters

Examples of the types of enquiry received

- Purchase of EUROCAT Data Management Program
- Data requests (website or EUROCAT Central Database)
- To obtain copies of publications/reports
- Sources of case and denominator data, when next year's data will be available
- Coding and registration methodology

- Specific enquires requiring expertise knowledge, e.g. about prenatal screening
- Requesting graphs, tables etc. that could be used and referenced in reports, presentations etc.
- Asking for further information or comment in response to media attention

(e) **Media Interest**

EUROCAT Central Registry makes an effort to trace reference to EUROCAT data/activity in the media and to log this media interest on the member's only section of the EUROCAT website. For related press releases see WP2 report.

Examples include:

EUROCAT Subject: Termination of pregnancy for fetal anomaly after 23 weeks of gestation: a European Register-based study

Related paper
Garne E, Khoshnood B, Loane M, Boyd P and Dolk H (2010). Termination of pregnancy for fetal anomaly after 23 weeks of gestation: a European Register-based study. British Journal of Gynaecology. 117: (6). 660-666.
Media coverage:

2 newspaper articles
Die dritte Dimension belebt die Supernova-Simulation, Natur und Wissenschaft, 19/05/2010 available at http://www.eurocat-network.eu/content/Media-German-Newspaper-Article-1.pdf
Schwangere und Artze spuren den Druck, Natur und Wissenschaft, 19/05/2010 available at http://www.eurocat-network.eu/content/Media-German-Newspaper-Article-2.pdf
EUROCAT Subject: Misinterpretation of TOPFA data on website tables

Media coverage:
Why are there discrepancies in the DoH abortion data? (The Ministry of Truth, 08/03/2013) http://www.ministryoftruth.me.uk/2013/03/08/why-are-there-discrepancies-in-the-doh-abortion-data/
Reports on abortions over minor foetal abnormalities incorrect, says Eurocat (thejournal.ie, 06/02/2013)
http://www.thejournal.ie/reports-on-abortions-over-minor-foetal-abnormalities-incorrect-says-eurocat-784069-Feb2013/
Dozens of abortions carried out on foetuses with minor imperfections such as cleft lip or club foot (Mail Online, 03/02/2013)
http://www.dailymail.co.uk/news/article-2272783/Dozens-abortions-carried-foetuses-minor-imperfections-cleft-lip-club-foot.html
Cleft lip abortions '10 times as common as reported' (The Telegraph, 03/02/2013)
http://www.telegraph.co.uk/health/healthnews/9845780/Cleft-lip-abortions-10-times-as-common-as-reported.html
Number of abortions for fetal "abnormality" vastly underreported in England and Wales (National Right to Life, 06/02/2013)
http://www.nationalrighttolifenews.org/news/2013/02/number-of-abortions-for-fetal-abnormality-vastly-underreported-in-england-and-wales/
UK: Cleft Lip, Club Foot Abortions 10X More Than Reported (LifeNews.com, 05/02/2013)
http://www.lifenews.com/2013/02/05/uk-cleft-lip-club-foot-abortions-10x-more-than-reported/
Major disparity in British abortion statistics revealed (The Irish Catholic, 07/02/2013)
http://irishcatholic.ie/20130207/news/major-disparity-in-british-abortion-statistics-revealed-S30402.html

Abortions for cleft lip '10 times more than recorded' (The Christian Institute, 11/02/2013)
http://www.christian.org.uk/news/abortions-for-cleft-lip-10-times-more-than-recorded/
Health department fails to reveal extent of abortions for correctable deformities (Christian Concern, 07/02/2013)
http://www.christianconcern.com/our-concerns/abortion/health-department-fails-to-reveal-extent-of-abortions-for-correctable-deformit
We must be given an accurate picture of abortion for disability (Life)
http://www.lifecharity.org.uk/lifenewseurocatdisability
27 Irish Babies Killed for Being Disabled (Prolife.ie, 11/02/2013)
http://www.prolife.ie/news/2013/02/11/27-irish-babies-killed-being-disabled
British reports show law abused and more babies aborted for disabilities (Life Institute, 04/02/13)
http://www.thelifeinstitute.net/latest-news/british-reports-show-law-abused-and-more-babies-aborted-for-disabilities/
Reports on abortions over minor foetal abnormalities incorrect, says Eurocat (The Daily Edge, 06/02/2013)
http://www.dailyedge.ie/reports-on-abortions-over-minor-foetal-abnormalities-incorrect-says-eurocat-784069-Feb2013/
UK Parliamentary Business
http://www.publications.parliament.uk/pa/ld201213/ldhansrd/text/130212w0001.htm (12/02/2013)

EUROCAT Subject: Trends in the prevalence, risk and pregnancy outcome of multiple births with congenital anomaly: a registry-based study in 14 European countries 1984-2007.

Related paper
Boyle B, McConkey R, Garne E, Loane M, Addor M-C, Bakker M, Boyd P, Gatt M, Greenlees R, Haeusler M, Klungsoyr Melve K, Latos- Bielenska A, Lelong N, McDonnell R, Metneki J, Mullaney C, Nelen V, O'Mahony M, Pierini A, Rankin J, Rissmann A, Tucker D, Wellesley D and Dolk H (2013). Trends in the prevalence, risk and pregnancy outcome of multiple births with congenital anomaly: a registry-based study in 14 European countries 1984-2007. *British Journal of Gynaecology*

Media coverage:
Channel 4 News (06/02/13) Birth defects linked to twins and triplets double
http://www.channel4.com/news/birth-defects-linked-to-twins-and-triplets-double

ITV News (06/02/13) Risk of birth defects higher for twins and triplets
http://www.itv.com/news/update/2013-02-06/risk-of-birth-defects-higher-for-twins-and-triplets/

Medical Xpress (06/02/13) The number of multiple births affected by congenital anomalies has doubled since the 1980s
http://medicalxpress.com/news/2013-02-multiple-births-affected-congenital-anomalies.html

Science Daily (06/02/13) Number of Multiple Births Affected by Congenital Anomalies Has Doubled Since the 1980s
http://www.sciencedaily.com/releases/2013/02/130205200237.htm

Scotsman (06/02/13) Birth defects are on the rise in twins
http://www.scotsman.com/the-scotsman/health/birth-defects-are-on-the-rise-in-twins-1-2776616

Telegraph (06/02/13) Older mothers driving up birth defect rate
http://www.telegraph.co.uk/health/healthnews/9850091/Older-mothers-driving-up-birth-defect-rate.html

Medical News Today (07/02/13) Multiple Births Affected By Congenital Anomalies Have Doubled Since The 1980s
http://www.medicalnewstoday.com/releases/255939.php

Medi Lexicon (07/02/13) Multiple Births Affected By Congenital Anomalies Have Doubled Since The 1980s
http://www.medilexicon.com/medicalnews.php?newsid=255939

Express (06/02/13) Rise in multiple birth defect rate
http://www.express.co.uk/posts/view/375894/Rise-in-multiple-birth-defects-rate

On Medica (06/02/13) ART link to rise in birth anomalies
http://www.onmedica.com/NewsArticle.aspx?id=745f4b32-5da5-45e6-b7ed-93d74c9ba6e3

Jezebel (06/02/13) Older Mothers Cause Spike in Birth Defect Rate
http://jezebel.com/5982095/older-mothers-possibly-cause-spike-in-birth-defect-rate

Inquisitir (06/02/13) Older Mothers And IVF Pushing Up Birth Defect Rate, Says Study
http://www.inquisitr.com/511189/older-mothers-and-ivf-pushing-up-birth-defect-rate-says-study/

West Welfare Society Territory (06/02/13) Alarming increase of birth defect in Europe
http://www.west-info.eu/alarming-increase-of-birth-defect-in-europe/

El Mundo (06/02/13) El número de niños con defectos congénitos se duplica
http://www.elmundo.es/elmundosalud/2013/02/06/noticias/1360153135.html

Reuters (07/02/13) Birth defects in multiples on the rise: study
http://uk.reuters.com/article/2013/02/07/us-birth-defects-idUKBRE91616T20130207

The Telephone (10/02/13) Are Test Tube Babies at Greater Health Risk
http://www.telegraph.co.uk/women/mother-tongue/9860773/Are-test-tube-babies-at-greater-health-risk.html

(f) **Web-based Evaluation Survey**

EUROCAT aimed to obtain constructive feedback (relating to the EUROCAT Joint Action January 2013 through December 2013) about its website and other outputs via a web-based evaluation survey tool of 10 short questions taking no longer than 10 minutes to complete (Annex 2) in order to determine if EUROCAT was reaching its intended user and if EUROCAT's output is appropriate for that user.

In addition to a pop-up invitation to take the survey upon visiting the EUROCAT website, a link to the survey was disseminated via the EUROCAT Central Registry mailing list (continuously updated throughout the timeframe of the EUROCAT Joint Action) and the EUROCAT Newsletter. EUROCAT Central Registry encouraged further dissemination of the survey link by the broader EUROCAT membership via reminders in EUROCAT communication emails and via an evaluation strategy presented at the final EUROCAT Registry Leaders' Meeting in Zagreb, Croatia, June 2013.

Responses to Web-based Evaluation Survey

The web-based evaluation survey received 133 respondents of which 36 requested to receive the EUROCAT Newsletter in the future and provided their email address to enable this.

1. Which professional or user perspective represents your interest in EUROCAT?

	Answer	Response	%
1	Affected person or parent of affected person	0	0%
2	National government official with responsibility for public health or health services	4	3%
3	National government official with responsibility for rare diseases	3	2%
4	National government official with responsibility for environment	0	0%
5	Regional or municipal government or health authority official	7	5%
6	European Commission, WHO or other international organisation official	0	0%
7	Hospital or health service manager	2	2%
8	Paediatrician	12	9%
9	Obstetrician	12	9%
10	Medical geneticist	12	9%
11	Nurse	1	1%
12	Midwife	4	3%
13	Public health or epidemiology academic	29	22%
14	Other academic	10	8%
15	Pharmacovigilance officer within industry	0	0%
16	Student	9	7%
17	Other (please specify)	28	21%
	Total	133	100%

Other responses included:

Other (please specify)	Response	Other (please specify)	Response
Programmer	1	Anesthesiologist	1
Registry leader of regional CA register	2	Paediatrician and Medical geneticist	1
Midwife studying for MSc	1	Regional government official charge of sanitary responsibility	1
EUROCAT registry leader (public health doctor)	1	Pediatric neurologist	1
Working for disability charity	1	Patient organisation	1
Corporate Researcher in Epidemiology	1	Research	2
Non-profit staff member	1	Urologist and coordinator of spina bifida centre of excellence in France	1
Epidemiologist	1	Medical sonographer	1
Pediatric surgeon	1	Public health, working in NGO	1
Charity	1	Teratology information service scientist	1
Clinical radiologist	1	Researcher in CA registry	1
Associate Dean of Undergraduate Medicine and Professor of Medical Education	1	Private company involved in prevention of birth defects intitiatives/programmes	1
Laboratory clinic	1	Neonatologist	1
		Total	28

2. Which country do you live in?

Answer	Response	%		Response	%
Argentina	2	2%	Hungary	2	2%
Armenia	1	1%	India	1	1%
Austria	3	2%	Ireland	10	8%
Bahamas	1	1%	Italy	8	6%
Belgium	8	6%	Latvia	3	2%
Belize	1	1%	Netherlands	4	3%
Bosnia and Herzegovina	1	1%	Norway	1	1%
Canada	1	1%	Portugal	1	1%
Chile	2	2%	Saudi Arabia	3	2%
Croatia	6	5%	Spain	14	11%
Cyprus	1	1%	Switzerland	1	1%
Denmark	5	4%	The former Yugoslav Republic of Macedonia	2	2%
Finland	1	1%	Turkey	1	1%
France	12	9%	Ukraine	1	1%
Germany	7	5%	United Kingdom of Great Britain and Northern Ireland	25	19%
Greece	1	1%	United States of America	1	1%
			Total	131	100%

3. Which one or more of the following topics is relevant to you?

Answer	Response	%
Prevalence of congenital anomalies	106	82%
Causes and primary prevention of congenital anomalies	87	67%
Prenatal screening for congenital anomalies	83	64%
Other (please specify)	22	17%

Other (please specify)	
Management and outcomes of congenital disorders	neonatal screening
EUROCAT documents	epidemiologic research
studies	Survival; Time to surgery;
Outcome	Coding of congenital anomalies
cluster investigation protocols	Diagnosis of C.A.
termination of pregnancy after prenatal screening	surveillance system
Effectiveness of interventions	Cancer research
pregnancies interruption for congenital anomalies	termination of pregnancy for foetal anomaly
mortality and morbidity	Statistics of Congenital anomalies
Asthma Drugs in Pregnancy	registration of exposure to medication in pregnancy
methodology	

4. Which one or more types of congenital anomalies are of particular interest to you in terms of your need for information from EUROCAT?

Answer	Response	%
Congenital Heart Defects	73	57%
Neural Tube Defects	78	60%
Orofacial Clefts	55	43%
Chromosomal including Down Syndrome	72	56%
Genetic Syndromes	64	50%
Abdominal Wall Defects (e.g. gastroschisis)	48	37%
Limb Defects	48	37%
Other (please specify)	39	30%

Other (please specify)	Response
All	16
Renal	3
Genital anomalies & sex development	2
None	2
Rarer anomalies/entities	2
All the anomalies listed above but just in the cases that require immediate hospitalization in a neonatology unit or NICU after birth	1
All. I am the author of a textbook on Medical Embryology	1
Cluster investigation protocols	1
Cystic fibrosis	1
Esophageal atresia	1
Hearing Loss	1
Hypospadias	1
Inborn errors of metabolism	1
Neurogenetica	1
Overgrowth syndromes	1
Respiratory malformations	1
Vascular defect in neural t	1

5. Which one or more of the following risk factors for congenital anomaly are of particular interest to you in terms of your need for information from EUROCAT?

Answer	Response	%
Folic Acid	61	54%
Medicinal Drug Exposures During Pregnancy	67	60%
Environmental Pollutants	53	47%
Genetic factors	61	54%
Other (please specify)	18	16%

Other responses include:

Other (please specify)	Response
Lifestyle (alcohol, smoking, obesity, diabetes)	6
All types	2
Maternal risk factors	1
Radiation exposure	1
Sociocultural	1
Stress	1
Drug use during pregnancy	1
I'm not interested in the risk factors	1
Policies of abortion	1

6. Please rate how useful you have found the following outputs from EUROCAT in the period 2011-2013

Question	Very Useful	Useful	Not Useful	Did Not Know About It	Not Relevant to Me	Total
Interactive Website Prevalence Tables	68	31	2	8	4	113
Website Prenatal Diagnosis Tables	45	44	2	12	10	113
Website Perinatal Mortality Tables/Key Public Health Indicators	43	39	4	17	10	113
Annual Statistical Monitoring Reports Concerning Time Trends and Clusters	50	32	7	12	12	113
Guide 1.3/1.4: Instruction for the Registration of Congenital Anomalies	47	27	7	14	18	113
EUROCAT Special Report (2012): Congenital Anomalies are a Major Group of Mainly Rare Diseases	53	32	5	14	9	113
EUROCAT Publication List	47	39	7	10	10	113
EUROCAT Newsletter (if you would like to receive the Newsletter please include your email address below)	34	37	5	30	7	113

7. Have you received information about or from one or more EUROCAT member registries in your country?

Answer	Response	%
Yes	48	43%
No	38	34%
Not applicable	25	23%
Total	111	100%

8. Do you know about the European Scientific Symposium on the Prevention of Congenital Anomalies which EUROCAT organises every two years?

27 respondents indicated they would like to find out more information about future symposia and provided their email addresses to enable this.

Answer	Response	%
Yes I participated in Antwerp, Belgium (2011) or Zagreb, Croatia (2013)	34	31%
No, but I would like to find out more and perhaps participate in future (if so please provide your email address below)	38	35%
I would not be interested or able to participate	38	35%
Total	110	100%

9. Do you find the EUROCAT website easy to use?

Answer	Response	%
Yes	95	86%
No (if no please specify why)	15	14%
Total	110	100%

Why responses included:

No (if no please specify why)
I just want to see fetal anomalies comparisons across countries. Rates for Ireland of specific anomalies and how Irish rates compare to other counties. Too much information here and hard to find exactly what I want
Access to prevalence tables are not so easy to find when we want to specify own search criteria.
I've yet to use it - this survey popped up on entering the site before I had a chance to look at anything else!
Need quick search box
The menu structure. It is not always clear where to find certain information, or to see what is available, for instance, workshop abstracts. Also, the option to download these abstracts would be appreciated.
I am in first time in your webs
I don't know yet
Yes for professional; no for people not specialists of congenital anomalies
Not always easy to find what you want. The menu is difficult to click on
Frames are awkward, contact information difficult to get from pdf-files
Mainly because of the menu structure
Difficult to read and make comparisons between countries e.g. Ireland has three registries

10. Please give any further comments you may have about EUROCAT. In particular what information you would like to see on the EUROCAT website.

Positive feedback

"Databases such as this are invaluable. I am researching for a small assignment on public health campaigns on folic acid and neural tube defects and EUROCAT data is often quoted."

"Very interesting surveillance system. We are using your reporting system as a model for our reports on prevalence rates."

"It is really helpful to find all these statistical data, so trustworthy and easy. Thanks for your efforts."

"Been of great use for research purposes and widening my general knowledge on other topics."

"The actual set up is fine; please do not change it. I appreciate enormously the excellent help and information I get from EUROCAT ANTWERP from Vera Nelen and Guy Thys."

"I found this site useful and was able to answer the questions I wished to research."

"EUROCAT is doing a great job in a very important field!"

"Website is excellent. Continued access to a wide variety of information on congenital anomalies."

"The website is comprehensive and very easy to navigate."

"Congratulations. Our surveillance system was established in 2012. Thanks for the sharing. Appreciated."

"The pages look good and it is easy to find information."

"Well structured and easy to access. Very important work."

"I think is very well done."

"Fully satisfied."

"No further comment apart from my thanks to all EUROCAT Central Registry's staff for all their support and concerns. Eva Bermejo-Sanchez."

"The website is OK for me."

"The EUROCAT website is an essential resource. The new style of tables are well-designed and much easier to use."

"Information on EUROCAT website is very complete and of great interest. Queries are helpful."

"EUROMEDICAT is a great initiative. Improved ascertainment of medication exposure data from multiple sources across all member registries is an important goal."

"The updated website is a great improvement on the earlier one. I particularly appreciate presentation of data by country as well as registry, and of pregnancy outcomes for non-chromosomal as well as total anomalies."

"This is an excellent website. The older format of the prevalence tables was simpler to use but I guess the current format allows more interaction."

"The prevalence tables have become much easier to use - eg. figures by country etc - in the past year."

"Basically no comment or suggestions on the EUROCAT website. About EUROCAT as such, I would strongly propose to continue the program especially in my country to widen it to include all of Austria."

"It's a very good site, easy to access and informative."

"Great to have this information available but when it was less interactive it was easier to find what I wanted."

More information on or the addition of ……

"While it will be very difficult to monitor, to get the full picture of the incidence of neural tube defects, it would be good to know the number of spontaneous, early losses (miscarriages; stillbirths) from before 20 weeks of gestation as well. It may improve governments willingness to invest in primary prevention measures."

"detailed info regarding drugs and congenital anomalies"

"prevalence of rare or very rare anomalies"

"detection rates by antenatal ultrasound per country"

"EUROCAT Data Management Program"

"prevalence Data"

"the etiology of congenital anomalies and on the follow up of cases"

"congenital hearts defect and specially in prenatal diagnosis in ultrasound of 11- 14 weeks"

"graphical information"

"please could Cystic Fibrosis be included - unless I cannot see the data at present"

"longitudinal data on the impact of congenital anomalies in adult disability and dependency"

"prenatal screening policies in the different registries"

"the experiences, attitudes and knowledge of parents on the application of preventive public health action in prenatal pregnancy"

"prenatal detection rates"

"Ideally I would like to see rates for affected individuals, but understand that this could be difficult."

"heart defects prevalence"

"WHO official definition of congenital anomalies that can be proven useful for scholars working about perinatal health and need to give definitions in their work"

"free access to full text articles written by the Eurocat members"

"PowerPoint Presentations regarding specific group of congenital anomalies"

"genetic causes of neurogenetic diseases"

"The new information for neurogenetic diseases"

"Could differing prevalence rates across Europe be shown by maps? Maybe these could build into a European Atlas of congenital anomalies"

"Interactive prevalence tables"

"Composite report on the state of evidence in Europe on congenital anomaly and alcohol, tobacco, obesity, nutrition etc and recommendations for action. Recommendation on the integration of prevention of congenital anomaly into public health strategies on alcohol, tobacco, maternity strategy, womens health etc. European consensus statement on prevention of congenital anomaly???"

"better compatibility of the european data: More information about the coding of congenital anomalies in ICD 10 and talk about the coding in ICD 11"

"Improvement in capture of eye anomalies by the registers"

"spain prevalences"

"prenatal detection rate of corpus callosum agenesis"

"I would like to be able to access more of the denominator data too, such as maternal age in the member registries, by individual year."

"Live birth prevalence of congenital anomalies [to be made available without a special query as for total prevalence]"

"If it becomes possible to provide data/estimates for isolated (and multiple) malformations, this would be particularly helpful."

"I'm specially interested in major and minor fetal anomalies rates, because I develop my job in Prenatal Ultrasound Diagnosis."

"I'm interested mainly in European records of fetal anomalies and prenatal diagnosis rates"

"In what way is now possible to include in EUROCAT register small regions such as the Canton of Tuzla in Bosnia and Herzegovina, which has about 500,000 residents and about 4500-5000 births a year."

"For the development/implementation of effective prevention of congential anomalies, it would be helpful to have access to persons and institutions involved in practical actions and (all types of) evaluation reports of those actions. This would require information gathering beyond the EUROCAT region and networks. Some other national and European institutions are already involved in prevention (like IF-SBH, EFCNI, EURORDIS, PfL. etc) : so a first step might be to see how existing information could be brought together?"

"It would be good to have a link for newly publications and statistics regarding congenital anomalies as well as recent public health measures for congenital anomalies prevention performed worldwide"

"local national reference centers"

Constructive Feedback

"I think it is not a very "visual" website"

"For a paediatric clinic to much information."

"I would suggest to open the registry to more interested professional outside Europe"

"It would be useful to have country icons on home page with links to local registries so visitors can get information on their own country in their own language"

"There is a "double entry" to perform prevalence tables. One selecting "Prevalence Tables" (left menu) and the second on selecting the anomaly (upper right of web page). We don't find the same information doing this (at a first glance). Searching a little more we found how to do it, and it works! Thank you very much for this great job."

"Looking at your website after reading news article in the Daily Telegraph, for background information on abortion and disability. A little less jargon on the website would help, not everyone coming here is a statistician. Thank you"

"Dear all, the website is great. But I could not mention my registry leadership in the test or when registering on the website. All the best!"

"But as far as my work is concerned I would like to have a quick search box in selecting Anomalies (after entering through ID and password), rather than selecting it in the list which consumes time. Thank u."

(g) Evaluation of EUROCAT Symposiums

Belgian Symposium in Antwerp (June 2011)

The 17th of June, the 11th international Eurocat symposium took place in the city of Antwerp, Belgium. The main topics of the symposium were prenatal and preconceptional care, environmental risks and long term outcome. The venue was the Antwerp provincial building. The symposium was organized by Eurocat and the province of Antwerp.

The symposium was advertised in the website www.Eurocat2011.com , also used for online registration. Evaluation showed the website had 1522 visitors, 2963 visits and 9367 pages visited. 1/3 visits came directly through the address, 1/3 through search engines and 1/3 through referral from other websites. Most visitors came from Belgium, followed by the UK and the Netherlands and France. There were 86 visitors from USA.

The symposium was a success with 232 participants from 25 countries. Participants came mainly form Belgium, the UK and the Netherlands but also from Saudi Arabia and Canada. The call for abstracts resulted in 40 abstracts that were orally presented or posters.

The symposium was evaluated with an E-mail questionnaire. We received 110 evaluation questionnaires from 224 participants, a response of 49%. 92 % evaluated the scientific value as good or very good, 96% for the choice of topics. For 28% it was too difficult. The organisation was good for 90% or more of the responders. Only translation to Dutch and accessibility scored lower.

Croatian Symposium in Zagreb (June 2013)

How did you evaluate:	**Poor**	**Satisfactory**	**Good**	**Excellent**	**Total number of answers**
Choice of topics	○	○	21 (51,3)	20 (48.7%)	41
Difficulty	2 (4,8%)	7 (17,1%)	19 (46,3%)	13(31,7%)	41
Scientific value	○	○	7 (18,9%)	30 (81,1%)	37

STYLE:					
How did you evaluate the:	**Poor**	**Satisfactory**	**Good**	**Excellent**	**Total number of answers**
Welcome	○	2 (5%)	14 (34,1%)	25 (60,9%)	41
Timing	3 (7%)	8 (18,6%)	10 (23,2%)	22 (51,3%)	43
Accessibility	○	○	13 (32,5%)	27 (67,5%)	40
Conference rooms	2 (4,8%)	3 (7,1%)	9 (21,4%)	28 (66,7%)	42
Lunch	○	○	13 (30,9%)	29 (69,1%)	42
Coffee breaks	○	○	13 (32,5%)	27 (67,5%)	40

Conference map	1 (2,3%)	2 (4,6%)	17 (39,5%)	23 (53,6%)	43
Poster placing	◯	5 (11,9%)	16 (38,1%)	21 (50%)	42

Do you have additional remarks?

The sound in conference room (which is beautiful, beside that) was quite poor so we couldn't hear speakers properly.

Good variety at the conference, with presentation topics organsied well to create a base level of understanding for varying backgrounds attending the conference.

Timekeeping should be enforced more readily. One of the presentations took much more time than allowed and as a result, there was hardly any time left for the poster session.

Very good conference. Probably a few too many presentations within the time available as questions at the end of each session great for timing but not ideal for the speakers.

(h) Independent Evaluation

A questionnaire (created by an external subcontractor and accessed via an online survey tool) was employed to focus on perception of "value" of the EUROCAT outputs/outcomes by targeted end-users (A list of the questions asked is provided for quick reference in Annex 3 and the full report provided by the independent evaluation subcontractor is provided in Annex 4).

EUROCAT Independent Evaluation Panel Members

191 individuals received the EUROCAT Independent Evaluation Questionnaire Survey link. Of those 93 responded giving a 49% response rate.

EUROCAT Member Registry Leaders

52 EUROCAT Member Registry Leaders (and the former EUROCAT President, who retired in 2013) were asked to be part of the evaluation panel and were in receipt of the questionnaire. Respondents are listed in the table below and included 19 of 31 Full Member Registry Leaders, 2 of 6 Associate Member Registry Leaders, 4 of 9 Affiliate Member Registry Leaders, 2 of 2 Applicant Member Registry Leaders and 4 of 4 World Affiliate Member Registry Leaders, in addition to the former EUROCAT President.

Only 22 of the EUROCAT registry leader respondents identified themselves in the category of registry leader. Therefore it was only possible to exclude the 22 when analysing responses separately.

Name	**Registry**
Full Members	
Ester Garne	**Odense (Denmark)**

Babak Khoshnood	Paris (France)
Bob McDonnell	Dublin (Ireland)
Hermien De Walle	N Netherlands
Elisa Calzolari	Emilia Romagna (Italy)
Marie-Claude Addor	Switzerland
Ingeborg Barisic	Zagreb (Croatia)
Miriam Gatt	Malta
Larraitz Arriola	Basque Country (Spain)
Anke Rissmann	Saxony Anhalt (Germany)
Martin Haeusler	Styria (Austria)
Mary O'Mahony	Cork and Kerry (Ireland)
Wladimir Wertelecki	Ukraine
Catherine Rounding	Thames Valley (UK)
Diana Wellesley	Wessex (UK)
Judith Rankin	Northern England (UK)
Judit Beres	Hungary
Carmel Mullaney	SE Ireland
Oscar Zurriaga	Valencia Region (Spain)
Associate Members	
Annukka Ritvanen	Finland
Maria Luisa Martinez Frias	Spain Hospital Network (Spain)
Affiliate Members	
Goradz Rudolf	Slovenia
Vladimir Egorov	Moldova
Joan Morris	National Down Syndrome Cytogenic Register (UK)
Katya Kovacheva	Bulgaria
Applicant Members	
Helga Sol Olafsdottir	Iceland
Elena Szabova (no longer proposed RL)	Slovakia

World Affiliate Members	
Boris Groisman	Argentina
Barry Norman	New Zealand
Saeed Dastgiri	Iran
Ahmed Kurdi	Saudi Arabia
Former EUROCAT President	
Lorentz Irgens	

Individuals nominated by EUROCAT Member Registry Leaders

52 EUROCAT Member Registry Leaders were asked to nominate (and obtain prior agreement of intent to respond), 1 national relevant representative from a (i) Public Health Authority, (ii) Patient Organisation (or patient), (iii) Department of Health e.g. in Rare Diseases field, and also to nominate a relevant national clinician. 27 EUROCAT Member Registry Leaders provided a total of 94 nominees and obtained confirmation of intent to respond. 34 of those 94 responded and are detailed in the table below.

EUROCAT Registry Leader who provided the nominees (EUROCAT Registry)	**Respondent Name**	**Respondent Type**	**Affiliation**
Babak Khoshnood (Paris, France)	Veronique Goulet	Public Health Authority	InVS – French Institute for Public Health Surveillance
	Francois Goffinet	Clinician (Obstetrician)	Director of INSERM U953
	Sylvie Rey	Department of Health (Medical Epidemiologist)	Chargée de mission auprès de la Sous-directrice Direction de la Recherche, des Etudes, de l'Evaluation et des Statistiques Sous-Direction "Observation de la Santé et de l'Assurance Maladie"

Mary O'Mahony (Cork and Kerry, Ireland)	Keelin O'Donoghue	Clinician (Consultant Obstetrician and Gynaecologist)	Cork University Maternity Hospital
	Richard Greene	Clinician (Consultant Obstetrician and Gynaecologist)	Health Service Executive, National Perinatal Epidemiology Centre, University College Cork
	Elizabeth Keane	Public Health Authority	Health Service Executive South (Cork and Kerry)
Carmel Mullaney (SE Ireland)	Orlaith O'Reilly	Public Health Authority	Department of Public Health, Health Service Executive Southeast Area, Kilkenny, Ireland
	Eibhlin Mulroe	Patient Organisation	Irish Platform for Patient Organisations, Science and Industry (IPPOSI)
Vera Nelen (Antwerp, Belgium)	Jenneke van den Ende	Clinician	Centre of Medical Genetics Antwerp
	Dirk Wildemeersch	Public Health Authority	Head of Division, Flemish Agency for Health and Care
Katia Kovacheva (Bulgaria)	Angelika Velkova	Public Health Authority	Faculty of Public Health, Medical University of Pleven
	Victoria Atanasova	Clinician (Neonatologist Consultant)	Neonatal Clinic
Antonin Sipek (Czech Republic)	Pavel Calda	Public Health Authority	Section of Foetal Medicine of the Czech Society of Gynaecology and Obstetrics

	Milan Macek	Department of Health	Czech Coordination Board for Rare Diseases
Annukka Ritvanen (Finland)	Mika Gissler	Public Health Authority	THL National Institute for Health and Welfare
	Carola Saloranta	Clinician (Geneticist)	Helsinki University Hospital, Department of Clinical Genetics/Women's Hospital, Prenatal Diagnostic Unit
Anke Rissmann (Saxony-Anhalt, Germany)	Matthias Schiener	Public Health Authority	Department of Public Health, Ministry of Labour and Social Affairs Saxony-Anhalt
	Claudia Spillner	Clinician (Paediatrician)	Early Intervention Centre, Magdeburg, Saxony-Anhalt
	Jenetzky Ekkehart	Patient Organisation and Clinician	Member of German Network for Congenital Uro-Rectal malformations CURE-Net http://www.cure-net.de/
Miriam Gatt (Malta)	Louisa Grech	Patient Organisation	Equal Partners Foundation
Ahmed Kurdi (Saudi Arabia)	M.A. Majeed-Saidan	Clinician	Prince Sultan Military Medical City
	Maha Al-Rakaf	Clinician	Prince Sultan Military Medical City
	Amal Al-Hashem	Clinician	Prince Sultan Military Medical City
Anna Barakova	Tana Foltanova	Patient Organisation	Slovakian Orphanet

(Slovakia)			Representative
Larraitz Arriola (Basque Country, Spain)	Isabel Portillo	Department of Health	Prenatal Screening Programme Basque Country
Maria-Luisa Martinez-Frias (Spain Hosptial Network, Spain)	Pilar Soler Crespo	Department of Health	General Sub-Directorate of Quality and Cohesion
Oscar Zurriaga (Valencia Region, Spain)	Herme Vanaclocha	Public Health Authority (Deputy Director of Epidemiology and Public Health Surveillance)	Regional Public Health Administration, Valencia, Spain
	Consuela Garcia Vicent	Clinician (Senior Registrar Paediatric Department)	Consorcio Hospital General Universitario de Valencia, Spain
	Claudia Delgado	Patient Organisation	Director of the Spanish Rare Diseases Patients Association (FEDER), Madrid, Spain
	Antonio Perez Aytes	Department of Health	Representative of the Valencia Region in the Spanish National Strategy on Rare Diseases, Hospital Politecnico Universitario La Fe (Valencia, Spain)
Wladimir Wertelecki (Ukraine)	Viktor Dolhov	Clinician	Khmelnytsky medical genetic centre, clinical geneticist
Catherine Rounding (Thames Valley, UK)	Ron Gray	Public Health Authority	National Perinatal Epidemiology Unit, University of Oxford

Ingeborg Barisic (Zagreb, Croatia)	Miroslav Dumic	Clinician	Professor emeritus of Pediatrics, University of Zagreb
Ieva Grinfelde (Latvia)	Antra Valdmane	Department of Health	Ministry of Health, Health Care Department

Individuals External to the EUROCAT Network (Central Registry and Member Registries)

44 relevant experts (external to EUROCAT Membership) were invited to participate in the evaluation panel. 27 responses (detailed in the table below) were received from individuals external to the EUROCAT Network Membership.

Name	**Affiliation**
Peter Soothill	Emeritus Professor of Maternal and Fetal Medicine at the University of Bristol (UK)
Sukhwinder Kaur	Representative of WHO Collaborating Centre for Nursing and Midwifery Development (India)
Briege Lagan	Representing FP7 – funded EUROmediCAT Project (Safety of Medication Use in Pregnancy)
Peter Mossey	Director of WHO Collaborating Centre for Public Health Issues on Congenital Anomalies and Technology and Professor of Craniofacial Development, Dundee University Dental School (UK)
Manual Antonio Fernandez Fernandez	Representative of the European Paediatric Neurology Society
Jana Lepiksone	Latvian National Representative of Orphanet
Ruitai Shao	Responsible Officer for relevant WHO Collaborating Centres, Programme Management Advisor, Department of Management of Non-Communicable Diseases
Julio Cesar Leite	Responsible Physician for the Birth Defects Monitoring Program at the Genetic Medical Services in Latin America
Ysbrand Poortman	World Alliance of Organisations for the Prevention and Treatment of Congenital Anomalies
Mittal Suneeta	Director of WHO Collaborating Centre for Human Reproduction (India), Director and Head of the Department of Obstetrics and Gynaecology

Carol Bower	Australian member of ICBDSR/Western Australian Register of Developmental Anomalies
Renee Jopp	Representative of the International Federation for Spina Bifida and Hydrocephaly
Melba Gomes	WHO Geneva
Bernadette Modell	Representing the Global Burden of Disease Project
Lolkje De Jong-van den Berg	Representing FP7 – funded EUROmediCAT Project (Safety of Medication Use in Pregnancy)
Marco Martuzzi	WHO Regional Office for Europe, European Centre for Environment and Health (Germany)
Sue Jordan	Representing FP7 – funded EUROmediCAT Project (Safety of Medication Use in Pregnancy)
Cristina Rusu	Romanian National Representative of Orphanet
Domenica Taruscio	Representing EUROPLAN (European Project for Rare Diseases National Plans Development) and EPIRARE (European Platform for Rare Diseases Registries) projects
Elisabeth Mason	Director of WHO's Department of Maternal, Newborn, Child and Adolescent Health
Christel Nourissier	Representative of EURORDIS (European Rare Disease Organisation)
Pierpaolo Mastroiacovo	Director of WHO Collaborating Centre on the Control of Birth Defects (Italy) also Director of the Central Office of the International Clearinghouse for Birth Defects Surveillance and Research (Italy)
Tarik Derrough	Expert Vaccine Preventable Diseases, European Centre for Disease Control (Sweden)
Alison MacFarlane	UK representative of EUROPERISTAT Project (Better Statistics for Better Health for Pregnant Women and their Babies)
Jan Cap	Slovakian representative of EUROPERISTAT Project (Better Statistics for Better Health for Pregnant Women and their Babies)
Frances Murphy	Contact A Family UK (Patient Organisation)
Anna David	University College London - Institute for Women's Health (UK)

Responses

Q1. List of countries

The 93 respondents were distributed across 33 different countries as detailed in the table below.

Answer	Including Registry Leaders		Excluding Registry Leaders	
	Response	%	Response	%
Argentina	1	1%		
Australia	1	1%	1	1%
Austria	1	1%	1	1%
Belgium	3	3%	3	4%
Brazil	1	1%	1	1%
Bulgaria	3	3%	2	3%
Croatia	2	2%	2	3%
Czech Republic	2	2%	2	3%
Denmark	1	1%		
Finland	3	3%	2	3%
France	5	5%	4	6%
Germany	5	5%	4	6%
Hungary	1	1%		
Iceland	1	1%	1	1%
India	2	2%	2	3%
Iran, Islamic Republic of...	1	1%	1	1%
Ireland	8	9%	5	7%
Italy	3	3%	2	3%
Latvia	2	2%	2	3%
Malta	2	2%	1	1%
Netherlands	3	3%	2	3%
New Zealand	1	1%	1	1%
Norway	1	1%	1	1%
Republic of Moldova	1	1%	1	1%
Romania	1	1%	1	1%
Saudi Arabia	4	4%	4	6%
Slovakia	3	3%	3	4%
Slovenia	1	1%	1	1%

Spain	10	11%	7	10%
Sweden	1	1%	1	1%
Switzerland	4	4%	3	4%
Ukraine	2	2%	1	1%
United Kingdom of Great Britain and Northern Ireland	13	14%	9	13%
Total	93	100%	71	100%

Q2. Which of the following best describes who you represent (select one option)?

Answer	Response	%
EUROCAT Registry Leader	22	24%
Public Health Authority	11	12%
Paediatrician or Paediatric Surgeon	5	5%
Obstetrician	8	9%
Nurse or Midwife	2	2%
Medical Geneticist	9	10%
Other Healthcare Professional (Please Specify)	6	6%
Patient or Patient's Family	1	1%
Patient Organisation	6	6%
Academic or Researcher	12	13%
Regional/National Department of Health	3	3%
European Commission	1	1%
WHO or Other International Organisation	3	3%
Other (Please Specify)	4	4%
Total	93	100%

Other Healthcare Professional (Please Specify)	Other (Please Specify)
Fetal Medicine	Clinical geneticist
Researcher	State birth defects register
Neonatologist	Italian National Institute of Health
pharmacoepidemiologist	world affiliated member

Public Health Researcher	
Consultant in Public Health Medicine	

Q3. Based on what you currently know about EUROCAT……..Would you recommend all new registries (registering cases of CA) become a member of the EUROCAT Network?

Answer	Including Registry Leaders		Excluding Registry Leaders	
	Response	%	Response	%
Yes	81	95%	60	94%
No	4	5%	4	6%
Total	85	100%	64	100%

Q4. On a scale of 1-5, to what extent is EUROCAT's ongoing CA surveillance important in providing information on the following? (select one option for each statement, 1 = Not useful, 5 + Very useful)

Question	1	2	3	4	5	Total Responses
Detection and investigation CA time clusters	0	3	9	39	42	93
Early warning of increases in prevalence of CA	1	4	7	35	46	93
Environmental risks	1	5	25	40	22	93
Epidemiological data about rare diseases (rare CA)	1	1	13	33	45	93
Impact of prevention measures in decreasing CA prevalence	2	2	15	45	29	93
Mortality related to CA	2	3	22	35	31	93
Prenatal screening	3	4	13	38	35	93
Prevalence of CA	0	2	3	19	69	93
Safety of medication use during pregnancy	0	5	20	41	27	93
Verifying the absence of CA time clusters	0	3	22	37	31	93

Excluding respondents who identified themselves as EUROCAT member registry leaders.

Question	1	2	3	4	5	Total Responses
Detection and investigation CA time clusters	0	2	9	32	28	71
Early warning of increases in prevalence of CA	1	4	4	30	32	71
Environmental risks	1	4	17	30	19	71
Epidemiological data about rare diseases (rare CA)	1	1	10	28	31	71
Impact of prevention measures in decreasing CA prevalence	2	2	14	35	18	71
Mortality related to CA	2	3	18	28	20	71
Prenatal screening	3	4	10	28	26	71
Prevalence of CA	0	2	3	16	50	71
Safety of medication use during pregnancy	0	5	13	29	24	71
Verifying the absence of CA time clusters	0	2	17	31	21	71

Q5. Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local EUROCAT registry)? (select one option)

Answer		Response	%
Yes		71	76%
No		22	24%
Total		93	100%

Excluding respondents who identified themselves as EUROCAT member registry leaders.

Answer		Response	%
Yes		49	69%
No		22	31%
Total		71	100%

Q6. For what purpose did you use this information or data? (tick as many options as apply)

Answer	Including Registry Leaders		Excluding Registry Leaders	
	Response	%	Response	%
Comparing between countries	58	82%	38	78%
Comparing between Registries in the same country	26	37%	14	29%
Improving coding and classification methodology	22	31%	11	22%
Informing clinical practice	31	44%	21	43%
Informing national policy	34	48%	23	47%
Informing regional or local policy	24	34%	14	29%
Research or publications (please provide examples)	24	34%	15	31%
Understanding CA at a European level	44	62%	28	57%
Other (please specify)	6	8%	4	8%

Research or publications (please provide examples)	Other (please specify)
Euro-Peristat report (non RL)	Medicolegal (non RL)
Research projects on: small intestinal atresia; congenital diaphragmatic hernia; hirschsprung's disease	Used to see what services our organisation would need to provide as intervention and support (non RL)
Used as major data source by Congenital Expert Group of the 2010 Global Burden of Disease exercise, and for March of Dimes 2006 report on Global Epidemiology of Birth Defects (non RL)	Understanding CA at the Global level (non RL)
Changing prevalence of CCAMs	discovering prevalence of certain anomalies

EUROCAT and local collaborative research	investigation of an apparent cluster
CA prevalence, Prenatal markers of chromosomal anomalies (non RL)	Attempted study of work linked chemical exposure (non RL)
I always cite EUROCAT as the gold standard for CA surveillance (non RL)	
prenatal diagnosis and prevalence of CA (non RL)	
in collaboration (non RL)	
The last one was on publications of orofacial clefts and also on NTD. Screening also	
Used for quick access to relevant published CA papers and reports	
we use EUROCAT data on many of our research and publications (non RL)	
trends in neural tube defects	
comparing methods of different registers (non RL)	
Mainly students and doctors undertaking research studies/assignments	
Author and Coauthor (non RL)	
IMER Annual Report	
frequency of CA after IVF (non RL)	
risks of AED-drugs on CAs (non RL)	
Jordan S. 2010 'Pharmacology for midwives: the Evidence Base for Safe Practice' Palgrave/ Macmillan, Basingstoke 2nd edition ISBN-13: 978-0-230-21558-0 pp. 486 Adopted in most midwifery departments in UK, Australasia, Malta. Translated into Indonesian & Faris Morgan M., de Jong van den Berg L., Jordan S. 2011 Drug Safety in Pregnancy: Monitoring congenital anomalies Journal of Nursing Management. 19, 305-310 (non RL)	
brief summary of EUROCAT publication in local newsletter to clinical staff	

Time of TOP between countries for example (non RL)	
Publications about CHD (non RL)	
Mortinatality (non RL)	

Q7. On a scale of 1-5, how useful were the following for assessing EUROCAT information? (select one option for each you have used, 1 = Not useful, 5 = Very useful, Blank = Not used)

Question	1	2	3	4	5	Total Responses
Annual Statistical Monitoring Reports	2	4	9	17	29	61
EUROCAT Central Registry team	4	8	10	20	15	57
EUROCAT newsletter	4	8	12	21	13	58
EUROCAT scientific journal publications	1	2	2	27	34	66
EUROCAT website including prevalence tables	2	2	3	12	46	65
Your local EUROCAT Registry	2	1	7	13	32	55

Excluding respondents that identified themselves as EUROCAT member registry leaders.

Question	1	2	3	4	5	Total Responses
Annual Statistical Monitoring Reports	2	4	5	10	19	40
EUROCAT Central Registry team	4	7	7	14	4	36
EUROCAT newsletter	4	6	9	12	7	38
EUROCAT scientific journal publications	1	2	2	20	20	45
EUROCAT website including prevalence tables	2	2	2	9	29	44
Your local EUROCAT Registry	1	1	5	11	19	37

Q8. Can you provide an example of how the information or services provided by EUROCAT has helped support or inform your work?

58 respondents provided an answer

Text Response
Proportion of prenatally diagnosed fetal limb abnormalities that chose termination of pregnancy. (non RL)
Citing birth prevalence & mortality in GBD (non RL)
prevalence tables on website, risk factors collected, but some rough classification on specific anomalies, e.g. anorectal malformation (non RL)
Comparing Down syndrome prevalence between different countries and our country (non RL)
The availability of figures for the number of NTD pregnancies across Europe has useful to demonstrate that the prevalence has not fallen
To research and compare prenatal screening strategies To Know and compare prevalence of CA between registries and countries (non RL)
I used CA prevalence data to compare indicators between countries. (non RL)
To comparative analyses on different frequencies (non RL)
Prevalence information. Cluster information. Different Studies and Publications. Meetings of register Leaders. Classification and coding. EDMP. Data quality indicators. Excellent help from the Central Registry Team whenever needed.
Our organisation was looking at prevalence of CA and the support and intervention that might be needed in the future. (non RL)
Information on Neural Tube Defects - differences in incidence rates in Europe and in specific countries has been used to inform national policy on folic acid fortification.
Examining relation between use of folic acid and neural tube defects; promotion of use before pregnancy. (non RL)
As a major source of data for CA, we use EUROCAT on almost every research, report and paper. (non RL)
Work in EURO Peristat (non RL)
1. EUROCAT data were very helpful in informing policy decisions regarding changes in prenatal screening of Down syndrome in France by the High Authority of Health in 2009. 2. The recently created perinatal unit at the French health surveillance body (InVS) has used EUROCAT methods and data extensively and is in the process of obtaining EUROCAT validated data on French registries who are members of EUROCAT. These data will in turn be used to provide epidemiological data on congenital anomalies in France (website of InVS) and is also intended to foster collaborative projects between registries in France.
Used when considering upper age limit for inclusion in registers. BDRA 2010;88:251-255 (non RL)

To compare data from other regional registries allow us to verify our data (quality, prevalence...) EUROCAT standards made easier the beginning of our registry

It has helped highlight the increased prevalence and hence challenge posed by livebirths with certain congenital anomalies in countries with no or limited access to termination of pregnancy.

The information help to enlarge or specify some questions of " Announcement of CA" in the National Center of Health Information (non RL)

In 2010, our organisation and Bayer HealthCare Pharmaceuticals published a joint report on the prevention of NTDs, "Act against Europe's most common birth defects: The right advice at the right time can reduce Neural Tube Defects now". Followed in 2011 by "Act against Europe's most common birth defects: one year on - Defining Neural Tube Defect prevention strategies in Europe". Both publications were made possible thanks to the contribution of EUROCAT, which provided most of the data used in the reports. http://www.ifglobal.org/en/what-we-do/if-projects/europe?layout=edit&id=224 (non RL)

We have used information about the prevalence, prenatal diagnosis and outcome of pregnancies for different anomalies in genetic counselling practice and for publishing papers (non RL)

We could see our data in comparison with the other European data and build scientific evidence from it. Especially on efficiency and effects of Prenatal Diagnosis.

I find the website informative, clear, transparent. I find the EUROCAT Registry personnel to be dedicated, helpful, technically competent and very supportive in making links/networking particularly with persons in developing countries. (non RL)

- it gave me actual prevalence data for writing scientific articles - I used it for showing the position of the Netherlands with respect to prenatal screening

EUROCAT is the reference point for the prevalence of congenital anomalies in Europe and is used in all comparisons made by the IMER Registry. Particularly useful the Primary Prevention Recommendations recently published

In my speciality: neonatal mortality and morbidity consequence of CA, nosological structure of neonatal morbidity (non RL)

We are updating the National Strategy of Rare Diseases in Spain and the document on recommendations on policies to be considered for the primary prevention of congenital anomalies in National Plans and Strategies on rare diseases was very useful for the updating. (non RL)

Comparing our data with the pooled and individual registries data. Data are available for presentation and discussions. (non RL)

improve coding, cleaning data , visibility of "my" registry in scientific publications

I am responsible for data modelling for the Congenital Expert Group of the Global Burden of Disease project. The detailed EUROCAT data available on the web has a central role in this undertaking, explained as follows in the article (in preparation) on the general method used in the underlying Global Database of Constitutional Congenital Disorders.

EUROCAT data is used for three purposes in the Global Database. • For neural tube defects and oro-facial clefts, country-specific data are entered directly. Rates for other populations are based on the literature. • For all other malformation groups, average EUROCAT birth prevalences are

used world-wide as best available baseline rates. • Average EUROCAT rates for pregnancy outcomes (livebirths, stillbirths, terminations of pregnancy) are used as best available estimates for populations with access to services but no readily-available observational data. (non RL)

The EUROCAT data are incredible important to evaluate the risks of CAs related to medicines used in pregnancy. The data can generate signals and also evaluate a signal published in literature. (non RL)

Statistics on prevalences to make comparisons within Europe (non RL)

Published evidence is crucial for practitioners and their teachers. (non RL)

Assistance to coding of congenital anomalies by the EUROCAT Coding guide; Coding workshops; Expert support to coding of difficult cases. EUROCAT provided the software for the local registry database – the EUROCAT Data Management Programme (EDMP). Regular training and support to EDMP is provided at the annual Registry Leaders Meeting. The software is supported and developed centrally with economies thus achieved. It enables standard computer data entry to allow comparisons between areas; ease of data analysis across registries; a data validation programme/ facility; and user friendly data analysis options. Research is enabled in this important area of rare diseases. Congenital anomalies have been identified as one of the major groups of mainly rare diseases. Central Registry collates the pooled database, and carries out surveillance and research using the common database. For rare diseases a large population base is needed to generate the required number of subjects for valid research. The Central EUROCAT Registry enables research into the aetiology of individual congenital anomaly and can identify possibilities for primary prevention. Extracts of the common database can be requested by external researchers. The Registry Leaders are drawn from diverse disciplines public health medicine; paediatrics; medical genetics; paediatric cardiology; statistics; epidemiology etc The resultant pooling of expertise provides a comprehensive review of this area. This expertise is seen e.g. in the Coding Committee which provides advice to the World Health Organisation (WHO) International Classification of Diseases (ICD-10) to improve the coding and classification of congenital anomalies. Central Registry undertakes surveillance activity to report on the epidemiology of Congenital Anomaly in Europe. It provides a benchmark against which to compare local surveillance data. The cluster advisory service and protocol assist with the evaluation of clusters. This is a useful local application. EUROCAT Special Reports can inform national policy on the prevention of congenital anomaly; and on service needs and service evaluation.

I have used the information to be able to compare literature data in order to write an article (non RL)

We did start our registry (non RL)

Data provided for research projects like chromosome abnormalities, rare syndrome prevalence etc. Website data and prevalence figures useful for lectures, comparison with local data etc

Improve knowledge on CA through EUROCAT publications and thus informing patients (non RL)

International cooperation with EUROCAT helps us to show the importance of surveillance of congenital anomalies to the local health authorities (e.g. Ministry of Health). (non RL)

Regular information via website as well as scientific publications (non RL)

Informing patients about prevalence of CAs. (non RL)

I work in academic research so the data provided by EUROCAT has enabled me to investigate

the epidemiology of specific congenital anomalies. I have also been able to offer Masters projects using this data so good for training purposes.
Comparing the prevalence tables Using the information about the teratogenic drugs (non RL)
prevalence of certain congenital defects in my region (examples: gastroschisis; congenital cardiac anomalies; oral clefts): useful to design strategies for medical genetics clinic (non RL)
To compare prevalence of registries of my country to Eurocat prevalence, and prevalence of different registries (i.e. neural tube defects), in order to assess the accuracy of a higher prevalence (non RL)
We are using the EUROCAT Statistical Monitoring Protocol (2010) and the EUROCAT Statistical Monitoring Report (2010) as models to develop surveillance protocols in our registry
As registry leader I have provided standardised information for a national study of prevalence of a congenital anomaly subgroup. The website prevalence data graphs and tables have been useful for presentations to clinicians locally. I have used information from published studies from EUROCAT and from the website in demonstrating the work of EUROCAT at the national EUROPLAN conference. The ability to link and network with other registries and access expertise not readily available locally has been extremely valuable in running the registry.
Commonly used to get prevalence data for clinical advice. Particular interest - tried to use to assess employment related chemical exposure to assess neuroendocrine CA - problems with some data fields (non RL)
Information about the prevalence of birth defects. Information on prenatal diagnosis (non RL)
definition of public health indicators for congenital anomalies in France (new annual report on the state of health in France) (non RL)
Create a Protocol of cluster detections; Useful list of publications,
When we have to do annual report about CA, we use information provided by EUROCAT (non RL)
Analysis of trends and clusters, prevalence rates of CA in my local EUROCAT registry comparing to rates from other registries/countries (non RL)
providing annual reports to local health care authorities; development of preventive initiatives and its evaluation; local CA trends and clusters evaluation comparing to Europe
Our registry is a modified EUROCAT registry. All our registry data are entered according to EUROCAT. we are in process of updating our registry to Guide 1.4 which introduced some new variables and also some new values for existing variables. (non RL)
It made an important contribution to the Euro-Peristat report (non RL)
differences in time of TOP to understand our motinatality rate in France comparing with other countries prevalences of CHD in European countries compared in our EPICARD cohort (non RL)
Comparing prevalence of gastroschisis cases for a study (non RL)

Q9. On a scale of 1-5, to what extent are the following EUROCAT data quality assurance and standardization activities important? (select one option for each statement, 1 = Not important, 5 = Very important)

Question	1	2	3	4	5	Total Responses
A consistent Coding and Classification system is used for CA	0	1	6	22	64	93
Data Quality Indicators are used to highlight conformity	0	2	11	39	41	93
Data is collected in a standardised way using common software	1	1	9	31	51	93
Experts are available to Registries for advice and guidance	2	1	9	34	47	93
The EUROCAT Central Registry team and EUROCAT website providing a central place for information	1	1	6	31	54	93

Excluding respondents who identified themselves as EUROCAT Member Registry Leaders.

Question	1	2	3	4	5	Total Responses
A consistent Coding and Classification system is used for CA	0	1	5	19	46	71
Data Quality Indicators are used to highlight conformity	0	2	10	27	32	71
Data is collected in a standardised way using common software	0	1	8	24	38	71
Experts are available to Registries for advice and guidance	2	1	9	27	32	71
The EUROCAT Central Registry team and EUROCAT website providing a central place for information	1	1	6	26	37	71

Q10. On a scale of 1-5, to what extent are the following attributes of EUROCAT CA data valuable? (select one option for each statement, 1 = Not valuable, 5 = Very valuable)

Question	1	2	3	4	5	Total Responses
Comparable across countries	0	1	10	29	53	93
Europe's largest and longest running database	0	2	9	25	57	93
Pan-European	0	5	6	33	49	93
Designed and interpreted by a multidisciplinary network	0	2	14	29	48	93
Provides a platform for CA research	0	1	12	34	46	93
Provides a research infrastructure for rare diseases	1	5	15	28	44	93
Publically available	0	0	10	34	49	93
Quality Assured	0	2	5	37	49	93
Represents a high proportion of EU births	0	0	11	38	44	93
Represents a high proportion of EU countries	0	0	10	36	47	93
Up-to-date	0	1	4	43	45	93
Uses population-based Registries	0	1	5	26	61	93

Excluding respondents that identified themselves as EUROCAT Member Registry Leaders.

Question	1	2	3	4	5	Total Responses
Comparable across countries	0	1	8	20	42	71
Europe's largest and longest running database	0	2	8	20	41	71
Pan-European	0	5	6	25	35	71
Designed and interpreted by a multidisciplinary network	0	2	14	22	33	71
Provides a platform for CA research	0	1	12	25	33	71
Provides a research infrastructure for rare diseases	1	4	12	23	31	71
Publically available	0	0	10	26	35	71
Quality Assured	0	1	4	30	36	71
Represents a high proportion of EU births	0	0	10	30	31	71
Represents a high proportion of EU countries	0	0	9	29	33	71
Up-to-date	0	1	3	35	32	71
Uses population-based Registries	0	1	4	24	42	71

Q11. Do you believe there is a continuing need for the work that EUROCAT does? (select one option) If No, please explain

Answer		Response	%
Yes		92	99%
No		0	0%
Don't know		1	1%
Total		93	100%

Excluding individuals who identified themselves as EUROCAT Member Registry Leaders

Answer		Response	%
Yes		70	99%
No		0	0%
Don't know		1	1%
Total		71	100%

Q12. Do you have any suggestions for improving the work of EUROCAT?

54 respondents provided an answer

Text Response
to give guidance and recommendations unified for all European countries
Give the opportunity to integrate patient group activities and registries, be open for longitudinal studies (non RL)
public engagement / primary prevention / research are examples of what could be maybe enhanced
to involve more staff and professionals in Europe to increase the budget
EUROCAT would collaborate more tightly within EU activities regarding rare diseases.
The inability to implement secondary prevention measures (abortion) in my country, for the majority of congenital anomalies, except anencephaly allows the birth of numerous children with anomalies, and so have a larger data set. The birth rate is still high in some regions of my country and the level of access to this population is very difficult. Their database is based on immigrant populations? For the largest number of birth comes from these groups, or not? Know that the education level is lower and whose religious beliefs often prevents the interruption of pregnancy. Suggest that data on these populations to be more transparent and responsive to their realities. It would be an honest and ethical manner to prevent the birth of babies with congenital anomalies untreatable. The best way, and cheaper, are still educational programs for young people early in their reproductive life.
Communicate more to the public in particular patient organisations. EUROCAT is very academic and more patients need to understand the valuable work done by EUROCAT
Change the central registry database to a new platform ie relational tables. (non RL)
Keep up the good work
Continuity in the excellent work of the EUROCAT Central Registry and the EUROCAT network as a whole. (non RL)
I see that many of the registries facing financial problems. This has to be solved if we intend to continue. This needs a full discussion in one of annual meetings. I will have some suggestions on this matter.

There needs to be a stable funding at the European level for the essential tasks of data collection, validation, and analyses for surveillance (detection of clusters and trends) that are done by the current structure of the Eurocat Central Registry in a very competent, cost- and time-efficient manner. I think with a funding of 300,000 € per year the substantial added-value of the EUROCAT Central Registry can be preserved. In the absence of such funding mechanism, there is a real danger of losing the years of efforts that have gone into building, maintaining and continued development of the infrastructure that allows such efficient and high-quality work in the Central Registry. Even in the best of circumstances and with far more funding, a new team would need a long time before achieving what has already been accomplished by the Central Registry. (non RL)
Seek to ensure that all contributing registers operate to the same high standard and hence are very similar
To improve the data reports (graphics and interactive) (non RL)
More public awareness / publicity. Making information more accessible to the lay person (non RL)
Increase the number of registries to increase the areas of coverage (for instance, the Netherlands only provides data from the 3 northern provinces; not registering important data from highly populated urban areas; only 3% of Germany is covered; 15,5% of France; 13% of Italy).
networking with other networks, biobanks and databases
The work is already great and would just need permanent financial support by the EU. To involve more international participants and wider distribution within countries.
Extend it to include Northern Ireland data
The website and database is extremely useful. I think one the most important aspects of Eurocat is the uniformity of data collection. So in the future, please keep choosing for quality of data and not quantity (non RL)
To stimulate Registries to use quality indicators and registry data for public health evaluation and policies. To maintain the quality of the database through high level scientific publications. (non RL)
include /expand cooperation of EUROCAT with other Ukrainian registries
provide special regular courses for people involved in running the registries. To have more world affiliate registries.
Regular linkage of EUROCAT data on affected births with data on survival of the disorders included, and with national cause of death data, could significantly enhance the public health value of EUROCAT data (e.g. for assessing life-time burden, and the effectiveness of therapeutic interventions).
Important to keep well trained and critical people of all different disciplines involved. I work with EUROCAT data since 2006 and I am impressed by the way the EUROCAT-team is working together.
New registers should be added to the data collection, but only if their quality is assured.
More funding is needed for the registries to find the cases and collect the relevant data.

EUROCAT should fund the registries.
The longevity of the network , the low turnover rate of participants, the generous support and input of individual registry leaders, the consistent performance of the database and its high productivity in terms of reports and peer reviewed research in the area of congenital anomaly - are testamont to its usefulness to its members and successful management by Central Registry. (non RL)
I think the work should be extended in some countries
Collect more exposure information on a European basis
Continued funding that is easy to administer leaving time to collect, analyse and report the data and consider further research. (non RL)
involve even more countries make biological material (eg. DNA, blood, tissue) collection. use a common code (Orphacode) communicate more the outcome of registries analysis take part into actions of National Plans for rare diseases
Improving EDMP form. Prepare to connect to environmental data falling in the scope of the INSPIRE directive.
Maintain the quality of website and the annual monitoring report The website could be more attractive, with tables easier to understand (for instance columns' labels)
It would be good to increase the activities in which world affiliates could participate (non RL)
At present the only coding workshop or training is at the RLM, although we can access advice via email etc when we need it. Coding can be difficult and an online coding training module would be very useful, both for new and established registries. Feedback on the quality of coding recently has also been very helpful. (non RL)
Ongoing review of the data fields to ensure valuable epidemiological questions can be considered.
Look at the National Birth Defects Prevention Study implemented in US
improving quality of data : - comparability of data across European countries using same coding process and same definition of population based registry - used of administrative data set for improving completeness in ascertainment of cases
Develop our registry, (non RL)
Continuous support and development to improve data collection from remote EUROCAT members.
It would be better if larger countries had a fuller coverage of registers, but that is not within Eurocat's control
more implications in clinicians areas (congress, journals, associated publications ...)
More dissemination of your activities to the general public so that they understand its value
I think that there is huge and consolidated trend in rare diseases in EU and also in the world and registries of CA are part of the develpments in RD registries , so I think that EUROCAT would be work as part of ORPHANET or in very close coopration with networks of RD.

Q13. Are there any emerging areas that EUROCAT should focus on in the future?

51 respondents provided an answer

Text Response
unification and standardization in data collection and analysis (non RL)
Home Management of congenital malformed children after discharge from hospital i.e. Meningomyelocele, anorectal malformation, colostomy care , tracheoesophageal fistula etc. (non RL)
genomics / GEI / epigenetics research (non RL)
don't think so (non RL)
Monitoring the increasing use of cell free fetal DNA tests
Other CA as Cystic Fibrosis and Neurologic congenital diseases (non RL)
I do not have any specific suggestions. (non RL)
I do not know (non RL)
It would be great if they could look at genetic diseases (non RL)
Preconceptional care.
Possibly link with cancer registries where there is a question of increased congenital anomaly incidence after an environmental incident - frequently the possibility of a rise / cluster is relevant to both.
Suspected environmental pollutants. (Micro) nutrients. Paternal age. (non RL)
We have worked (and published) on an outbreak of some types of birth defects in one of the Iraqi areas. I believe that international organizations (including EUROCAT and ICBDSR) should play a strong role to investigate this problem in the field. As the world leading experts on CA, I think EUROCAT and ICBDSR have a major responsibility on this. I would be happy to provide more information/co-operation if it is principally agreed by EUROCAT. (non RL)
New techniques for prenatal screening and diagnosis - such as those based on fetal DNA analyses
ART and link with CA, as well as ageing maternal population (non RL)
The increasing use of microarray may throw up diagnostic dilemmas regarding what to include as cases (non RL)
The rarer conditions - as much information as possible of these conditions should be made available both to the professional and lay community.
Eurocat data from Eastern Europe would be most welcome. (non RL)
collecting more data on genetic testing, explore more environmental causes of CA and outcome of children with CA (non RL)
EUROCAT has already a lot of focuses. I cannot imagine additional ones. - Given the

available dataset and financial support. (non RL)
I am trying to involve them in supporting work in developing countries where far less is known, and far more drugs/exposures occur without information being obtained. This would complement and increase sample size of information available, and yield data on genetic risk factors. (non RL)
Perhaps stimulating registries collecting DNA data as well. I think epigenetics is becoming more and more important
Maintenance and strengthening of the European network of registers in order to have reliable information in the field of congenital malformations and rare diseases.
Focus on the spontaneous abortions and dead births - better registration of accompanying CA (non RL)
to run a multinational case control studies to assess the role of some of the risk factors for CA (non RL)
Possibility of linkage with other data sources, e.g. cause of death data (as already mentioned) and other registries e.g. congenital hypothyroidism, Down syndrome, phenylketonuria etc. Expansion of global interest, e.g. through a web-based education/training course to support extension of methodology and standards to other parts of the world. (non RL)
pollution and adverse pregnancy outcomes as congenital anomalies (non RL)
Use of medication during pregnancy, environmental risk factors (non RL)
Breastfeeding (non RL)
Primary prevention of congenital anomaly Continue research into the aetiology of congenital anomaly especially following lifestyle / common exposures such as prenatal alcohol intake by pregnant women Continue pharmocovigilance Continue to provide data to inform the assessment of the risk to public health arising from / following exposure during pregnancy to infectious or to non-infectious environmental hazards
All the country (non RL)
Prenatal diagnosis
Making biological sample bank (non RL)
coding and classification of rare diseases new diagnosis on high speed sequencing platforms link with environmental data (non RL)
Bioethics (non RL)
environmental influence, medication in pregnancy (non RL)
I think there may be (first trimester screening, survival) but not at the expense of maintaining high quality data
Study of the genetic causes of congenital anomalies
Birth defects and Reproductive technologies (non RL)

Real etiological studies using high quality information on risk factor exposure (non RL)
environmental health : providing data of prevalence and developing research on environmental risk factors (non RL)
Cluster detection (including spatial and time clusters).
prevention of CA, environmental risk factors, teratogens
anomalies from congenital exposure that manifest later in life i.e early childhood, adolescence (non RL)
Effects of alcohol use in pregnancy (non RL)
preventive strategies
Establishing links with WHO. (non RL)
Prenatal diagnostics (not only CA, but also chromosomal and monogenic diseases) and influence of Prenatal diagnostics in CA statistics. (non RL)
clinical practices and CA outcomes (non RL)
Access to social care for families with disabled children (non RL)
Public patient engagement (non RL)

Q14. Any other comments?

Text Response
Public awareness regarding prevention of congenital malformation. (non RL)
EUROCAT is a wonderful system and initiative and it simply must continue - it makes an outstanding contribution to perinatal and infant health as well as research into the causes of birth defects (non RL)
EUROCAT an example of European excellence (non RL)
you have sent this questionnaire in multiple rules to various e-mail addresses and I did not agreed in the past to participate as you said in the request letter... (non RL)
I would like Eurocat will work further (non RL)
EUROCAT is an excellent and important organization worth while continuing in the future and deserves all support it needs. It produces important and valuable information on congenital anomalies, their surveillance and prevention, for patients and their families, clinicians, authorities at all stages and for media.
As I mentioned in my previous comment, EUROCAT (and ICBDSR) should play a role on the problems occurring in different parts of the world related to birth defects. I do not think it is enough to register/disseminate data on CA only. As far as I have studied, there are some clusters on CA in different parts of the world needing accurate investigation/intervention by experts. This really needs EUROCAT (and ICBDSR). (non RL)
As I have noted below, I am very much concerned about the future of EUROCAT and the real risk of loss of much has been accomplished through the efforts of EUROCAT's project leader Pr. Helen Dolk and her excellent team at the Central Registry. My fear is that with the dismantling of the Join Action the current funding and institutional environment would not be conducible, at least not in the short or middle term to an uninterrupted functioning of the Eurocat activities (please see also my comments in response to a previous question).
Eurocat is a great exemplar of how to manage a network of registers (non RL)
Only compliments for providing such important and valuable information freely. (non RL)
EUROCAT is great, central registry is awesome. (non RL)
EUROCAT is a unique resource for large-scale quality epidemiological data on congenital anomalies. In view of increasing recognition of their global importance, secure long-term funding is more important than ever. (non RL)
EUROCAT is of great importance to evaluate the possible risks of medicines taken by pregnant women. Every day new medicines are coming on the market and we do not have any information related to the safety in pregnancy when a medicine is marketed. This knowledge comes available when a drug is taken by pregnant women. The EUROmediCAT project shows the importance of EUROCAT. (non RL)
Keep up the good work

Thank you
A very valuable resource that has taken years to get to a really useful, stable place. Any major disruption to its funding, or manner of administration, would undo much of this and would be highly regrettable. EUROCAT has the longest experience as an European registry and should share its experience with other registries in the field of rare diseases, in particular, how to motivate clinicians throughout the EU to enter data (non RL)
I appreciate the quality of the monitoring process to analyze clusters, of the guidelines, of the website. Eurocat is also an opportunity for registers to maintain and gain quality. (non RL)
Keep up the good work and expand it! (non RL)
Thanks everybody for a great job! The work done within EUROCAT activities is necessary and has a great local and international influence.
It is very important and funding should continue (non RL)

Other

Tracing Impact

During the time of the EUROCAT Joint Action Project, EUROCAT Central Registry was also responsible for developing an impact case study, entitled "Antiepileptic Drug Safety in Pregnancy - epidemiological surveillance of congenital anomalies (birth defects)", for submission to the Research Excellence Framework (REF) the new system for assessing the quality of research in UK higher education institutions (HEIs) such as the University of Ulster (Co-ordinating Centre of the EUROCAT Joint Action and home of EUROCAT Central Registry).

1. Summary of the impact

(1) Enhancing the awareness of (i) women of childbearing age suffering from epilepsy and prescribed new and/or older generation AEDs, and (ii) their healthcare professionals. Empowering both to make informed decisions through evidence-based practice that will reduce/prevent the risk of harm to unborn children potentially exposed to AEDs in early pregnancy.

(2) A change in the process by which GlaxoSmithKline (GSK) practices post-marketing epidemiological surveillance of the new generation AED 'lamotrigine' in pregnancy.

(3) Benefit to the methodological practice of other researchers in Europe involved with AEDs and epidemiological surveillance

(4) Contribution to building European system for reproductive safety evaluation

2. Underpinning research

The EU-funded European Surveillance of Congenital Anomalies network (EUROCAT), is coordinated and led by *Professor Dolk* since 2000. Surveillance to ensure early detection of new teratogens (i.e. birth defect causing exposures) originated following the thalidomide tragedy when thousands of children were born with limb defects due to a medication used in early pregnancy. EUROCAT population-based registries (42 in 23 European countries covering 1.7M births annually) annually transmit a dataset to a central database (Centre for Maternal, Fetal and Infant Research, Institute of Nursing and Health Research, University of Ulster) where quality validation, and epidemiologic surveillance and research are conducted relating to causes and prevention. This case study concentrates on AED safety in pregnancy. *Maria Loane* leads EUROCAT Central Database Management and Surveillance since 2002. *Professor Lolkje de Jong van den Berg*, led until recently the Medication Safety in Pregnancy Working Group, and collaborates closely on related research.

For newly licensed medications, safety information is limited to pre-marketing animal studies (with limited ability to predict harm in human pregnancy), since pregnant women are excluded from clinical trials. Post-marketing surveillance (pharmacovigilance) is essential for early detection of safety concerns, particularly difficult for birth defects due to the rarity of cases. Very large study populations are needed to provide sufficient statistical power. This research is relevant to regulatory decisions regarding medication safety and product safety information, and to clinical decision making regarding risk and benefit of treatment options.

The research output relates to case-control studies performed 2007-2009 using EUROCAT data to address hypotheses (or evaluate signals) from the literature regarding teratogenicity of AEDs (new -lamotrigine[1] and old - valproic acid[2]/carbamazepine[3])). An AED database was created for this referring to 3.9M births (19 registries, 1995-2005) including 98,075 livebirths, stillbirths or terminations with birth defects.

(i) The lamotrigine study[1] responded to a signal indicating an over 10-fold raised risk of orofacial clefts associated with lamotrigine, from the North American AED cohort. The study did not support the original signal, nor have subsequent updates[4].
(ii) Valproic acid was known to be teratogenic, but which birth defects were specifically associated was unknown as reports include chance associations. 7 of 14 birth defects suggested in the literature were confirmed as significantly associated with valproic acid exposure, with up to 13-fold risk. This is the first study to specifically identify types of birth defect caused, with implications beyond clinical practice to elucidating teratogenic mechanisms of action[2].
(iii) The carbamazepine study proceeded as for valproic acid, and in contrast confirmed only one significantly associated birth defect - spina bifida, with a much less raised risk than for valproic acid[3].

EUROCAT Guide 1.3: Instructions for the Registration of congenital anomalies[5], a methodological guide developed by a process of consultation and consensus for EUROCAT research and surveillance, includes standardised congenital anomaly inclusion/exclusion criteria and classification and coding, used in the AED studies and all other EUROCAT studies, and available to other researchers in this field. This Guide also introduced the International Anatomic Therapeutic Classification coding of medication exposure for EUROCAT data, which, after a period of training and data source validation, has enabled the subsequent AED research.

Details of the Research Team
Professor Dolk (Professor of Epidemiology and Health Services Research) since 2000.
Maria Loane, Public Health Lecturer since 2002.
Visiting Professor of Pharmacoepidemiology, Lolkje de Jong van den Berg since 2010.

3. References to the research

Impact factors (IFs), citation reports, related funding, and google analytics have been included as quality indicators of the underpinning research.

1. Dolk H, Jentink J, Loane M, Morris J, de Jong-van den Berg LTW and on behalf of the EUROCAT AED Working Group (2008), Does lamotrigine use in pregnancy increase orofacial cleft risk relative to other malformations? *Neurology,*71, 714-722

2. Jentink J, Loane M, Dolk H, Barisic I, Garne E, Morris J, de Jong-van den Berg L for the EUROCAT Antepileptic Study Working Group (2010), Valproic Acid Monotherapy in Pregnancy and Major Congenital Malformations, *The New England Journal of Medicine*, 362, 2185-2193 http://www.nejm.org/doi/pdf/10.1056/NEJMoa0907328

3. Jentink J, Dolk H, Loane M, Morris JK, Wellesley D, Garne E, de Jong-van den Berg L for the EUROCAT Antiepileptic Study Working Group (2010), Intrauterine Exposure to Carbamazepine and Specific Congenital Malformations: Systematic Review and Case-Control Study, *British Medical Journal*, 341, c6581
http://www.bmj.com/content/341/bmj.c6581.pdf%2Bhtml

4. Wang H, Garne E, Loane M, Dolk H, Morris JK, de Jong-van den Berg L (2012), Lamotrigine Use in Pregnancy and Risk of Orofacial Cleft, an Update. Poster presentation at The International Society of Pharmacoepidemiology's 28th International Conference on Pharmacoepidemiology and Therapeutic Risk Management, August 2012, Barcelona, Spain URL: http://onlinelibrary.wiley.com/doi/10.1002/pds.3324/pdf abstract 691 (page 321)

5. EUROCAT (2005). EUROCAT Guide 1.3. Instructions for the registration and surveillance of congenital anomalies [Online], available at: http://www.eurocat-network.eu/ABOUTUS/DataCollection/GuidelinesforRegistration/Guide1_3InstructionManual

4. Details of the impact

The WHO recognises "The Importance of Pharmacovigilance – Safety Monitoring of Medicinal Products" for impacting drug regulation, clinical practice and international health (http://apps.who.int/medicinedocs/en/d/Js4893e/). Pharmacovigilance imparts an impact by empowerment and reassurance through knowledge. Negative results (results not supporting a signal regarding medication risk) are as important as positive results, but the impacts are less demonstrable, and do not attract media attention. In 2006, the European Medicines Agency adopted a guideline on "The exposure to medicinal products during pregnancy: Need for post-authorisation data". Within, EUROCAT is listed as a source of information for human pregnancy data collected post-authorisation (pg 9) (http://www.ema.europa.eu/docs/en_GB/document_library/Regulatory_and_procedural_guideline/2009/11/WC500011303.pdf). Our research does not and should not stand alone. It is a component of the wider global pharmacovigilance agenda.

Evidence of impact described as:
1. Enhancing awareness of women of childbearing age suffering from epilepsy and prescribed AEDs, and their Healthcare Professionels (HCPs), empowering both to make informed decisions through evidence-based practice that will reduce/prevent the risk of teratogenic harm to unborn children potentially exposed to AEDs.

The valproic acid study[2] based on birth defect information of nearly 4M births had visibility in a high impact medical journal (NEJM) and in media. The reemphasis and further clarification and quantification of the known teratogenicity, was an important part of changing awareness and practice. The lamotrigine study[1] which was largely negative was also important in helping women/HCPs make optimal medication choices based on updated patient information (i.e. the pregnancy and lactation section of the Global Data Sheet).

Our research has been included in systematic reviews which inform evidence-based practice for women with epilepsy, but also with bipolar disorder, now a more common indication for use of some AEDs.

Contribution to Medscape, Motherisk and Patient.co.uk – authoritative, trusted, accessible online information for pregnant women/HCPs regarding the safety/risk to the developing foetus associated with maternal exposure to drugs. Rigorous literature reviewing allows rapid integration of new practice-changing evidence, such as our research on carbamazepine and lamotrigine.

2. A change in the process by which GSK practices post-marketing pharmacovigilance in relation to lamotrigine

In 2006, based on a signal from emerging data of the North American AED Pregnancy Registry, GSK alerted HCPs to a possible association of lamotrigine exposure with orofacial clefts. A US Federal Drugs Agency (FDA) warning followed (http://www.fda.gov/Drugs/DrugSafety/PostmarketDrugSafetyInformationforPatientsandProviders/ucm126225.htm). GSK, FDA, UK Medicines and Health Regulatory Agency (MHRA) and European regulators (EMA) revised patient information, and agreed new research was needed to independently confirm this finding. GSK approached Prof. Dolk (2006) to establish the possibility of funding feasibility research by EUROCAT. EUROCAT offered more appropriate and powerful observational research methods, than heretofore possible using the GSK run International Lamotrigine Pregnancy Registry (1992-2010). The results of the EUROCAT research that ensued were disseminated by confidential report to GSK and scientific paper[1]. GSK shared the results with regulators who endorsed a revision to the core safety information provided in the Pregnancy and Lactation section of GSKs Global Data Sheet for lamotrigine, by insertion of - "*A case control study did not demonstrate an increased risk of oral clefts compared to other defects following exposure to lamotrigine*". Based on our data, the regulators expressed

an interest in monitoring a potential signal for club foot and lamotrigine, a study now underway (http://www.eurocat-network.eu/content/Poster-Lamotrigine-Mejnartowicz.pdf) within the GSK-funded research. Following closure of GSK's Registry, Prof. Harden, Director of Comprehensive Epilepsy Care Center, published a commentary explaining its replacement by further use of EUROCAT data.

3. Benefit to the practice of other researchers in Europe.

EUROCAT guidelines[5] are utilised as the gold standard methodology by others when conducting research into birth defects and AEDs and are helping overcome non-comparability between studies. Another important impact is strengthening the system of signal generation and signal evaluation in AED pharmacovigilance among research groups worldwide, such that signals generated by one research study are evaluated by one or more others, as our methodology clearly adheres to and has promoted this approach. An indirect impact of this research is that, in order to ensure scientific independence and transparency in industry-sponsored research, Prof Dolk chaired, for four years to 2012, the European Medicines Agency Working Group which developed a Code of Conduct for Scientific Independence and Transparency (http://www.encepp.eu/documents/encepp_studies/ENCePP%20Code%20of%20Conduct_20100507.pdf).

4. Sustainable Impact

These studies were the first to use EUROCAT data for investigation of specific drugs. Since then we have had many queries from pharmaceutical companies/researchers requesting data/information on other medications. A PhD student at the University has analysed the EUROCAT data in relation to antidepressant safety; an ongoing GSK funded study continues to study lamotrigine [4]. EUROmediCAT is looking further at newer generation AEDs, insulin analogs, antidepressants and antiasthmatic drugs, is testing new methodologies and has a wider aim of building a European system for reproductive safety evaluation.

Annex 1 EUROCAT Evaluation Plan

1. Process Evaluation

Process evaluation relates to planning, organisation and assuring quality of implementation of project activities, identifying and overcoming obstacles and verifying that the stated objectives have been met. This will included determining that the process/output indicators, the milestones and the deliverables have been met.

The process indicators measure the progress of activities in the EUROCAT Joint Action and the way these are carried out (e.g. annual data transmission by EUROCAT registries, by how many?, did they meet the deadline?).

Output indicators measure the quantity, quality and timeliness of the products of the EUROCAT Joint Action activity (e.g. update of website prevalence tables for all congenital anomaly subgroups, how many EUROCAT registries provided data to enable this?, was this achieved annually as planned?).

Process evaluation will be internally conducted by the Project Management Committee and the Steering Committee.

2. Effect Evaluation

Effect evaluation relates to evaluation of the outcome and impact during the period of the EUROCAT Joint Action and for a five year period predating commencement of the EUROCAT Joint Action.

Outcome indicators measure the intermediate results generated by the EUROCAT Joint Action outputs (e.g. improvement in Data Quality Indicators in EUROCAT registries).

Impact indicators measure the quality and quantity of long-term results generated by the EUROCAT Joint Action output (e.g. citations of EUROCAT prevalence data/published papers).

Methods of effect evaluation:

(a) **Citation Tracking**

Citation tracking (of peer-reviewed journal publications) will be performed in preparation of the final report, using Scopus Citation Tracker (SciVerse). Publications to be tracked include:

1. A selection of collaborative key peer-reviewed journal publications published as a result of activity undertaken by the EUROCAT Network during the last funding contract (2007-2010)

- Boyd P, Haeusler M and Barisic I (2011). EUROCAT Report 9: Surveillance of Congenital Anomalies in Europe 1980-2008. Birth Defects Research (Part A). 91: S1
- Boyd P, Barisic I, Haeusler M, Loane M, Garne E and Dolk H (2011). Paper 1: The EUROCAT network: organization and processes. Birth Defects Research (Part A). 91: 2-15.
- Khoshnood B, Greenlees R, Loane M, Dolk H, EUROCAT Project Management Committee and EUROCAT Working Group (2011). Paper 2: EUROCAT public health indicators for congenital anomalies in Europe. Birth Defects Research (Part A). 91: S16-S22.
- Loane M, Dolk H, Garne E, Greenlees R and EUROCAT Working Group (2011). Paper 3: EUROCAT Data Quality Indicators for population-based registries of congenital anomalies. Birth Defects Research (Part A). 91: S23-S30.

- Loane M, Dolk H, Kelly A, Teljeur C, Greenlees R, Densem J and EUROCAT Working Group (2011). Paper 4: EUROCAT Statistical Monitoring: Identification of ten year trends of congenital anomalies in Europe. Birth Defects Research (Part A). 91: S31-S43.
- Garne E, Dolk H, Loane M, Wellesley D, Barisic I, Calzolari E and Densem J (2011). Paper 5: Surveillance of multiple congenital anomalies: implementation of a computer algorithm in European registers for classification of cases. Birth Defects Research (Part A). 91: S44-S50.
- Greenlees R, Neville A, Addor M-C, Amar E, Arriola L, Bakker M, Boyd P, Calzolari E, Doray B, Draper E, Vollset S E, Garne E, Gatt M, Haeusler M, Kallen K, Khoshnood B, Latos- Bielenska A, Martinez-Frias M-L, Materna-Kiryluk A, Dias C M, McDonnell R, Mullaney C, Nelen V, O'Mahony M, Pierini A, Queisser-Luft A, Randrianaivo-Ranjatoelina H, Rankin J, Rissmann A, Ritvanen A, Salvador J, Sipek A, Tucker D, Verellen-Dumoulin C, Wellesley D and Wertelecki W (2011). Paper 6: EUROCAT member registries: organization and activities. Birth Defects Research (Part A). 91: S51-S100.
- Jentink J, Loane M, Dolk H, Barisic I, Garne E, Morris J, de Jong-van den Berg L and EUROCAT Antiepileptic Study Working Group (2010). Valproic acid monotherapy in pregnancy and major congenital malformations. The New England Journal of Medicine. 362: (23). 2185-2193.
- Jentink J, Dolk H, Loane M, Morris J, Wellesley D, Garne E, de Jong-van den Berg L and EUROCAT Antiepileptic Study Working Group (2010). Intrauterine exposure to carbamazepine and specific congenital malformations: systematic review and case-control study. British Medical Journal. 341: C6581
- Dolk H, Jentink J, Loane M, Morris J, de Jong-van den Berg L and EUROCAT Antiepileptic Drug Working Group (2008). Does Lamotrigine use in pregnancy increase orofacial cleft risk relative to other malformations. Neurology. 71: 714-722.
- Dolk H, Loane M, Garne E and EUROCAT Working Group (2011). Congenital heart defects in Europe: Prevalence and perinatal mortality, 2000 to 2005. Circulation. 123: 841-849.
- Best KE, Tennant P, Addor M-C, Bianchi F, Boyd P, Calzolari E, Dias C M, Doray B, Draper E, Garne E, Gatt M, Greenlees R, Haeusler M, Khoshnood B, McDonnell R, Mullaney C, Nelen V, Randrianaivo-Ranjatoelina H, Rissmann A, Salvador J, Tucker D, Wellesley D and Rankin J (2012). Epidemiology of small intestinal atresia in Europe: a register-based study. Archives of Disease in Childhood - Fetal and Neonatal Edition. 97: F353-F358.
- Garne E, Loane M, Dolk H, Barisic I, Addor M-C, Arriola L, Bakker M, Calzolari E, Dias C M, Doray B, Gatt M, Klungsoyr Melve K, Nelen V, O'Mahony M, Pierini A, Randrianaivo-Ranjatoelina H, Rankin J, Rissmann A, Tucker D, Verellen-Dumoulin C and Wiesel A (2012). Spectrum of congenital anomalies in pregnancies with pregestational diabetes. Birth Defects Research (Part A). 94: 134-140.
- Khoshnood B, Loane M, Garne E, Addor M-C, Arriola L, Bakker M, Barisic I, Bianca S, Boyd P, Calzolari E, Doray B, Draper E, Gatt M, Haeusler M, Klungsoyr Melve K, Latos- Bielenska A, McDonnell R, Mullaney C, Nelen V, O'Mahony M, Pierini A, Queisser-Luft A, Randrianaivo-Ranjatoelina H, Rankin J, Rissmann A, Salvador J, Tucker D, Verellen-Dumoulin C, Wellesley D, Zymak-Zakutnya, N and Dolk H (2012). Recent decrease in the prevalence of congenital heart defects in Europe. Journal of Pediatrics. 162: (1). 108-113.
- Pedersen R, Calzolari E, Husby S, Garne E and EUROCAT Working Group (2012). Oesophageal atresia: prevalence, prenatal diagnosis and associated anomalies in 23 European regions. Archives of Disease in Childhood. 97: 227-232.

- Wellesley D, Dolk H, Boyd P, Greenlees R, Haeusler M, Nelen V, Garne E, Khoshnood B, Doray B, Rissmann A, Mullaney C, Calzolari E, Bakker M, Salvador J, Addor M-C, Draper E, Rankin J and Tucker D (2012). Rare chromosome abnormalities, prevalence and prenatal diagnosis rates from population based congenital anomaly registers in Europe. European Journal of Human Genetics. 20: (5). 521-526.
- Boyd P, Loane M, Garne E, Khoshnood B, Dolk H and EUROCAT Working Group (2011). Sex chromosome trisomies in Europe: prevalence, prenatal detection and outcome of pregnancy. European Journal of Human Genetics. 19: 231-234.
- Loane M, Dolk H, Morris J and EUROCAT Working Group (2009). Maternal age-specific risk of non-chromosomal anomalies. British Journal of Gynaecology. 116: 1111-1119.

2. All peer-reviewed journal publications that have arisen as a result of activity undertaken by the EUROCAT Network during the Joint Action (2011-2013)

3. EUROCAT Guide 1.3 is extensively cited in peer-reviewed publications. As this document is made available on the EUROCAT website, there are no formal mechanisms to trace citation of this document. We will conduct web-based searches to determine the breadth and reach of the citation profile of EUROCAT Guide 1.3. We will also rely on members of the EUROCAT Network to inform us of citations of EUROCAT Guide 1.3.

(b) **Google Analytics**

Google analytics will be employed to determine the visitor profile to the EUROCAT website between 2010 and 2013 (e.g. number of returning and new visitors).

(c) **Website Tables Registration**

EUROCAT Central Registry traces and profiles use of the website tables (since registration began in March 2012) by tracking the number of registrations by country and by type and the number of website table reports generated. An overview of website table registration will be provided in the final evaluation report.

(d) **External Enquiries/Feedback**

EUROCAT Central Registry logs external enquiries or feedback regarding EUROCAT data and/or activity - An overview of those received during the time period of the EUROCAT Joint Action will be provided in the final evaluation report.

(e) **Media Interest**

EUROCAT Central Registry makes and effort to trace reference to EUROCAT data/activity in the media – An overview of which will be provided in the final evaluation report.

(f) **Web-based Evaluation Survey**

EUROCAT is obtaining constructive feedback (January 2013 through December 2013) about its website and other outputs via a web-based evaluation survey tool (of 10 short questions taking no longer than 10 minutes to complete) in order to determine if EUROCAT is reaching its intended user and if EUROCAT's output is appropriate for that user.

In addition to a pop-up invitation to take the survey upon visiting the EUROCAT website, a link to the survey has been disseminated via the EUROCAT Central Registry mailing list (that continues to be updated throughout the timeframe of the EUROCAT Joint Action) and the EUROCAT newsletter. EUROCAT Central Registry will encourage further dissemination of the survey link by the broader EUROCAT membership via reminders in EUROCAT communication emails and via an evaluation strategy presentation at the final EUROCAT Registry Leader's Meeting in June 2013.

Results from the web-based evaluation survey will be included as part of the final evaluation report.

(g) **Evaluation of EUROCAT Symposiums**

The Antwerp based symposium (in June 2011) was evaluated by E-mail questionnaire. The Zagreb based symposium (in June 2013) will also be evaluated by E-mail questionnaire. Summaries of both evaluations will be included in the final evaluation report.

(h) **Independent Evaluation**

A questionnaire (created by an external subcontractor and accessed via an online survey tool) will be employed to focus on perception of "value" of the EUROCAT outputs/outcomes by targeted end-users (split across 3 evaluation panels).

Members of all three evaluation panels will have to provide their email addresses and agree to have their names published in the evaluation report, but all responses will remain anonymised.

Evaluation Panel 1 - Each Registry Leader will respond to the questionnaire and will be asked to identify 4 additional respondents (within their country/region) and confirm their intent to participate and provide the following information to the independent evaluator via Central Registry. Those registries/countries that have established National Committees will use them where possible.

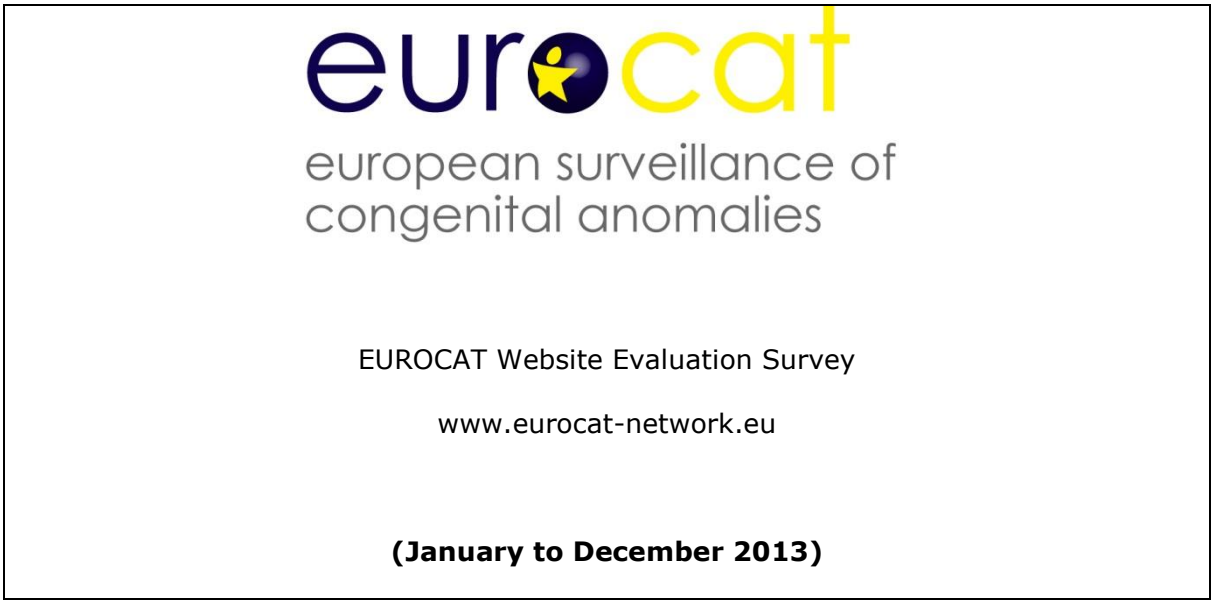
Registry:				
Respondent Group	**Name**	**Role**	**Affiliation**	**Email**
Registry Leader				
Public Health Authority				
Clinician				
Patient/Patient Organisation				
Department of Health – Rare Diseases (e.g. EUCERD rep)				

Evaluation Panel 2 – will consist of representatives from WHO, Europe (DG Sanco/EAHC), EMA, EUROPERISTAT, EURORDIS, Orphanet, and Professional Societies.

Evaluation Panel 3 – will consist of proactive volunteers identified by putting a request out via the EUROCAT Central Registry emailing database.

Annex 2

EUROCAT Website Evaluation Survey



INTRODUCTION

EUROCAT will finish its current Joint Action contract with the EU Public Health Programme (2008-2013) in December 2013.

The purpose of EUROCAT's dissemination activity is to raise awareness of the importance of congenital anomaly registries and databases coordinated at European level, on the possibilities that they offer in terms of collecting data, coding and classification of rare disorders, public health planning, primary prevention, and research in the field of congenital anomalies. The purpose of the dissemination activity is also to raise the profile of the EUROCAT network, and to gain wider support for the setting up congenital anomaly registries across Europe. The results of the EUROCAT Joint action will serve to inform and educate the wider community on the importance of prevention strategies for congenital anomalies. Through dissemination of the EUROCAT Joint Action results we would like to actively engage the community in relation to improving the health status of women of childbearing age.

EUROCAT wishes to obtain constructive feedback about its website and other outputs in order to determine if we are reaching our intended user and if our output is appropriate for that user. You are likely to be in receipt of this survey because you accessed the EUROCAT website, or you have been identified by EUROCAT or an associate of EUROCAT as a target user. We welcome the opportunity to gain a better understanding of your expectations of and opinions on EUROCAT. This questionnaire (of 10 short questions) should take no longer than 10 minutes to complete.

The results of the website evaluation survey will be made publically available on the EUROCAT website by February 2014.

WE THANK YOU IN ADVANCE OF YOUR PARTICIPATION.

1. Which professional or user perspective represents your interest in EUROCAT?

- ☐ Affected person or parent of affected person
- ☐ National government official with responsibility for public health or health services
- ☐ National government official with responsibility for rare diseases
- ☐ National government official with responsibility for environment
- ☐ Regional or municipal government or health authority official
- ☐ European Commission, WHO or other international organisation official
- ☐ Hospital or health service manager
- ☐ Paediatrician
- ☐ Obstetrician
- ☐ Medical geneticist
- ☐ Nurse
- ☐ Midwife
- ☐ Public health or epidemiology academic
- ☐ Other academic
- ☐ Pharmacovigilance officer within industry
- ☐ Student
- ☐ Other (please specify)

2. Which country do you live in?

3. Which one or more of the following topics is relevant to you?

- ☐ Prevalence of congenital anomalies
- ☐ Causes and primary prevention of congenital anomalies
- ☐ Prenatal screening for congenital anomalies
- ☐ Other (please specify)

4. Which one or more types of congenital anomalies are of particular interest to you in terms of your need for information from EUROCAT?

- ☐ Congenital heart defects
- ☐ Neural tube defects
- ☐ Orofacial clefts
- ☐ Chromosomal including Down syndrome
- ☐ Genetic syndromes
- ☐ Abdominal wall defects (e.g. gastroschisis)
- ☐ Limb defects
- ☐ Other (please specify)

5. Which one or more of the following risk factors for congenital anomaly are of particular interest to you in terms of your need for information from EUROCAT?

- ☐ Folic acid
- ☐ Medicinal drug exposures during pregnancy
- ☐ Environmental pollutants
- ☐ Genetic factors
- ☐ None of the above
- ☐ Other (please specify)

6. Please rate how useful you have found the following outputs from EUROCAT in the period 2011-2013:

	very useful	useful	not useful	did not know about it	not relevant to me
Interactive Website Prevalence Tables	○	○	○	○	○
Website Prenatal Diagnosis Tables	○	○	○	○	○
Website Perinatal Mortality Tables/Key Public Health Indicators	○	○	○	○	○
Annual Statistical Monitoring Reports concerning time trends and clusters	○	○	○	○	○
Guide 1.3/1.4: Instruction for the registration of congenital anomalies	○	○	○	○	○
EUROCAT Special Report (2012): Congenital Anomalies are a Major Group of Mainly Rare Diseases	○	○	○	○	○
EUROCAT Publication List	○	○	○	○	
EUROCAT Newsletter (if you would like to receive the Newsletter please include your email address below)	○	○	○	○	○

7. Have you received information about one or more EUROCAT member registries in your country?

○ Yes

○ No

○ Not applicable

8. Do you know about the European Scientific Symposium on the Prevention of Congenital Anomalies which EUROCAT organises every two years?

Yes, I participated in Antwerp, Belgium (2011) or Zagreb, Croatia (2013)

No, but I would like to find out more and perhaps participate in future (if so please provide your email address below)

I would not be interested or able to participate

9. Do you find the EUROCAT Website easy to use?

Yes

No (if no please specify why)

10. Please give any further comments you may have about EUROCAT. In particular what information you would like to see on the EUROCAT website.

Annex 3

EUROCAT: Independent Evaluation Survey

Q1 In which country do you reside?

Q2 Which of the following best describes who you represent? (select one option)

- ❍ EUROCAT Registry Leader
- ❍ Public Health Authority
- ❍ Paediatrician or Paediatric Surgeon
- ❍ Obstetrician
- ❍ Nurse or Midwife
- ❍ Medical Geneticist
- ❍ Other Healthcare Professional (Please Specify) _____
- ❍ Patient or Patient's Family
- ❍ Patient Organisation
- ❍ Academic or Researcher
- ❍ Regional/National Department of Health
- ❍ European Commission
- ❍ WHO or Other International Organisation
- ❍ Other (Please Specify) _____

Q3 Based on what you currently know about EUROCAT.....Would you recommend all new Registries (registering cases of CA) become a member of the EUROCAT network?

- ❍ Yes
- ❍ No
- ❍ Don't know

Q4 On a scale of 1-5, to what extent is EUROCAT's ongoing CA surveillance important in providing information on the following? (select one option for each statement, 1 = Not useful, 5 = Very useful)

	1	2	3	4	5
Detection and investigation CA time clusters	❍	❍	❍	❍	❍
Early warning of increases in prevalence of CA	❍	❍	❍	❍	❍
Environmental risks	❍	❍	❍	❍	❍
Epidemiological data about rare diseases (rare CA)	❍	❍	❍	❍	❍
Impact of prevention measures in decreasing CA prevalence	❍	❍	❍	❍	❍
Mortality related to CA	❍	❍	❍	❍	❍
Prenatal screening	❍	❍	❍	❍	❍
Prevalence of CA	❍	❍	❍	❍	❍
Safety of medication use during pregnancy	❍	❍	❍	❍	❍
Verifying the absence of CA time clusters	❍	❍	❍	❍	❍

Q5 Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local EUROCAT registry)? (select one option)

❍ Yes
❍ No

If Yes Is Selected, Then Skip To For what purpose did you use this inf...If No Is Selected, Then Skip To On a scale of 1-5, to what extent are...

Answer If Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local EUROCAT registry)? (select one option) Yes Is Selected

Q5a For what purpose did you use this information or data? (tick as many options as apply)

- ❑ Comparing between countries
- ❑ Comparing between Registries in the same country
- ❑ Improving coding and classification methodology
- ❑ Informing clinical practice
- ❑ Informing national policy
- ❑ Informing regional or local policy
- ❑ Research or publications (please provide examples) _____
- ❑ Understanding CA at a European level
- ❑ Other (please specify) _____

Answer If Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local EUROCAT registry)? (select one option) Yes Is Selected

Q5b On a scale of 1-5, how useful were the following for assessing EUROCAT information? (select one option for each you have used, 1 = Not useful, 5 = Very useful, Blank = Not used)

	1	2	3	4	5
Annual Statistical Monitoring Reports	❍	❍	❍	❍	❍
EUROCAT Central Registry team	❍	❍	❍	❍	❍
EUROCAT newsletter	❍	❍	❍	❍	❍
EUROCAT scientific journal publications	❍	❍	❍	❍	❍
EUROCAT website including prevalence tables	❍	❍	❍	❍	❍
Your local EUROCAT Registry	❍	❍	❍	❍	❍

Q9 Can you provide an example of how the information or services provided by EUROCAT has helped support or inform your work?

Q6 On a scale of 1-5, to what extent are the following EUROCAT data quality assurance and standardisation activities important? (select one option for each statement, 1 = Not important, 5 = Very important)

	1	2	3	4	5
A consistent Coding and Classification system is used for CA	❍	❍	❍	❍	❍
Data Quality Indicators are used to highlight conformity	❍	❍	❍	❍	❍
Data is collected in a standardised way using common software	❍	❍	❍	❍	❍
Experts are available to Registries for advice and guidance	❍	❍	❍	❍	❍
The EUROCAT Central Registry team and EUROCAT website providing a central place for information	❍	❍	❍	❍	❍

Q7 On a scale of 1-5, to what extent are the following attributes of EUROCAT CA data valuable? (select one option for each statement, 1 = Not valuable, 5 = Very valuable)

	1	2	3	4	5
Comparable across countries	❍	❍	❍	❍	❍
Europe's largest and longest running database	❍	❍	❍	❍	❍
Pan-European	❍	❍	❍	❍	❍
Designed and interpreted by a multidisciplinary network	❍	❍	❍	❍	❍
Provides a platform for CA research	❍	❍	❍	❍	❍
Provides a research infrastructure for rare diseases	❍	❍	❍	❍	❍
Publically available	❍	❍	❍	❍	❍
Quality Assured	❍	❍	❍	❍	❍
Represents a high proportion of EU births	❍	❍	❍	❍	❍
Represents a high proportion of EU countries	❍	❍	❍	❍	❍
Up-to-date	❍	❍	❍	❍	❍
Uses population-based Registries	❍	❍	❍	❍	❍

Q8 Do you believe there is a continuing need for the work that EUROCAT does? (select one option) If No, please explain your answer

- ❍ Yes
- ❍ No _____
- ❍ Don't know

Q10 Do you have any suggestions for improving the work of EUROCAT?

Q11 Are there any emerging areas that EUROCAT should focus on in the future?

Q12 Any other comments?

Annex 4

Fistral
Training and Consultancy Ltd

EUROCAT Questionnaire

A consultancy engagement for the EUROCAT network

Version: 3.0 Final
12th December 2013

+44 (0)141 636 0290
info@fistraltraining.com
www.fistraltraining.com

Background & Scope

Fistral Training and Consultancy Ltd have been engaged by EUROCAT to develop a 'customer-service' style questionnaire by 13th December 2013. This will determine the 'value' of the EUROCAT network to 'end-users', and reinforce or identify new areas in which the network can continue to provide value to its' users and the European Rare Diseases community into the future.

This questionnaire will ultimately be disseminated online in English, and based on online experience and best-practice, it should be written in simple language and comprise no more than 20 questions.

Purpose of this document

The final questionnaire and related information is provided in fulfilment of the consultancy work, and has been updated to reflect comments from the Steering Committee given 04/12/13.

In addition to the EUROCAT feedback, Fistral has undertaken usability[1] testing of the questionnaire and comments have been incorporated as appropriate.

Out of Scope

Fistral was not engaged to:

- Upload the questionnaire to the survey tool
- Identify the target audience or engage with them in any way
- Test/pilot the questionnaire with the target audience, test how the questionnaire functions on the survey tool etc.
- Disseminate the questionnaire or chase-up responses
- Analyse or draft a report on the questionnaire responses

[1] *Usability: using a set of individuals to check the 'ease of use' of the questionnaire in terms of design, format,flow, clarity, comprehension, length/timing etc. Not subject-specific testing or functional or usability testing in any other format other than its' delivery format (PDF / Microsoft / paper-based to be agreed).*

Table of Contents

Terminology used regarding this Questionnaire

Customer-service style questionnaire	A questionnaire designed to focus on customer satisfaction to help improve and give feedback on products or services. Presented in a 'friendly' way, using simple language, format and navigation. Underpinned by marketing/commercial questionnaire approaches; not necessarily using an academic research approach.
End-users	Individuals from representative groups who use EUROCAT information or services. Not EUROCAT Registry members; however Registry Leaders may be a sub-group.
EUROCAT Vision Statement[2]	"The values that guide EUROCAT's strategic definition of aim and objectives: • **POPULATION-BASED**: We collect epidemiological data in geographically defined populations to represent the unselected experience of all who live in the population. • **REDUCTION OF INEQUALITIES**: We highlight the preventive and service needs for a group of individually mainly rare conditions which together constitute a significant but neglected public health problem; highlight differences between countries and identify high risk groups; work to improve data on socioeconomic inequalities; expand capacity for registries across EU. • **EARLY WARNING**: We aim to monitor and respond to emerging health threats and exposures in a timely manner, and communicate the results to public health authorities. • **ACCURACY**: We invest considerable effort in assuring high quality data, and transparency regarding data quality deficiencies • **PRIMARY PREVENTION AS THE ULTIMATE GOAL**: We use epidemiologic data to raise awareness of the need and potential to accelerate the very slow progress in recent decades towards reducing the number of affected livebirths, prenatal deaths and terminations of pregnancy. • **TRANSPARENCY**: We make all our information available to health care professionals, researchers, policy makers and the public. • **COLLABORATION AND MUTUAL INTERDEPENDENCE**: We recognise all members of the European network as having a valuable contribution to make irrespective of disciplinary, geographic, institutional or other origin; all contribute to and derive benefit from the collaborative network. • **SUSTAINABILITY AND EFFICIENCY**: We design data systems (software, data documentation) to ensure the efficiency and sustainability of the network."
Fistral	Fistral Training and Consultancy Ltd.
Importance	An individual's perception or measure of value based on its' placement in an overall context. Can be assigned to a person, group, product, service, knowledge or type of information.

[2] Extract taken from RC email to Fistral 26/06/13.

Usability testing	Using a set of individuals to check the 'ease of use' of the questionnaire in terms of design, format, flow, clarity, comprehension, length/timing etc. Not subject-specific testing or functional or usability testing in any other format other than its' agreed delivery format.
Usefulness	An individual's perception or measure of value based on its' practical use. Can be assigned to a person, group, product, service, knowledge, or type of information.
Utility	A personal desire, want or preference for something; a measure of relative satisfaction.
Value	In this questionnaire, value is an assessment of the perceived Importance, Usefulness and/or Worth of a person, group, product, service, knowledge or type of information.
Worth	An individual's perception or measure of value based on their utility. Can be assigned to a person, group, product, service, knowledge or type of information.

Approach

Fistral has been managing projects and undertaking project evaluations and audits in the UK, Europe, Africa and Asia, in both commercial and academic sectors, for over 15 years. The team also has a vast experience of managing European-funded projects comprising a variety of UK and European university partners.

This practical expertise combined with online questionnaire experience, and that of Fistral's Associates working in the commercial Marketing arena for large multi-national corporations, ensures the robustness of research undertaken into current "best practice" and industry standards to identify the best approach for EUROCAT.

Questionnaire and Mapping

Meetings, telephone conversations and focus sessions with key EUROCAT staff and various online and face-to-face discussions were undertaken to understand the EUROCAT network/project aims and objects and activities. Sessions achieved understanding of the key areas in which EUROCAT adds value, and therein the unique value of EUROCAT and sub-topics that questions should address.

These areas were aligned to question approaches and subsequent question responses in terms of assessing respondents' Knowledge, Behaviour, Attributes and Opinions. This led to a comprehensive set of 'draft-draft' questions, which were refined to a 'first draft' and then a 'revised draft' on which comments were received from the EUROCAT Steering Committee.

Subsequently a review and focus on the key areas that would demonstrate the 'value' of EUROCAT, has led to the following questionnaire that is aligned to the four attributes given in the statement box below (in no particular order).

Figure 1

In the European Public Health arena, EUROCAT provides 'value' through the following four attributes...

1. **Surveillance of congenital anomalies**
2. **Assuring data quality and standardisation**
3. **Being a European network**
4. **Providing unique data and services**

'Value' being ascertained in the degree to which respondents place:

- Importance;
- Usefulness; and/or
- Worth

on a person, group, product, service, knowledge or type of information.

In addition to the final questionnaire, a map has also been provided to indicate how each question links back to the statement and concept of 'value' given in Figure 1 above. It also provides a keyword indication of the responses, and suggested alignment to EUROCAT 'Vision Statement'.

Disclaimer

Please note that Fistral Training and Consultancy Ltd makes no warranties, express or implied, in this document or subsequent work. In no event shall Fistral Training and Consultancy Ltd be liable for damages of any kind arising out of the work undertaken, changes made post-delivery, deliverables, reports, recommendations or any information; or in the use of and any actions arising (current or future) linked to this work.

Target Audience

The following list indicates the representative categories of audience for the questionnaire.

1. EUROCAT Registry Leader
2. Public Health Authority
3. Clinician
4. Paediatrician or Paediatric Surgeon
5. Obstetrician
6. Nurse or Midwife
7. Medical Geneticist
8. Other Healthcare Professional
9. Patient or Patient's Family
10. Patient Organisation
11. Academic or Researcher
12. Regional/National Department of Health
13. European Commission
14. WHO or Other International Organisation

Technical Note

- Words in blue are for direction – not inclusion in the survey.
- Pagination and question-type has been suggested throughout.
- The default for all response options when transferred online should be blank.
- Number ratings (1-5) have replaced words to avoid confusion or over-complication.
- For radio buttons and tick boxes, responses are ordered alphabetically to avoid leading respondents.

Email Introduction

DEAR XX,

EUROCAT has engaged an independent organisation - Fistral Training and Consultancy Ltd - to develop a questionnaire exploring the 'value' of the network.

This will allow EUROCAT to:

1. Gain user feedback on how they use the network and what areas they find most valuable, to support these better into the future
2. Allow clearer reporting to funders on current progress, and support new funding applications
3. Look to the future and identify areas in which the network can continue to provide value to its' users and the European Rare Diseases Community

You have been selected by EUROCAT to provide user feedback on behalf of XX.

You have agreed to complete the following questionnaire developed by Fistral Training and Consultancy Ltd. There are XX questions which you should answer as fully and honestly as possible by DEADLINE.

The responses you give will inform the future direction of the network, and will also demonstrate to funders the importance of the EUROCAT network and how it can continue to provide value to European partners and the Rare Diseases Community into the future.

Plus EUROCAT to provide

- CONFIDENTIALITY AND USE STATEMENT/LINKS
- WHO TO CONTACT WITH QUESTIONS
- STATEMENT ABOUT ACCESS TO RESPONSE EVALUATION (IF REQUIRED)

LINK TO FISTRAL WEBSITE www.fistraltraining.com

Thank You

Final Questionnaire

INTRODUCTION

Since 1979, the EUROCAT network has pooled and shared information and expertise from across Europe to provide a network of population-based Registries for epidemiologic surveillance of congenital anomalies ("birth defects"), the majority of which are Rare Diseases, and to provide a joint approach to European public health questions.

EUROCAT's mission is to support the primary prevention of congenital anomalies (CA) and the provision of appropriate services to pregnant women, affected children and their families by the ongoing collection, analysis, interpretation and dissemination of epidemiologic data i.e. by epidemiologic surveillance. Surveillance should inform policies and interventions to reduce the size of, and inequalities in, the public health burden of congenital anomalies.

Currently surveying more than 1.7 million births per year in Europe, EUROCAT offers the longest running and largest database that follows a common protocol with efficient data systems for global evaluation.

Data is collected from members in 37 Registries across 21 countries (almost all of the population-based CA Registries in Europe) using the same standard dataset. This means that data is quality assured and comparisons can be made between countries.

EUROCAT provides access to anonymised up-to-date CA data via customisable prevalence tables on www.eurocat-network.eu, or by formal request to EUROCAT.

You have been asked to provide your thoughts on the work undertaken by EUROCAT. This will input into future EUROCAT planning, and will help with responses to funders and ongoing work in the Rare Diseases community.

Please note that throughout this questionnaire, congenital anomalies will be referred to as "CA".

Thank you for your time and input.

EUROCAT Final Questionnaire and Mapping

PAGE 1

1. **Which country do you represent?** (select one option)

	Select one via dropdown menu
Country list to be provided by EUROCAT	

2. **Which of the following best describes who you represent?** (select one option)

	Select one via dropdown menu
EUROCAT Registry Leader	
Public Health Authority	
Clinician	
Paediatrician or Paediatric Surgeon	
Obstetrician	
Nurse or Midwife	
Medical Geneticist	
Other Healthcare Professional Please specify *(Free text)*	
Patient or Patient's Family	
Patient Organisation	
Academic or Researcher	
Regional/National Department of Health	
European Commission	
WHO or Other International Organisation	
Other Please specify *(Free text)*	

PAGE 2

Based on what you currently know about EUROCAT...

3. **Would you recommend all new Registries become a member of the EUROCAT network?** (select one option)

	Select one via dropdown menu
Yes	
No	
Don't Know	

4. **On a scale of 1-5, to what extent is EUROCAT's ongoing CA surveillance important in providing information on the following?** (select one option for each statement, 1 = Not useful, 5 = Very useful)

Select one for each via radio buttons 1=Not important --- 5=Very important	1	2	3	4	5
Detection and investigation CA time clusters					
Early warning of increases in prevalence of CA					
Environmental risks					
Epidemiological data about rare diseases (rare congenital anomalies)					
Impact of prevention measures in decreasing CA prevalence					
Mortality related to congenital anomalies					
Prenatal screening					
Prevalence of CA					
Safety of medication use during pregnancy					
Verifying the absence of CA time clusters					

PAGE 3

5. **Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local Registry)?** (select one option)

	Select one via radio buttons
Yes	
No	

If No, continue to the NEXT PAGE [create link].

If Yes...

a. **For what purpose did you use this information or data?** (tick as many options as apply)

	Tick many via tickbox matrix

	Tick many via tickbox matrix
Comparing between countries	
Comparing between Registries in same country	
Improving coding and classification methodology	
Informing clinical practice	
Informing national policy	
Informing regional or local policy	
Research or publications Please provide examples *(Free text)*	
Understanding CA at a European level	
Other Please specify *(Free text)*	

b. **On a scale of 1-5, how useful were the following for accessing EUROCAT information?** (select one option for each you have used, 1 = Not useful, 5 = Very useful, Blank = Not accessed)

Select one for each as appropriate via radio buttons Blank=Not accessed 1=Not useful --- 5=Very useful					
	1	2	3	4	5
Annual Statistical Monitoring reports					
EUROCAT central registry team					
EUROCAT newsletter					
EUROCAT scientific journal publications					
EUROCAT website					
Your local Registry					

PAGE 4

6. **On a scale of 1-5, to what extent are the following EUROCAT data quality assurance and standardisation activities important?** (select one option for each statement, 1 = Not important, 5 = Very important)

Select one for each via radio buttons 1=Not important --- 5=Very important					
	1	2	3	4	5
A consistent Coding and Classification system is used for CA					
Data Quality Indicators are used to highlight conformity					
Data is collected in a standardised way using common software					

Select one for each via radio buttons 1=Not important --- 5=Very important	1	2	3	4	5
Experts are available to Registries for advice and guidance					
The EUROCAT Central Registry team and website provide a central place for information					

7. **On a scale of 1-5, to what extent are the following attributes of EUROCAT CA data valuable?** (select one option for each statement, 1 = Not valuable, 5 = Very valuable)

Select one for each via radio buttons 1=Not valuable --- 5=Very valuable	1	2	3	4	5
Comparable across countries					
Europe's largest and longest running database					
Pan-European					
Part of a multidisciplinary network					
Provides a platform for CA research					
Provides a research infrastructure for rare diseases					
Publicly available					
Quality Assured					
Represents a high proportion of EU births					
Represents a high proportion of EU countries					
Up-to-date					
Uses population-based Registries					

PAGE 5

8. **Do you believe there is a continuing need for the work that EUROCAT does?** (select one option)

	Select one via radio buttons
Yes	
No	
Don't Know	

a. **If No, please explain your answer** *(Free text)*

9. **Can you provide an example of how the information or services provided by EUROCAT has helped support or inform your work?** *(Free text)*

10. **Do you have any suggestions for improving the work of EUROCAT?** *(Free text)*

11. **Are there any-emerging areas should EUROCAT focus on in the future?** *(Free text)*

12. **Any other comments?** *(Free text)*

CLOSING PAGE

Thank you for your participation in this questionnaire. All answers will be analysed and will help inform EUROCAT's decision-making and planning.

If you would like to discuss any of the above or learn more about becoming a Registry member, click on the links below or leave your email address/telephone number in the box below, or email XX and EUROCAT will get back to you. (Provide links e.g. website, scientific journal publications, newsletter, Annual Statistical Monitoring Reports plus Email and Telephone fields)

For more information visit the EUROCAT website: www.eurocat-network.eu

Sign-up to the EUROCAT mailing list to receive a copy of the annual newsletter and special reports, and to find out more about the annual EUROCAT Symposium: (Add email field)

For more information on the questionnaire or any of the work that EUROCAT performs, contact:

EUROCAT Central Registry

Room 12L09, University of Ulster
Newtownabbey, Co Antrim

Tel: +44 (0)28 90366639

Fax: +44 (0)28 90368341
Email: eurocat@ulster.ac.uk

Final Questionnaire – Questions and Mapping v3.0

Mapping Questions, 'Attributes' and 'Values'

The following tables indicates the way in which each question maps back to the overall statements in Figure 1 and demonstrates 'value' through Importance, Usefulness and Worth. In addition keywords indicating what each response centres on is given to provide a quick reference or cross-check regarding areas under discussion. This information will help when considering analysing and contextualising responses.

Revised Question	Attribute[3]	'Value'[4]	Response is about
1. Which country do you represent?	---	---	---
2. Which of the following best describes who you represent?	---	---	---
3. Would you recommend all new Registries become a member of the EUROCAT network?	Network	Importance	EUROCAT membership
4. **On a scale of 1-5, to what extent is EUROCAT's ongoing CA surveillance important in providing information** on **the following?**	Surveillance	Usefulness	Ongoing surveillance of e.g. clusters, prevalence, risks etc.
5. **Have you accessed information or data provided by EUROCAT (e.g. from the website, EUROCAT publications, provided by your local Registry)?**	Surveillance	Worth	EUROCAT services/information/data
If No, continue to the NEXT PAGE**.**			
If Yes...			
a. For what purpose did you use this information or data?	Surveillance	Usefulness	EUROCAT information/data
b. **On a scale of 1-5, how useful** were **the following for accessing EUROCAT information?**	Data	Usefulness	EUROCAT publications/resources
6. On a scale of 1-5, to what extent are the **following EUROCAT data** quality assurance and standardisation activities important?	Quality	Importance	Methodology and quality mechanisms
7. On a scale of 1-5, to what extent are the following attributes of EUROCAT CA data valuable?	Uniqueness	Importance	Attributes of the EUROCAT database, e.g. populations based, comparable, largest, quality assured
8. **Do you believe there is a continuing need for the work that EUROCAT does**?	Network	Usefulness	Network as a whole
9. **Can you provide an example of how** the information or services provided by **EUROCAT has helped** support or inform your work?	Network	-(impact)-	Examples of how benefit of using EUROCAT data/services

[3] See Figure 1: Four EUROCAT Attributes in the 'Approach' section of this document.
[4] Ibid

Final Questionnaire – Questions and Mapping v3.0

Revised Question	Attribute[3]	'Value'[4]	Response is about
10. Do you have any suggestions for improving the work of EUROCAT?	Network	Worth	Improving services/data
11. Are there any emerging areas should EUROCAT focus on in the future?	Network	Worth	Future activity
12. Any other comments?	---	---	---

Mapping 'Attributes' and 'Value'

Value:	Importance	Usefulness	Worth	Impact
1. Surveillance of congenital anomalies	Q4 Ongoing surveillance	Q5a Usefulness of data	Q5 Access services/info	
2. Assuring data quality and standardisation	Q6 Methodology and QA			
3. Being a European network	Q3 Recommend m/ship	Q8 Continuing need	Q10 Improvements Q11 Future areas	Q9 Example impact
4. Providing unique data and services	Q7 Data attributes	Q5b Publications/resources		

Final Questionnaire – Questions and Mapping v3.0

About Fistral

Fistral Training and Consultancy has been providing highly successful practical training courses and consultancy to many major organisations and universities throughout the UK and abroad since 1991. We specialise in delivering expert tuition and support to all members of an organisation from post-graduate students to board-level management by using our existing management training products or tailoring to specific customer requirements.
Visit Fistral at ***www.fistraltraining.com***

Contact Us

www.fistraltraining.com
+44 (0) 141 636 0290
info@fistraltraining.co.uk

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