

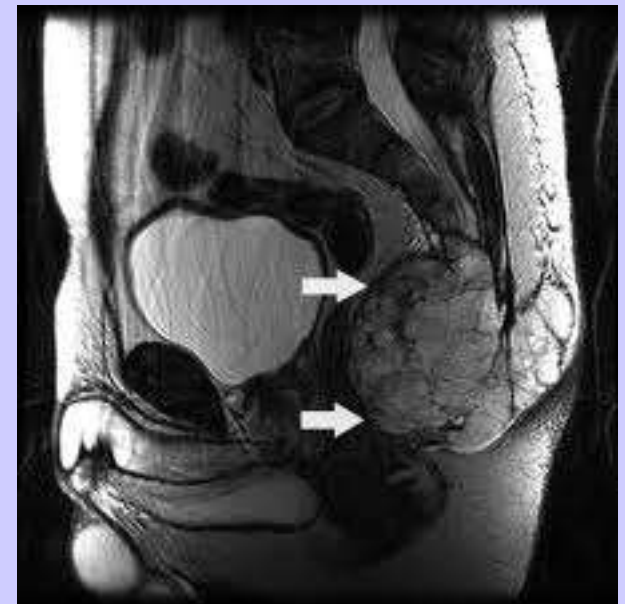
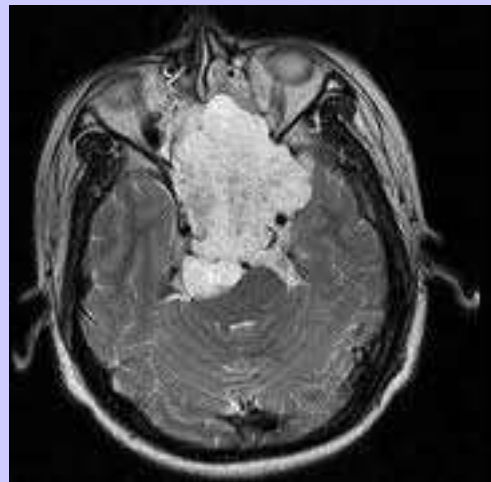
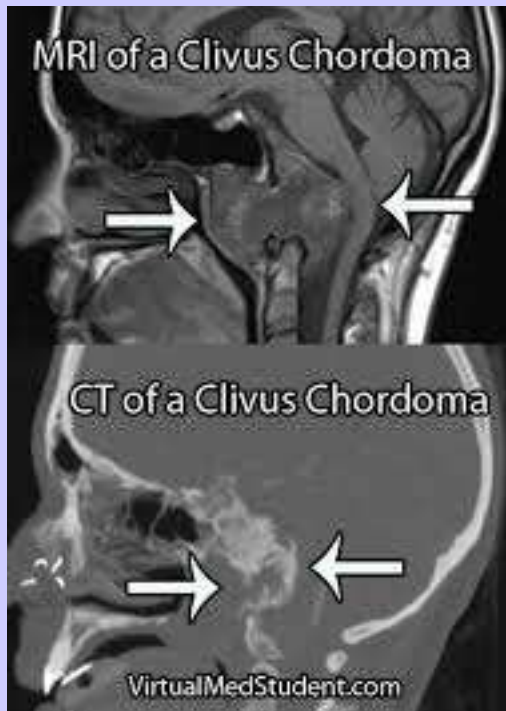
A photograph of several surgeons in an operating room, wearing blue scrubs and masks, focused on a surgical procedure. The scene is brightly lit, and the surgeons are using various instruments. The text is overlaid on the center of the image.

# Prospective chordoma patient database: a European initiative

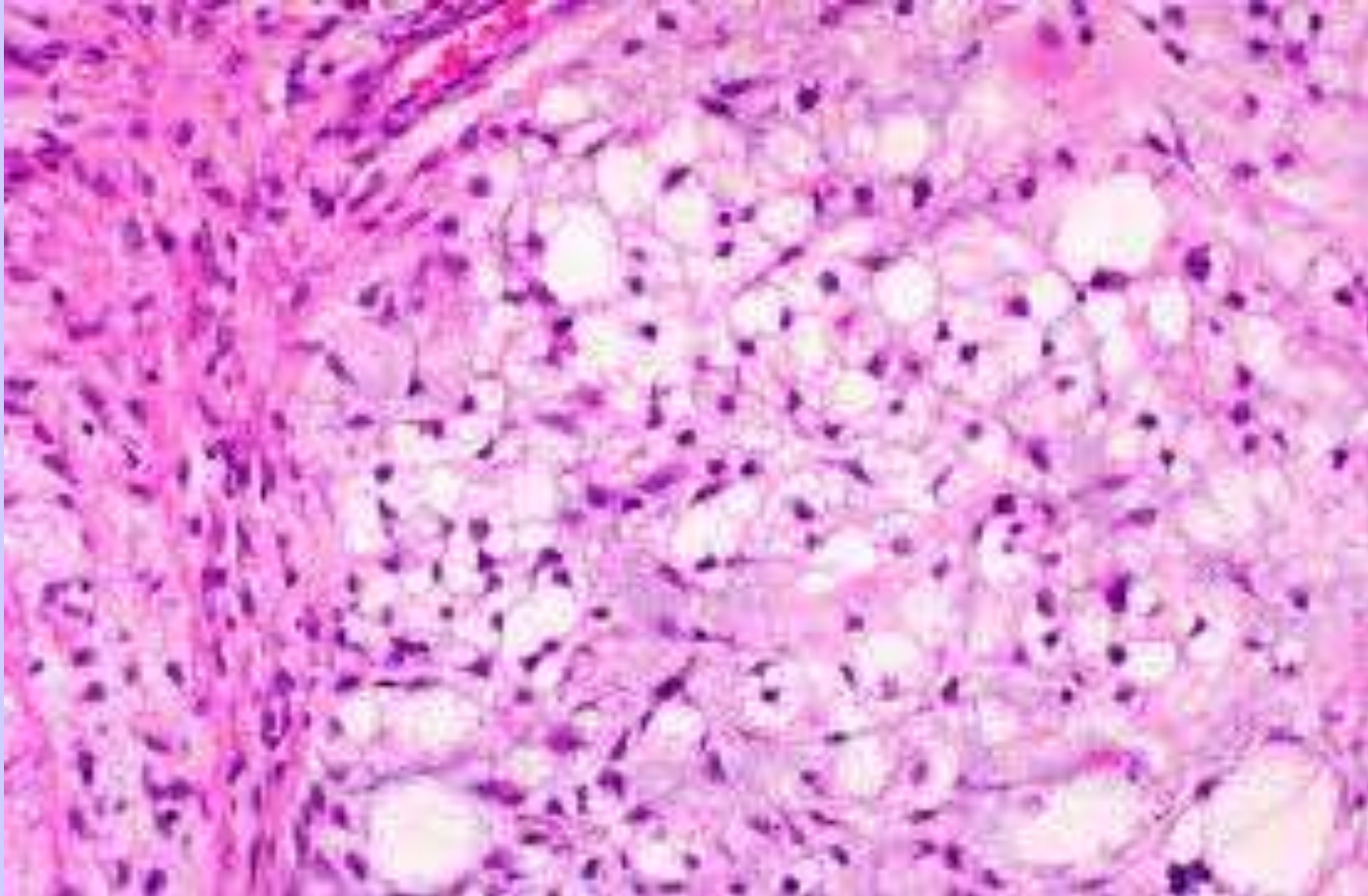
Sierk H. Bakker BSc<sup>a</sup>; Hans Gelderblom MD, PhD<sup>b</sup>; Pancras C.W. Hogendoorn MD, PhD<sup>c</sup>;  
Wilco C. Peul MD, PhD<sup>a</sup>; P.D. Sander Dijkstra MD, PhD<sup>d</sup>; Herman M. Kroon MD, PhD<sup>e</sup>; Remi  
A. Nout MD, PhD<sup>b</sup>; Hans Keulen MIM<sup>f</sup>; Judith V. Bovée MD, PhD<sup>c</sup>;  
Carmen L.A.M. Vleggeert-Lankamp MD, MSc, PhD<sup>a</sup>

- Doctor's problem:
  - Challenging surgery
  - Post surgery treatment is not well-described
- Researcher's problem:
  - Incidence is low
  - Outcome is variable

- Doctor's problem:



- Researcher's problem:





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- Herman M. Kroon MD, PhD: radiologist
- Remi A. Nout MD, PhD: radiotherapist
- Hans Keulen MIM: Chordoma Foundation
- Judith V. Bovée MD, PhD: pathologist
- Carmen L.A.M. Vleggeert-Lankamp MD, MSc, PhD: neurosurgeon

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- Herman M. Kroon MD, PhD: radiologist
- Remi A. Nout MD, PhD: radiotherapist
- Hans Keulen MIM: Chordoma Foundation
- Judith V. Bovée MD, PhD: pathologist
- Carmen L.A.M. Vleggeert-Lankamp MD, MSc, PhD: neurosurgeon



# Where do we stand?

Results: 1 to 20 of 330

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## **A comprehensive analysis of intracranial chordoma and survival: a systematic review.**

Jian BJ, Bloch OG, Yang I, Han SJ, Aranda D, Parsa AT.

Department of Neurological Surgery, University of California at San Francisco, San Francisco, California, USA.

### **Abstract**

**OBJECTIVE:** Despite the published information on cranial chordoma, most of the data regarding survival in these patients has come from a single institution. Here, we perform a systematic review of the literature to evaluate across multiple institutions the overall survival after treatment for intracranial chordoma.

**MATERIALS AND METHODS:** We systematically analysed every study published in English and found a total of over 2000 patients being treated for intracranial chordoma. The overall 5-year and 10-year survivals in these patients were stratified according to the age (<5 years vs. >5 years and <40 years vs. >40 years), treatment (surgery and radiation vs. surgery alone) and histological findings (chondroid vs. typical). Data were analysed via Pearson chi-square test and student t-test when appropriate.

**RESULTS:** A total of 560 non-duplicated patients treated for cranial chordoma met inclusion criteria for this systematic analysis. The survival rate among these patients was 63% (299 patients) and 16% (176 patients) for 5-year and 10-year survivals, respectively. There was no difference in overall survival between the two groups when a cut-off age of 40 years was used (<40 years=50% vs. >40 years=51% at 5-year survival;  $p=0.1$ ), but when 5 years was used as the cut-off age, then survival was better for patients in the group older than 5 years of age (<5 years=14% vs. >5 years=66%;  $p=0.001$ ). There was no difference between 5-year survival in patients with chordoma with histological chondroid features and those with chordoma possessing typical histology (45% vs. 67%;  $p=0.06$ ). When patients who only received surgery were compared to those patients who were treated with surgical intervention in combination with adjuvant radiation treatment, no difference in survival rate was found (54% vs. 56% at 5 years;  $p=0.8$ ).

**CONCLUSION:** The results of our systematic study provide data to predict the survival of intracranial chordoma patients across multiple institutions. Our data suggest that patients younger than 5 years of age may be associated with a worse prognosis, and adjuvant radiation therapy and histological type were not associated with the improvement of survival rates.

## Retrospective study of cases in the Netherlands

National pathology database

Find the patients

Gather data

Study follow up of those patients

Reconsider pathological diagnose

Evaluate 'medical pathway'

Next step is prospective research:

Conventional Reasoning:

Studies must have an adequate size ,  
usually hundreds/thousands of patients.

But:

Incidence of Chordoma < 1:1 million

Prevalence of Chordoma < 1: 150.000

(Paolo Bruzzi at Chordoma meeting in Amsterdam, June 21, 2012)



**This won't work**



- An international, prospective patient database can greatly enhance the number of patients being enrolled in trials
- Look for large treatment effects
- Recent statistical developments (< 10 years):

Amsterdam, June 21, 2012

## Methodology of Clinical Studies

Paolo Bruzzi  
Clinical Epidemiology Unit  
National Cancer Research Institute  
Genova - Italy



- Quality control mechanism
  - It sets a reference point
  - First: one can compare to the reference point
  - Second: (Anonymously) compare inter-institution differences in patient characteristics and outcome



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### Perspective

### What Is Value in Health Care?

Michael E. Porter, Ph.D.

N Engl J Med 2010; 363:2477-2481 | December 23, 2010 | DOI: 10.1056/NEJMp1011024

- Many issues to tackle
  - Which data?
  - Entry and maintenance of data
  - Access and ownership of data
  - Realization of a system

- Which data:

- Exhaustive list of parameters drafted by LUMC sarcoma team (syst review)
- List will be put online in questionnaire form for physicians present at Amsterdam/Prague meetings
- Results of the questionnaire will determine definition of data set

- Entrance and maintenance of data:
  - Physician treating the patient is responsible for entry of data
  - Start off with limited number of specialized treatment centers, build from there
  - Compliance with legislation on security, e.g. NHS, NEN 7510, HIPAA

- How to motivate for entrance and maintenance of data?
  - Only participants can use the data in the system
  - Participants get status of “preferred supplier” on CF’s doctors advisory
  - Money (e.g. from Rare Cancers Europe FP7 request)



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Patient participation

## MijnZorgnet.nl

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### Op MijnZorgnet.nl komen patiënten en zorgverleners samen

Als patiënt bouwt u hier uw eigen zorgnet, een beveiligde ontmoetingsplek waar het draait om uw gezondheid. Hier kunt u uw medische gegevens opslaan en beheren en u bepaalt u zelf met wie u deze informatie deelt. Ook kunt u zich aansluiten bij groepen, waaronder diverse digitale poli's of praktijken, die voor u belangrijk zijn. Als zorgverlener kunt u binnen MijnZorgnet een digitale poli of praktijk starten. Dit zijn besloten groepen voor uw patiënten en uw team. Daarnaast kunt u zich aansluiten bij het zorgnet van uw patiënt of deelnemen aan groepen die voor u van belang zijn.



### Patiënt

Als patiënt wilt u dat het gaat over uw eigen gezondheid. Contact hebben wanneer dat ú uitkomt. Snel antwoord op uw vragen.

#### Wat kunt u op MijnZorgnet

- ✓ Praten over uw zorg in een veilige omgeving
- ✓ Contact leggen met uw zorgverleners
- ✓ De regie hebben over uw zorgproces

Meer informatie



### Zorgverlener

Als zorgverlener wilt u meegaan in de ontwikkeling van de digitale zorg. Contact hebben met uw patiënten en hun vragen efficiënt beantwoorden.

#### Wat kunt u op MijnZorgnet

- ✓ Uw eigen digitale poli of praktijk starten
- ✓ Vragen van patiënten gemakkelijk beantwoorden
- ✓ Overleggen met collega's

Meer informatie

- Access and ownership
  - Individual patient data accessible only to patient and physicians treating the patient
  - Data is owned by the patient
  - Contributing institutions can request patient information for prospective studies/trial enrolment, provided that informed consent is acquired

- Realization of a system
  - Web application is implied
  - Connect with existing database structure:
    - Eurobonet
    - ProMISe
  - Funding
  - Differences in legislation:
    - If successful in Europe, extend to the U.S.?

- Goals
  - Trial enrolment
  - Tissuebanking
  - Value in healthcare
  - Quality control
- Many issues to tackle
- Work in progress:  
database online in 2013/2014



