QOMS Rare Benign Lesions of the Jaws registry – Protocol

PROJECT DETAILS

Project Title: QOMS Rare Benign Lesions of the Jaws (RLJ) registry

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RLJ Working Group:

Name	Position / Institution	Email
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Anticipated rollout Date: December 2023 **Review date:** TBC

Funding: British Association of Oral and Maxillofacial Surgeons (BAOMS)

INTRODUCTION

Numerous benign lesions, cysts or solid tumours may present in the jaws. They may be of either odontogenic (tooth-forming, in the dental alveolus) or non-odontogenic (mainly bone) origins in the mandible (ICD10: D16.4) and maxilla (ICD10: D16.5). From a diagnosis perspective, these lesions may have similar imaging features and their location, margins, internal contents, and effects on adjacent structures are important features to diagnose them. These rare benign lesions and tumours of the jaws (RLJ) can vary in behaviour, and despite their benign diagnosis, some can grow rapidly and result in destruction of surrounding structures. Some patients may require complex treatment to adequately treat these lesions/tumours adequately. In recent years / decades, less invasive and adjunctive treatments have become available to reduce the morbidity associated with surgical treatment. The long term safety and efficacy of these emerging modalities are still unclear. As the molecular and genomic pathogenesis of these lesions is better understood, there might yet be the potential for more personalised treatment approaches to optimise treatment strategies for patients.

QOMS

The Quality and Outcomes in Oral and Maxillofacial Surgery (QOMS) project is the quality improvement and clinical effectiveness programme for Oral and Maxillofacial Surgery (OMFS), initiated by the British Association of Oral and Maxillofacial Surgeons (BAOMS).

QOMS operates a series of clinical registries across several OMFS subspecialties (trauma, oncology, reconstruction, non-melanoma skin cancers and orthognathic surgery) as well as the dental specialty of oral surgery, either as audits / service evaluations to measure the quality of care provided to patients or as condition- or procedure-specific registries to look at medium to long-term patient outcomes to guide recommendations for patient treatment and management.

QOMS has an audit / service evaluation for maxillofacial reconstruction, patients affected by RLJ and require reconstruction to treat their jaw defects may already be included in QOMS. However, that registry does not collect specific data on RLJ.

The RLJ registry operates independently from the rest of the QOMS Project, overseen by the QOMS executive team. It will be dedicated solely to this group of rare disease patients.

RATIONALE, AIM & EXPECTED BENEFITS

The presentation of the RLJ through various healthcare professionals (dentists, oral surgeons and, oral and maxillofacial surgeons) in the primary and secondary care in the UK means that it is impossible to gather this new emerging knowledge, validate it in the clinical trial setting and then use it to transform care in the population. The key is to first establish the incidence of these lesions in the population, their distribution and current management. This national service evaluation and its subsequent analysis will create a reference guide on rare jaw tumours to aid diagnosis and open constructive discussions about perspectives on their management.

It is imperative to evaluate the situation, as the last decade has brought new techniques and drugs that can minimise treatment extent and reduce the need for major surgery with its attendant morbidity. The long term outcome of these new modalities will only be evident in the years to come.

The rarity and histological diversity (See Appendix 1 for full list) of RLJ mean that there is a lack of strong evidence to provide recommendations for their management and that randomised control trials (RCT) are not feasible. The next best level of evidence after RCT is observational evidence based on registry data.

A disease-specific registry allows for the collation of real-world data and in time can lead to benefits for patients, surgeons, participating institutions, and commissioners by measuring and improving quality of care. This can be achieved by better understanding the natural history of these tumours, generating future research hypothesis, raising awareness, assessing and informing clinicians and patients about the effectiveness of current treatments, their safety and tolerability profiles, providing a faster diagnosis and design care pathways.

Here, the overarching aim of the RLJ registry is to describe the incidence, distribution and management of rare tumours and lesions of the jaws treated in the UK. Its objectives are to (1) identify the incidence and national distribution of these tumours and lesions in the UK and (2) chart the current management practice of these tumours and lesions in the UK, and long term outcomes of each treatment modality.

The collection of patient reported outcomes (PRO) is also often included and proven to be valuable for disease-specific registries. When considering this aspect for the RLJ registry, we were faced with two problems. First, there is no specific PRO tool for these (rare) conditions, adoption of a head and neck cancer PROM tool was considered but most of them focus on cancer and the quality of life of patients with cancer. Second, the range of possible treatment from ‘simple’ cyst enucleation to tumour resection with or without reconstruction means that patient experience can be very different and therefore difficult to assess reliably with one PRO tool. Thus the Working Group decided that the collection of PRO for the RLJ registry would not be feasible at this time.

INFORMATION GOVERNANCE

These following principles of Information Governance (IG) apply to the Rare Benign Lesions of the Jaws registry:

- The RLJ is NOT a research project but a service evaluation and therefore does not require ethical approval (see Appendix 2). It however still requires approval form participating hospitals' IG departments / Caldicott Guardians…
- The RLJ registry will seek patient consent to collect data, including patient identifiable information (A patient information leaflet and consent form are available in Appendix 3a and 3b, respectively). Any change to the registry that modifies its content which was not included in the initial consent will require re-consenting of patients (for patients already included in the registry).
- The RLJ's lawful basis of processing patient data relies on Article 6 (1) (e) of GDPR: "processing is necessary for the performance of a task carried out in the public interest or in the exercise of official authority vested in the controller…"
- Data is collected directly either by dedicated members of staff (data coordinators) or by surgeons and stored in an instance of the Research Electronic Data Capture (REDCap) system, hosted and managed by the Barts Cancer Research UK Centre (BCC), Queen Mary University of London (QMUL). *The Barts CR-UK Centre (BCC) has a valid NHS Digital DSPT toolkit (EE133904-ECC04) and is ISO 27001 certified (Cert. No. 225111).*
- Data processing: see data flow in Appendix 4.
- Data is initially retained for 10 years **after the end** of collection of follow-up data. This policy will be reviewed on a regular basis.
- Population: Patients **newly diagnosed with** or **currently treated for** a primary or recurring rare lesion or tumour of the jaws.
- Registry access is under access control policy:
    - Local clinical lead(s) of participating departments are given full access (including patient identifiable information) to the records entered in the registry for their own institution only. They are able to view, edit and download that data to use it locally.
    - Access to the whole dataset is limited to the designated data manager (Project Manager), who is a non-clinical member of the QOMS team. Other members of the QOMS team will only have access to anonymised information.
- Access to the central dataset by any third party (individuals/institutions) will require a formal request, via the online data request form. Applicant must demonstrate that they will adhere to relevant information governance regulatory framework. Applications will be reviewed by the RLJ Working Group and the QOMS Information Governance Group. No patient confidential / identifiable information will be shared with third parties.

DATA COLLECTION PROCESS

Consent

The consent process will be set up online. The patient information leaflet will be available both as a printed document and electronically. A copy of the signed consent form will be sent to the patient's

email when a valid email address has been provided. Provision will be made to allow for consenting paediatric patients (parent, carer or legal guardian).

Clinical data

- Data collection will be done directly either by dedicated members of staff (existing QOMS data coordinators) or by surgeons. Each user will be provided with a unique username and password to access the online registry. User access to data will be limited to data collected in their respective institution.
- Patients will be followed up yearly until the registry ends or they decide to opt-out. Initially, the follow up will be set up for 10 years but this might be revised (reduced or extended) at a later date.
- Because of the diversity of those conditions, the range of treatment and the lack of specific PRO tools, it was decided that PRO would NOT be collected.

Forum

As part of the Rare Benign Lesions of the Jaws registry initiative, we intend to develop multidisciplinary collaborations at a regional (Integrated Care Board) and national levels. The quorate requirements for these multidisciplinary team meetings are Oral and Maxillofacial Surgeons (or other specialty involved in the treatment of these conditions), Oral/Head and Neck Pathologists and, Dental and Maxillofacial/Head and Neck Radiologists. Attendance of clinicians involved in care of the patients such as Oral Surgeons, Nurses, Clinical Nurse Specialists and Allied Health Professionals will be welcomed and encouraged. Due to the rarity of these conditions, it is anticipated that these meetings will be held on a quarterly basis through a virtual platform. The registry activities will be reviewed and any related operational issues discussed, in addition to review and discussion of complex patient cases as requested by their primary clinicians.

DATASET

See “Supporting document 1”

PATIENT AND PUBLIC INVOLVEMENT

A patient and public involvement meeting was organised by the Working Group through personal networks. A group of 5 patients and carers with Fabien Puglia met online on September 1st and September 6th, 2023. Prior to the meeting, the project protocol was shared with the group to be used as a starting points for discussions.

The aims of the meeting was for patients and carers to:

(1) Demonstrate that they were satisfied with the rationale, objectives and processes of the registry and

(2) Provide feedback to the working group on the registry form (e.g., scope of the dataset) and content (e.g., readability of the patient information leaflet).

In summary, patients and carers thought that creating a Rare Benign Lesions of the Jaws registry is positive and could see the benefits to patients, healthcare professionals and commissioners. A summary of the feedback from the group is provided in Appendix 5.

DATA OWNERSHIP

Participating organisations will retain the ownership of the data they entered, while the ownership of the central dataset will be BAOMS. BAOMS will curate data on behalf of participating organisations.

PUBLICATION POLICY

The British Journal of Oral and Maxillofacial Surgery (BJOMS) will have first refusal of any peer reviewed output from this initiative.

Individuals responsible for collecting data will be acknowledged as "collaborators" and listed in publications.

Document control version

Version / Date	Description	Sign off by / Date
1.0 – 22/06/2023	Initial protocol	
1.2 – 07/06/2023	Modifications to include comments and suggestions raised during PPI meeting	
1.3 – 20/09/2023	Proofread	
1.4 – 25/01/2024	Clarification between OMFs and OS	

APPENDICES

APPENDIX 1. LIST OF CONDITIONS COVERED BY THE REGISTRY

Source: Diagnosis (WHO)

Benign epithelial odontogenic tumours

- Ameloblastoma
  - **Ameloblastoma, unicystic type**
  - **Ameloblastoma, extraosseous/peripheral type**
  - **Metastasizing ameloblastoma**
- **Squamous odontogenic tumour**
- **Calcifying epithelial odontogenic tumour**
- **Adenomatoid odontogenic tumour**

Benign mixed epithelial and mesenchymal odontogenic tumours

- **Ameloblastic fibroma**
- **Primordial odontogenic tumour**
- Odontoma
  - **Odontoma, compound type**
  - **Odontoma, complex type**
- **Dentinogenic ghost cell tumour**

Benign mesenchymal odontogenic tumours

- **Odontogenic fibroma**
- **Odontogenic myxoma/myxofibroma**
- **Cementoblastoma**
- **Cemento-ossifying fibroma**

Malignant maxillofacial bone and cartilage tumours

- **Chondrosarcoma**
- **Mesenchymal chondrosarcoma**
- **Osteosarcoma**

Benign maxillofacial bone and cartilage tumours

- **Chondroma**
- **Osteoma**
- **Melanotic neuroectodermal tumour of infancy**
- **Chondroblastoma**
- **Chondromyxoid fibroma**
- **Osteoid osteoma**
- **Osteoblastoma**
- **Desmoplastic fibroma**

Fibro- and chondro-osseous lesions

- **Ossifying fibroma**
- **Familial gigantiform cementoma**
- **Fibrous dysplasia**
- **Cemento-osseous dysplasia**
- **Osteochondroma**

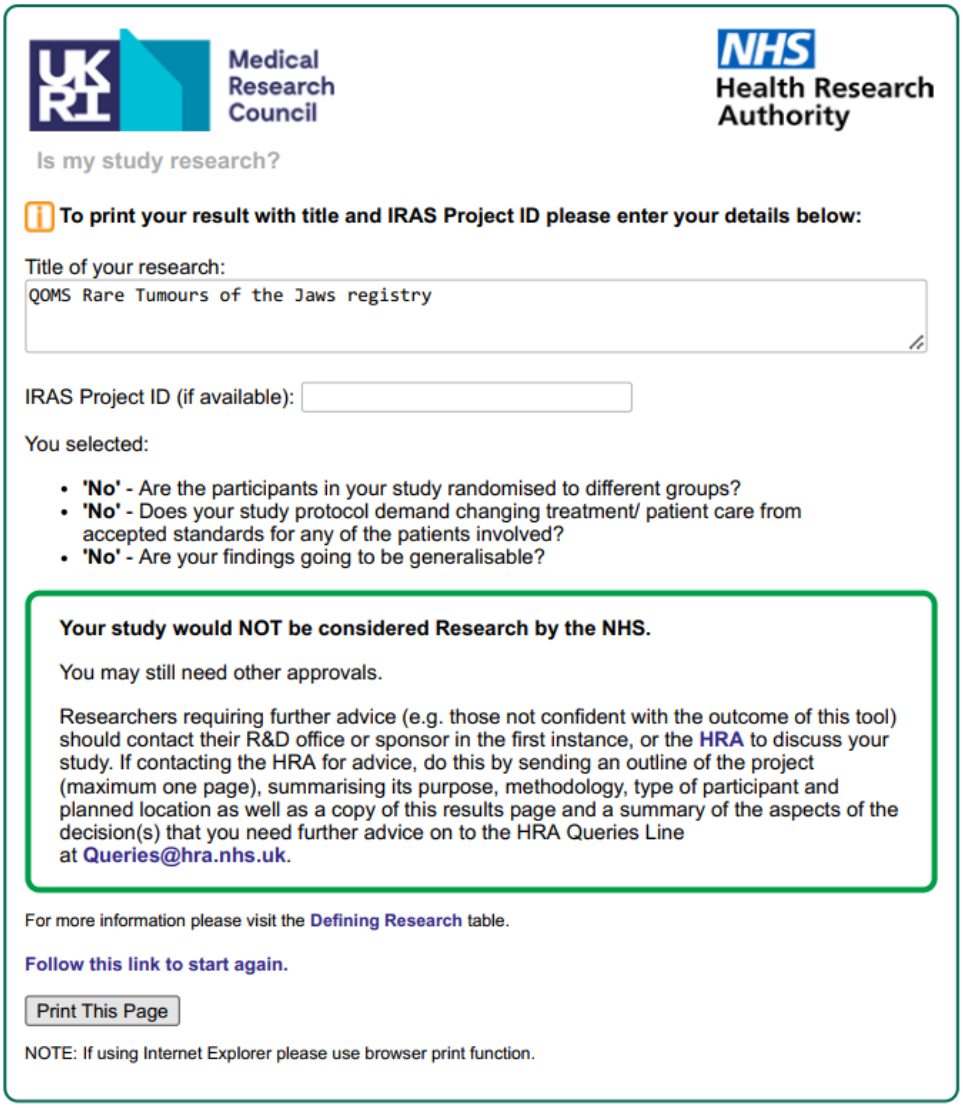
Giant cell lesions and bone cysts

- **Central giant cell granuloma**
- **Peripheral giant cell granuloma**
- **Cherubism**
- **Aneurysmal bone cyst**
- **Simple bone cyst**

Haematolymphoid tumours

- **Solitary plasmacytoma**

APPENDIX 2. HRA MRC TOOL KIT "IS MY STUDY RESEARCH?"



Medical Research Council

NHS Health Research Authority

Is my study research?

To print your result with title and IRAS Project ID please enter your details below:

Title of your research:

QOMS Rare Tumours of the Jaws registry

IRAS Project ID (if available):

You selected:

- **'No'** - Are the participants in your study randomised to different groups?
- **'No'** - Does your study protocol demand changing treatment/ patient care from accepted standards for any of the patients involved?
- **'No'** - Are your findings going to be generalisable?

Your study would NOT be considered Research by the NHS.

You may still need other approvals.

Researchers requiring further advice (e.g. those not confident with the outcome of this tool) should contact their R&D office or sponsor in the first instance, or the HRA to discuss your study. If contacting the HRA for advice, do this by sending an outline of the project (maximum one page), summarising its purpose, methodology, type of participant and planned location as well as a copy of this results page and a summary of the aspects of the decision(s) that you need further advice on to the HRA Queries Line at Queries@hra.nhs.uk.

For more information please visit the Defining Research table.

Follow this link to start again.

Print This Page

NOTE: If using Internet Explorer please use browser print function.

APPENDIX 3A. PATIENT INFORMATION LEAFLET

Version 1.0 Date: 20/09/2023

You have been given this leaflet because you have been diagnosed with a rare benign lesion or tumour of the jaws. The surgeons and other health professionals who care for you would like to invite you to take part in this registry dedicated to similar conditions to yours.

Please read this leaflet carefully. It explains who we are, what we are doing and how we treat your information to ensure confidentiality and anonymity.

WHY WAS I GIVEN THIS LEAFLET?

Benign lesions and tumours of the jaws are a rare and relatively varied group of conditions. Because they are rare, they are not very well understood and there are various recommendations for their management. One possible approach to this challenge is to collect information about these conditions in a specialised registry. A clinical registry collects organised information about patients affected by a condition and the treatment received. This can be used to find patterns in disease presentation, treatment outcomes and ultimately improve patient care.

WHY ARE YOU COLLECTING THIS INFORMATION?

We would like to find out how your condition was treated and followed up, if you have experienced any complications or recurrence. We would like to understand how frequent these tumours are and how they are treated in the UK. We hope this information about patients and their treatment will help clinicians and healthcare commissioners understand more about the best treatment for those tumours and improve care for patients in the future.

WHAT WOULD TAKING PART INVOLVE?

Taking part in this registry will not take up any of your time after you have consented to the study. Your surgical team will collect data directly from your medical records and pass it onto us securely.

WHAT INFORMATION ABOUT ME ARE YOU COLLECTING?

To be able to follow-up your progress over time, we need to collect your NHS number, date of birth and information about your conditions, treatments, and long-term outcomes. We would also like your permission to access and share any imaging or biological specimens routinely collected as part of your diagnosis, treatment or follow-up, to be reviewed by a panel of experts and potentially used for secondary research.

WHAT WILL HAPPEN TO MY INFORMATION?

Your information will be collected and stored on secure computers managed by the Barts Cancer Research UK Centre at Queen Mary University of London (BCC, QMUL). Access to your information will be restricted to your clinical team and a limited number of approved members from QMUL and the project team. No identifiable information will be shared.

IS MY INFORMATION SAFE?

Yes, your information is safe. Very strict rules and secure procedures are in place to ensure that your information is kept safe. The systems and procedures in place at QMUL comply with international standards and QMUL continuously monitor and adapt them as necessary to maintain security over the lifetime of the project.

Because this information is valuable, it may also be used for secondary research e.g. evaluation of treatment outcomes, surveillance strategy and translational studies. Should this be the case, data that can directly identify you (e.g. NHS or CHI number) will never be shared.

HOW LONG WILL MY DATA BE KEPT FOR?

Your information will be kept for 10 years after the end of data collection. Afterwards, it will either be anonymised (i.e., NHS number, date of birth… will be deleted) or completely deleted.

Can I not take part to this registry?

Participation is voluntary and you can change your mind at any time without it affecting the care that you receive.

If you decide to not take part, when you complete the consent form, simply select "I do not agree". This way, we will keep a record of your decision and we will not ask you again at a later stage.

If you change your mind about taking part, you can withdraw at any point without providing any reasons. Simply contact your treating team or email the project team and put "Opt-out" in the subject line (see email address below). You will be asked whether you want all your information removed or whether you are happy for us to keep the information we have so far but no new information will be collected.

Who is organising and funding this study?

This project was designed by oral and maxillofacial surgeons in collaboration with pathologists. The British Association of Oral and Maxillofacial Surgeons (BAOMS) leads this project and as data controller, is responsible for looking after your information and using it appropriately. The costs for the project are being supported by BAOMS (Registered charity number: 1062067).

Who has reviewed this initiative?

This project has been reviewed by clinicians and a group of patients and carers, and the audit department of this hospital and authorised by this hospital for data protection and security prior to their participation.

What if there is a problem?

You also have the right to lodge a complaint with the Information Commissioner's Office (ICO), the supervisory authority in the UK responsible for the implementation and enforcement of data protection law, if you have concerns about the way your personal data is being handled. You can contact the ICO via telephone (0303 123 1113) or email (W: https://ico.org.uk/concerns/).

Finding out more

If you would like further information or have any questions, please contact:

British Association of Oral and Maxillofacial Surgeons | Royal College of Surgeons of England, 38/43 Lincoln's Inn Fields, London WC2A 3PE | Project Team's email: qoms@baoms.org.uk | W: https://bit.ly/qoms-at-baoms

APPENDIX 3B. PATIENT CONSENT FORM

Version 1.0 Date: 20/09/2023

Consent form for patients aged 16 years and above, deemed to have capacity to consent

Before signing this consent form, please read the accompanying patient information leaflet (version: XX, Date: DD/MM/YYYY) carefully and ask questions to your clinical team. Once you are satisfied, please complete the consent form below to show whether or not you consent to the collection of your personal information and sign this form.

Please **initial** the boxes below

1. I confirm that I have read and understand the patient information leaflet (version XX, date: DD/MM/YYYY) describing the registry and potentially associated work and have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I am fully aware that the project collects personal information about me and that I will remain anonymous. ☐
4. I understand that I have the right to withdraw my consent at any time without giving a reason and that my care will not be affected. ☐
5. I agree to having my personal health data stored in this database at the Barts' Cancer Centre ☐
6. I agree to have images and samples of my blood/tissue collected from any procedures that would have been undertaken as part of my treatment, to be accessed for review and secondary research. ☐
7. I understand and agree that data from the study can be used in future research and that data would be completely anonymised. ☐
8. I am fully aware that data collected will be stored securely, safely and in accordance with Data Protection Act (2018) and the General Data Protection Regulation (GDPR). ☐
9. a. I AGREE to take part in this project and for my information to be collected. ☐

   b. I DO NOT AGREE to take part in this project and for my information to be collected ☐

_____	_____	_____
Name of Participant	Signature	Date
_____	_____	_____
Name of the person taking consent	Signature	Date

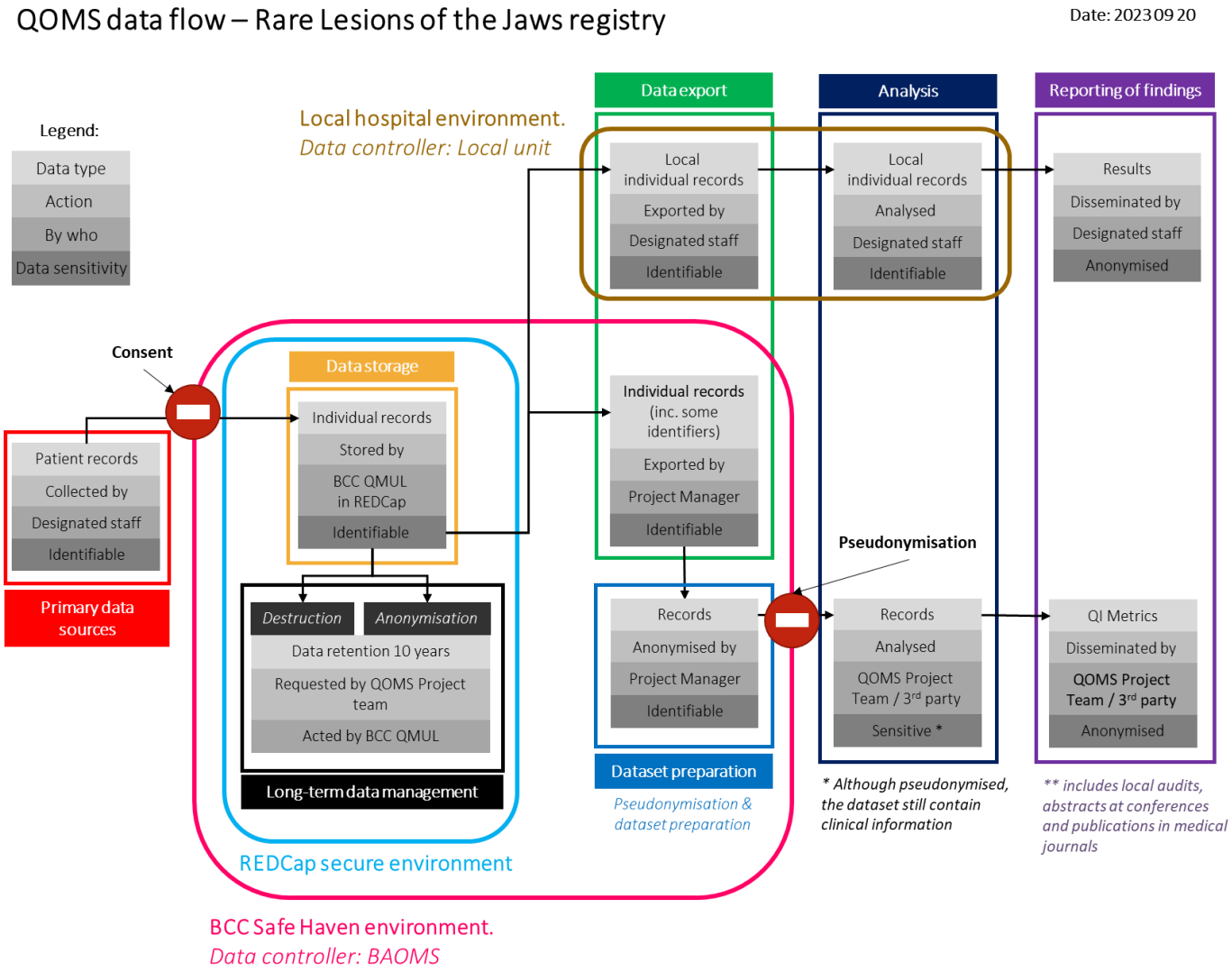
If you would like further information or have any questions, please contact:

BAOMS | Royal College of Surgeons of England, 38/43 Lincoln's Inn Fields, London WC2A 3PE |Project Team: qoms@baoms.org.uk | W: https://bit.ly/qoms-at-baoms

One copy of this form should be given to the patients, one copy kept in the patient's note and the original copy kept by the treating team.

Note: provision will be made to allow for consenting of paediatric patients (less than 16 years of age)

APPENDIX 4. DATA FLOW



APPENDIX 5. PATIENT AND PUBLIC INVOLVEMENT PANEL DISCUSSIONS

Area of concerns / discussions	Discussion points	Answers / Action points
Raising awareness of RLJ to Primary Care Dentist	Engage with the BDA and FDS when launching the registry	Yes
Engagement with dentists	Engage with PCD more widely so they can understand and see the patient's treatment…	Yes
Long-term patient / participant engagement	To contact by email patients / participants on a yearly basis with an update about the RLJ registry (e.g. recruitment, achievement…). This yearly update could be used to see if some patients would like to get in touch with each other to discuss their experience…	Yes
Long-term patient / participant engagement	To have resources available for patients about their conditions and treatment (long-term)	Yes. to be considered in the future
Data collection	Source of initial referral (PCD, GP, Hospital, Other specialty...)	Yes
Data collection	Capturing the time from 1st referral to diagnosis and possible to definitive treatment	Yes
Data collection	PROM	Yes, to be reviewed in the future
PIL & Consent form	• Change the "Where are you collecting this information?" as discussed. • Make it clearer that this email address is the project team's. • Modify the consent form to allow for paediatric consent. • Make the PIL less wordy / add pictures?	Done Done Done To be considered