

The ERNICA Registry Project and the OCELOT study

Introduction: This communication sets out the relationship between the ERNICA Registry project¹ and the OCELOT study², two separate activities which, while different in their origin and focus, are nevertheless significantly complementary.

ERNICA

As part of its network activities ERNICA has obtained financial support from the European Commission to create a comprehensive registry for all European children with rare inherited diseases and congenital anomalies. This registry will build upon the European Pediatric Surgical Audit (EPSA), that was developed by the Dutch Institute for Clinical Auditing (DICA) in 2014 in collaboration with the Dutch Pediatric Surgical Association. Diseases (eg Hirschsprung's disease, Congenital Diaphragmatic Hernia, Esophageal Atresia and Abdominal Wall Defects) seen in Dutch, new-born children are recorded in this registry.

Part of this project has been to establish a set of internationally applicable quality indicators for evaluation and comparison of the quality of care in EA^{3 4} patients. This is intended to provide insight into variation between hospitals, regions, or countries, and identify areas that could be focused on to improve care. The resulting quality indicator set will be implemented in the EPSA/ERNICA Registry⁵. The final round in the Delphi process to establish this set has recently been completed and an article will be soon finished in a first draft.

This project has been underway since early 2021, and is led by Nadine Teunissen, a clinical researcher at Erasmus MC in Rotterdam and project manager of the EPSA registry. She is supported by Dr Julia Brendel from the department of paediatric surgery at the Medizinische Hochschule Hannover (Hannover Medical School), and Dr Simon Eaton from UCL, London.

OCELOT

OCELOT is a study led by Dr. Rebecca Thursfield, a paediatric respiratory consultant at Alder Hey Children's Hospital, Liverpool. Sponsorship for this project is provided by Alder Hey NHS Foundation Trust and funding has been provided by TOFS, the UK Support group for EA families.

¹ **ERNICA** is the **E**uropean **R**eference **N**etwork for rare **I**nherited and **C**ongenital (digestive and gastrointestinal) **A**nomalies. The network was officially launched in March 2017

² **THE OCELOT STUDY: O**esophageal atresia **C**or**E**outcomes **L**ong**T**erm

³ NB: ERNICA tends to use the American spelling of esophagus, hence (o)esophageal atresia is abbreviated within this communication to EA

⁴ EA patients refers also to those (the majority) born also with tracheo-(o)esophageal fistula)

⁵ See also: <https://ern-ernica.eu/registry/introduction/>

The planning phase for this study commenced in 2021 and the study protocol was approved in March 2022. This study is therefore running 'behind' the ERNICA project, but has recently completed an interesting set of focus groups with patients and parents.⁶

The overall aim of the study is to develop an internationally agreed comprehensive *long-term* Core Outcome Set (COS) for EA relevant to all ages, from birth to transition and adulthood. The study's specific objectives are:

- To determine which outcomes are currently reported in studies of OA-TOF
- To identify which outcomes patients, families and healthcare professionals regard as important following surgical repair for EA
- To prioritise which outcomes patients, families and healthcare professionals think should be included in a COS for EA
- To reach final consensus on the outcomes to be included in the COS

Whilst this study is UK-based it is involving multiple centres and worldwide, including the ERNICA centres.

How are these projects connected?

At the outset of the OCELOT study, it was recognised that its impact would be increased by engaging with appropriate centres and clinicians internationally. Following an initial meeting in October 2021 between Dr Thursfield, Ms Teunissen and Prof. Wijnen (ERNICA Coordinator), it was further recognised that there was a degree of overlap in the projects and that unnecessary duplication could be avoided, for example, by not repeating a literature review. The review performed by N. Teunissen and J. Brendel was shared with R. Thursfield and the OCELOT study group, to reduce their efforts in a new review.

To ensure a collaborative approach, Ms Teunissen and Dr Brendel were invited to join the OCELOT steering group and the OCELOT team was invited to join the process of the Delphi procedure for the quality indicator set.

Significantly the two projects have a common set of stakeholders, i.e. clinicians, researchers and patient/parent representatives.^{7 8}

It should also be noted that the methodological approaches, in particular the use of the Delphi technique, are compatible, and that efforts are being made to ensure that participants in the ERNICA Delphi process are recruited to the forthcoming equivalent OCELOT activity.

⁶ See also: <https://tofs.org.uk/oa-tof-information/oa-tof-research/ocelot-study/>

⁷ In support of the collaboration between the two projects it should be noted that: the lead patient representative for the ERNICA EPSA/Registry project is Anke Widenmann-Grolig, President of EAT (EA Global Support Groups); the OCELOT steering group includes two representatives of TOFS (the UK EA patient group) these being Julia Faulkner (also an EAT Board member), and Graham Slater (also Special Patient Advisor to EAT, and the overall lead patient representative in ERNICA,

⁸ A further aid to collaboration is that Assoc. Prof. Nigel Hall, an EUPSA Executive Board member who leads the EUPSA Network Office, is also a member of the OCELOT steering committee.

Finally, the leads of both these projects will be presenting in the same session at the international EA conference to be held in October 2022⁹. In preparation they will collaborate to ensure their respective updates reinforce the complementary nature of their work, and emphasise the common goal of improving outcomes for EA patients by an agreed set of core outcomes, quality indicators and, in the longer term, an effective widely used registry.

In the coming years several publications and presentations at conferences will be delivered from both projects. Both, ERNICA and OCELOT declared that they will mention the cooperation between both projects in all dissemination such as publications, presentations and flyers and publications on websites and other social media platforms.

⁹ See: <http://www.inoea.org> and/or <https://www.quadconference.com/event/e083dbe3-fcb2-4e91-af46-b7de890e9f6f>