


BIRTH DEFECT CLUSTER INVESTIGATIONS: THE TEXAS EXPERIENCE AND SOME GENERAL QUESTIONS

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Outline

- Stages of a birth defects cluster investigation
- Summary of TX investigations conducted to date
- Some general questions
- Some lessons learned

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Included in This Talk


Cluster investigations

- When occurrence of birth defects initiates concern
- Most focus of this talk

Small area investigations

- When some exposure initiates concern

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Stages of a Birth Defects Cluster Investigation (the Texas Protocol)

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1: Gather Initial Information

- Gather and provide information
- Criteria to proceed:
 - 3+ cases of the same defect or defect group

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2: Are There More Cases Than We Expect?

- Assume all reported cases valid.
- Define time/place bounds, comparison area.
- Calculate rates using simple statistical software.
- Criteria to proceed:
 - Rate in target area is higher than comparison area AND

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3A: Define "Case", Find and Verify Cases



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Case Definition

- Diagnosis, time, place


Case Finding and Verification

- Use Registry if possible (MUCH easier)
- Otherwise use vital records, hospital records, service providers

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3B: Consider Environmental Info




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- In a cluster investigation:
 - Usually only if informant mentions a specific environmental concern
- In a small area investigation:
 - Can be very helpful
- Work with partners (e.g. environmental epi staff)

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3C: Describe Cluster, Compare Rates



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- Descriptive epidemiology:
 - Use final case definition
 - Person, place, time
 - Esp for small area investigations, see if cases live nearby
- Compare crude rates
- Adjust for established risk factors:
 - Often use indirect standardization, may use direct

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Criteria to Proceed

- Birth defects are the same or same type
- Rate in target area is higher than comparison area (or ratio > 1.00)
- Difference between rates is statistically significant
- Either of these is true:
 - 3+ cases with biologically plausible exposure
 - OR
 - 5+ cases and observed rate > 10x expected

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4: Feasibility Study

- Consider:
 - Possible study designs, power
 - Any modifications to case definition
 - Additional case finding, verification, controls
 - Data collection methods
 - Resources required and available
- Criteria to proceed: If warranted

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5: Try to Find What Caused the Cluster

- Base on feasibility study
- Use most cost-effective approach

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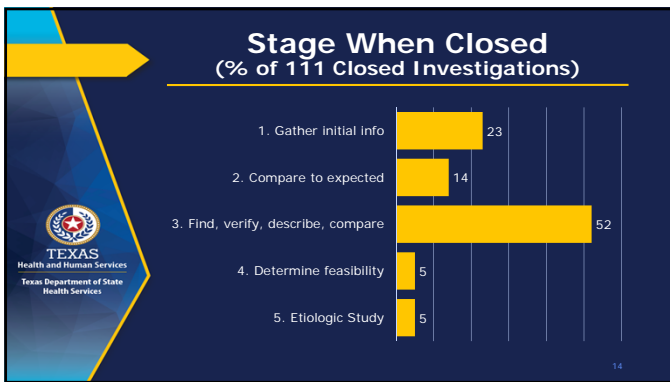


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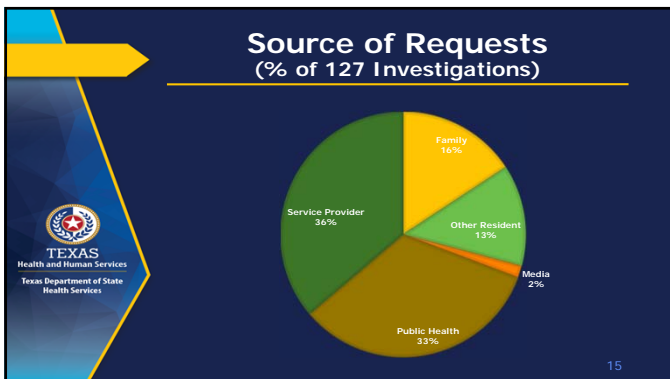
Summary of Investigations Conducted to Date

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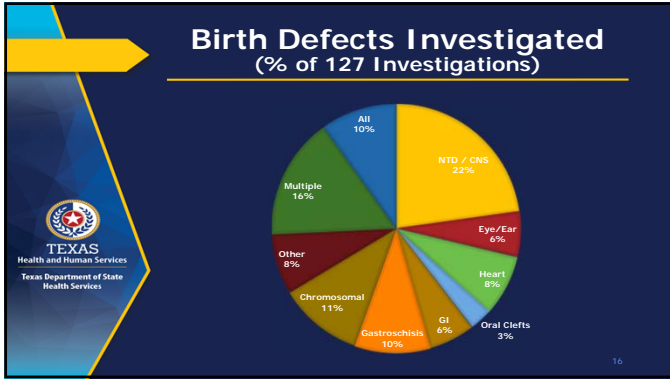
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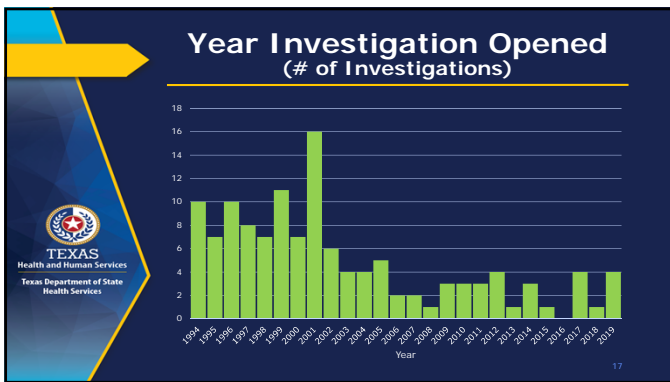
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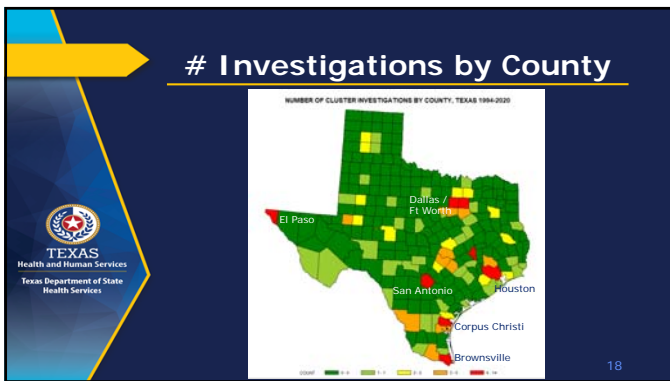
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Trisomy 18 Concern – Brazos Co

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Trisomy 18 by Estimated Date of Conception

Brazos County Fire, 7/30/2009

Surrounding Counties

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Some General Questions

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What Should Be the Comparison Area?




Some options:

- Registry area or state
- Rest of region containing the target area
- If very small target area: Rest of city or county containing the target area
- County or city similar demographically
- Other?

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The Challenge of Diagnostic Variability




- Some defects consistently diagnosed & recorded
 - Examples: anencephaly, gastroschisis
- Some vary with hospital, clinical practice, training, use of specialized equipment
 - Examples: ASD, microcephaly
 - Apparent "clusters" may be due to many

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Which Birth Defects Should be Investigated?




If specified by informant, use those

If not specified (especially in small area investigations):

- Short list of defects?
- Hand pick some? E.g. low diagnostic variability defects?
- NBDPN defects or annual report defects?
- All defects collected?

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Some Resources

- **Data collection**
 - Vital records (info on covariates, on denominators)
 - Epi Info: <https://www.cdc.gov/epiinfo/index.html>
 - Environmental Public Health Tracking
- **Data analysis**
 - WINPEPI (PEPI for Windows): <http://www.brixtonhealth.com/pepi4windows.html>
 - Epi Info: <https://www.cdc.gov/epiinfo/index.html>

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Should We Keep Doing These Investigations?

| CONS | PROS |
|---|---|
| Require significant time (e.g. wait for Registry) or resources; | Public health duty / try to address concern of residents |
| Rarely find cause of the cluster or easily interpretable link of envtl concern with BDs | May alleviate concern if occurrence: <ul style="list-style-type: none"> • Not significantly elevated • Not associated with envtl factor |
| Very rarely find a new cause of birth defects | May help keep BDs in public eye May help with program funding |

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Identify Areas Proactively as Well as Reactively?

- If previous answer = yes... Consider:
 - Worth resources? (staff time, etc.)
 - If do repeatedly, good to have protocol
 - One potential software program: SaTScan (Space And Time Scan Statistic)

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Some Lessons Learned

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Methodology

- Protocol can be helpful
- Probably don't do all birth defects
- Investigation MUCH easier using Registry data than other sources
 - Cases already found and verified
 - Comparison rates easy to calculate

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Communication

- Educate informants early; manage expectations
- Keep as open as possible with informants and media throughout investigation
- Get training in risk communication and dealing with the media

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Thanks



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- Program web site:
<https://www.dshs.state.tx.us/birthdefects/>

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