EVALUATION OF THE NEURAL TUBE DEFECTS SURVEILLANCE SYSTEM IN MEXICO CITY, 2012

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• Birth defects that result from the failure of the neural tube to close during embryogenesis.
  – *Anencephaly, encephalocele and spina bifida (meningocele and myelomeningocele)*

• Mexican Official Norm NOM-017-2012-SSA2 for Epidemiological Surveillance
  • *Special subsystem for Neural Tube Defects within Birth Defects.*

• 1993 → Neural Tube Defects Surveillance System (NTDSS).

**Objective:**
• *To identify the cases of Neural Tube Defects, evaluate changes and trends on its presentation.*
  • *Detect groups with elevated risk → Prevention and control actions.*

• Manual outlining the guidelines for the functioning of the surveillance system.
  • *Updates → 2005 and in 2012*
Neural Tube Defects Surveillance System

- **Local Level**
  - Neural Tube Defect case
  - Classification
  - Neural Tube Defect case
  - Weekly notification
  - Reference for specific treatment
  - Monthly notification
  - Negative or Positive
  - Monthly notification
  - Weekly notification
  - Physical examination
  - Imaging studies
  - Onset

- **Jurisdictional Level**
  - Collect, process, analysis, and report (max., 10 days)
  - Mother Follow up report (6 months)

- **State Level**
  - Collect, process, analysis, disseminates and report (max., 5 days)

- **National Level**
  - Collect, analysis and dissemination
  - Federal District
    - 16 health jurisdictions
    - Total:
      - 552 health units
      - 561 hospitals (81% private)
  - 32 states
    - 234 health jurisdictions
    - 18,185 health units
    - 4,259 hospitals (68% private)
  - 80% (448) attended births in 2012

**Flowchart**

1. **At birth**
   - Neural Tube Defect case
   - Classification
   - Monthly notification
   - Reference for specific treatment
   - Negative or Positive
   - Weekly notification
   - Physical examination
   - Imaging studies

2. **Report**
   - 0 to 60 days

3. **Feedback**
   - Collect, analysis and dissemination
   - Collect, process, analysis, disseminates and report (max., 5 days)
   - Collect, process, analysis and report (max., 10 days)
   - Mother Follow up report (6 months)
   - Monthly notification
   - Report
     - (Max. 30 days after birth)
   - Weekly notification
   - Negative or Positive
   - Reference for specific treatment

**Legend**
- **Blue Arrow**
  - Feedback
- **Green Box**
  - Collect, analysis and dissemination
- **Gray Box**
  - Collect, process, analysis, disseminates and report (max., 5 days)
- **Pink Box**
  - Collect, process, analysis and report (max., 10 days)
- **Light Blue Box**
  - Mother Follow up report (6 months)
- **Yellow Box**
  - Report
    - (Max. 30 days after birth)
- **Orange Box**
  - Monthly notification
- **Red Box**
  - Negative or Positive
- **Green Box**
  - Reference for specific treatment
- **Yellow Box**
  - Weekly notification
- **Green Box**
  - Physical examination
- **Green Box**
  - Imaging studies

**References**
- Anencephaly
- Encephalocele
- Meningocele
- Myelomeningocele
Neural Tube Defects Surveillance System

Formats and Sources

Case report format
Monthly report format
Mother follow up Report format

Sources:
• Hospital registries.
• Death Certificates.
• Fetal Death Certificates.
• Birth Certificates.
Objectives

• **General objective:**
  – Assess the attributes and usefulness of the Neural Tube Defects Surveillance System in Mexico City during 2012.

• **Specific objectives:**
  – Assess the qualitative attributes of NTDSS such as: acceptability, flexibility, and simplicity.
  – Assess the quantitative attributes of NTDSS such as: data quality, timeliness, representativeness, sensitivity and positive predictive value.
  – Assess the usefulness of the NTDSS.
  – Make recommendations to improve the quality, efficiency and usefulness of NTDSS.
Methodology

• CDC ➔ Guidelines for Evaluating Public Health Surveillance Systems \(^1\).

• Evaluation method proposed by Batista – Gonzalez\(^2\).

• Staff interviews ➔ national, state, jurisdictional and local level.

• Medical record review.

• Database:
  • Cases reported to the system.
  • Birth certificates.
  • Death certificates.
  • Fetal death certificates.

• Manuals, publications and report reviews.

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Methodology

- **Staff interviews** (qualitative attributes):
  - **Acceptability**: 5 – 6 questions.
  - **Simplicity**: 9 – 10 questions.
  - **Flexibility**: 3 questions.

- **Acceptability**: interview score, notification rate, format integrity, timeliness of notification.

- **Simplicity**: interview score, type and amount of data needed to fill the form, methods for data collection, analysis and dissemination, and monitoring of cases.

- **Flexibility**: interview score, case report form analysis, ability to adapt of the system.

<table>
<thead>
<tr>
<th>Table 1. Score and Categories of Qualitative Attributes</th>
<th>Score at Local level (17 questions)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Attribute</td>
<td>Poor</td>
</tr>
<tr>
<td>Acceptability</td>
<td>0-6</td>
</tr>
<tr>
<td>Flexibility</td>
<td>0-3</td>
</tr>
<tr>
<td>Simplicity</td>
<td>0-11</td>
</tr>
</tbody>
</table>

| Score at Jurisdictional and State level (18 questions) |
|--------------------------------------------------------|-----------------------------------|
| Attribute                                              | Poor | Fair | Good |
| Acceptability                                          | 0-7  | 8-16 | 17-24|
| Flexibility                                            | 0-3  | 4-8  | 9-12 |
| Simplicity                                             | 0-11 | 12-24| 25-36|

| Score at National level (19 questions)                 |
|--------------------------------------------------------|-----------------------------------|
| Attribute                                              | Poor | Fair | Good |
| Acceptability                                          | 0-7  | 8-16 | 17-24|
| Flexibility                                            | 0-3  | 4-8  | 9-12 |
| Simplicity                                             | 0-12 | 13-27| 28-40|
Methodology

**Quantitative attributes:**

- **Data quality:**
  - Integrity of the form.
  - Validity of data in the system → review of clinical records of reported cases.
  - Cases reported to the system and birth, death and fetal death certificates.

- **Timeliness:**
  - Elapsed time since event occurrence until it is reported to each level.
  - Elapsed time between report of cases at national level and its dissemination.
  - Timeliness among different NTDs.

- **Representativeness:**
  - Population covered by the system.
  - Births delivered in health institutions.

- **Sensitivity:**
  - Cases collected by the system and vital records, among the total cases reported in vital records.

- **Positive Predictive Value:**
  - Cases collected by the system and vital records, among the total cases reported to the system.

- **Usefulness:**
  - Use of the data provided at each level.
  - Outcomes reports.
  - Use of the data for research.
  - Contributions to health policy and prevention programs.
Acceptability

• The system is acceptable.

• Participation rate:
  • Jurisdictional Level (n=16) → 69%.
  • Local level (n=448) → 5%.
  • NHS Local level (n=104) → 22%.

• 42% of participants (n=31) reported that the data collected are not analyzed or used.

• Inconsistencies between manual procedures and their application
  – Request to report cases of other defects not under surveillance (56% other malformations → *Hidrocephaly, holoprosencephaly, schizencephaly*, etc).
  – Report not performed by the unit that attended the birth.
  – Data collection by review of clinical record or domiciliary visit, instead by interviewing to the mother in her hospitalary stay.
RESULTS

Simplicity

- The structure and operation is simple.

- The type and amount of data needed to fill the form is large and complex:
  - Form required 96 variables,
  - Includes maternal history section, parents history which includes addictions, occupation and pesticide exposure, diseases, etc.

- The Neural Tube Defects Surveillance System does not have an electronic platform for report the cases.
  - The information is emailed through all levels.
RESULTS

Flexibility

• Case report form allow the reporting of:
  – Other types of spina bifida,
  – Other malformations of encephalus,
  – Other malformations nervous system.

• Any modification to the actual report form or defects under surveillance will increase training requirements and system costs. → 35%.

• According to personnel in charge, the surveillance system is considered regularly flexible.
Data Quality

- Case report format integrity: 97%.

- No review of cases by staff.

- Of the 106 cases, we reviewed 89 clinical records (not allowed the review of six clinical records and 11 were not found):
  - 41 cases corresponded to a neural tube defect under surveillance;
    - 73% have been confirmed;
    - 27% have different diagnosis:
      - **Anencephaly**: 33% → *Schizencephaly*.
      - **Meningocele**:
        - 56% → *Myelomeningocele*,
        - 11% → *Encephalocele*,
        - 11% → *Hydrocephaly*.
      - **Myelomeningocele**:
        - 8% → *Meningocele*
        - 4% → *Heart defect*
RESULTS

Timeliness

- Timely notification in 77% of the reported cases.

- Range of notification: 6-309 days → Median: 33 days.

- Decreases by type of birth defect.
  - Anencephaly → Not notified by the unit of attention → Found by reviewing vital registries →
    → Request investigation of the case to the unit or jurisdiction corresponding.
  
   → Delay of the report.

Timeliness by Type of Neural Tube Defect, Mexico City, 2012. (n=47)

- Myelomeningocele: 81%
- Meningocele: 78%
- Encephalocele: 71%
- Anencephaly: 50%
Representativeness

- The system covers the entire population → operates in all states and units of national health system and private sector.

- Most of births were attended in a medical unit:
  - 144,955 births in Mexico City in 2012
  - Only 0.12% (183) were not born in a medical unit (0.02% on street, 0.09% at home and 0.01% elsewhere).
  - 83% (119,857) of births were attended in public medical units.
  - 17% (24,705) born in private hospitals.
  - 0.14% (210) unknown.
RESULTS

Sensitivity and PPV

- Sensitivity and Positive Predictive Value calculations were made from the comparison of cases on vital registries which were reported by NTDSS

<table>
<thead>
<tr>
<th>Cases in NTDSS</th>
<th>NTDs cases in vital records</th>
<th>Sensitivity</th>
<th>Positive Predictive Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yes</td>
<td>Yes</td>
<td>58</td>
<td>29%</td>
</tr>
<tr>
<td></td>
<td>No</td>
<td>48</td>
<td>55%</td>
</tr>
<tr>
<td></td>
<td>No</td>
<td>141 SD</td>
<td>199</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>106</td>
<td>55%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cases in NTDSS</th>
<th>NTDs cases in vital records</th>
<th>Sensitivity</th>
<th>Positive Predictive Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yes</td>
<td>Yes</td>
<td>21</td>
<td>22%</td>
</tr>
<tr>
<td></td>
<td>No</td>
<td>26</td>
<td>45%</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>73</td>
<td>22%</td>
</tr>
<tr>
<td></td>
<td>No</td>
<td>94</td>
<td>45%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Anencephaly, Encephalocele, Meningocele, Myelomeningocele</th>
</tr>
</thead>
<tbody>
<tr>
<td>NTDs cases in vital records</td>
</tr>
<tr>
<td>-----------------------------</td>
</tr>
<tr>
<td>Yes</td>
</tr>
<tr>
<td>No</td>
</tr>
<tr>
<td>Sensitivity</td>
</tr>
<tr>
<td>Positive Predictive Value</td>
</tr>
</tbody>
</table>
Usefulness

• The system is not being used.

  • Have not been conducted reports with system outcomes at any level.

  • Annually, in Mexico, a report of morbidity is carried out, which include neural tube defects, however, the information is not of the NTDSS, but the numerical week of probable cases.

  • No articles or publications with the results provided by the system.
Summary

**Strengths**
- Timeliness.
- Flexibility.
- Representativeness

**Weaknesses**
- Data quality
- Not accepted
- Low Sensitivity (22%)
- Low PPV (45%)
- Data not further used
Conclusions

• Underreporting cases from NTDSS and vital registries

• With the results obtained, it is considered that there should be a revision of the manual, the procedures and their application, as there is inconsistency between them.

  • Notification not by a treating physician.

  • Wrong use of sources → Search of cases not performed on vital registries, case report format filled with data obtained from clinical record or domiciliary visit.

  • Report of defects not under surveillance
Recommendations

- Review of the Manual
- Review of the report form
- Clarify defects under surveillance
- Improve the registration of cases in the NTDSS and vital registries
- Staff training for detection, classification, reporting, monitoring and data collection
- Provide feedback to participants
- Disseminate outcomes from system
- Involve the participation of stakeholders
- Enable an electronic platform for the registration of cases
- Improve linkage with vital registries
Limitations

• Method used as Gold Standard does not meet the requirements:
  – Not record all cases → Underreporting.
  – Diseases at birth and causes of death → Reporting failures or lack of it.
  – Diagnoses are not confirmed.

• Difficulties in the access to the evaluation of local units, mainly in the unrelated to the Secretariat of Health.

• Difficulties in obtaining information of the cases and vital registries.
Next Steps

• Final report ➔
  – Publish and disseminate.

  – Presentation of results to:
    • Staff
    • General Director of Epidemiology.
    • Secretary of Prevention and Health Promotion.

  – Insist ➔ Recommendations made.
Thank you.
## Sensitivity and PPV by type of NTD

<table>
<thead>
<tr>
<th>NTDs cases in vital records</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Cases in NTDSS</strong></td>
<td><strong>Yes</strong></td>
<td><strong>No</strong></td>
</tr>
<tr>
<td><strong>Yes</strong></td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td><strong>No</strong></td>
<td>33</td>
<td>SD</td>
</tr>
<tr>
<td><strong>34</strong></td>
<td>3</td>
<td></td>
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</table>

### Anencephaly

- **Sensitivity**: 3%
- **Positive Predictive Value**: 25%

<table>
<thead>
<tr>
<th>NTDs cases in vital records</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Cases in NTDSS</strong></td>
<td><strong>Yes</strong></td>
<td><strong>No</strong></td>
</tr>
<tr>
<td><strong>Yes</strong></td>
<td>12</td>
<td>15</td>
</tr>
<tr>
<td><strong>No</strong></td>
<td>18</td>
<td>SD</td>
</tr>
<tr>
<td><strong>30</strong></td>
<td>15</td>
<td></td>
</tr>
</tbody>
</table>

### Myelomeningocele

- **Sensitivity**: 40%
- **Positive Predictive Value**: 44%

<table>
<thead>
<tr>
<th>NTDs cases in vital records</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Cases in NTDSS</strong></td>
<td><strong>Yes</strong></td>
<td><strong>No</strong></td>
</tr>
<tr>
<td><strong>Yes</strong></td>
<td>5</td>
<td>2</td>
</tr>
<tr>
<td><strong>No</strong></td>
<td>13</td>
<td>SD</td>
</tr>
<tr>
<td><strong>18</strong></td>
<td>2</td>
<td></td>
</tr>
</tbody>
</table>

### Encephalocele

- **Sensitivity**: 28%
- **Positive Predictive Value**: 71%

<table>
<thead>
<tr>
<th>NTDs cases in vital records</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Cases in NTDSS</strong></td>
<td><strong>Yes</strong></td>
<td><strong>No</strong></td>
</tr>
<tr>
<td><strong>Yes</strong></td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td><strong>No</strong></td>
<td>17</td>
<td>SD</td>
</tr>
<tr>
<td><strong>18</strong></td>
<td>2</td>
<td></td>
</tr>
</tbody>
</table>

### Meningocele

- **Sensitivity**: 75%
- **Positive Predictive Value**: 33%
Timeliness

• Date of birth → Report at National level → 6 to 309 days.

• New Variable →
  – Cases reported before 45 days → Timely.
  
  – Cases reported after 46 days → Not timely.
Knowledge of the NTDSS operating guidelines

Knowledge of personnel in charge of the NTDSS about defects under surveillance, definitions of case and type of notification (n=31)

- 90% have reviewed the manual
- 58% know the defects under surveillance
- 35% know the type of notification
- 32% mention other birth defects
- 26% know the four operational definitions of case
Study Population

- National: 1
- State: 1
- Jurisdictional: 11
- Local: 23

Conducted: 106 cases

Estimated: 84% (n= 89 )

84% (n= 89 )

Conducted
Final Diagnosis

- **Anencephaly (n=3)**
  - 2 Anencephaly
  - 1 Other Malformations of encephalus (Schizencephaly)
  - Total: 3

- **Meningocele (n=9)**
  - 2 Meningocele
  - 1 Encephalocele
  - 5 Myelomeningocele
  - 1 Hydrocephalus
  - Total: 4

- **Myelomeningocele (n=24)**
  - 21 Myelomeningocele
  - 2 Meningocele
  - 1 Heart Defect
  - Total: 27

- **Encephalocele (n=5)**
  - 5 Encephalocele
  - Total: 6

In total, there are 36 cases.
## Final Diagnosis

<table>
<thead>
<tr>
<th>Category</th>
<th>Details</th>
<th>Count</th>
</tr>
</thead>
<tbody>
<tr>
<td>Other spina bifida</td>
<td>1 Hidrocephaly, 1 None</td>
<td>0</td>
</tr>
<tr>
<td>Other congenital malformations of encephalus (n=9)</td>
<td>8 Other congenital malformations of encephalus → Holoprosencephaly, 1 Hidrocephaly</td>
<td>16</td>
</tr>
<tr>
<td>Other congenital malformations of nervous system</td>
<td>1 Anencephaly, 1 Myelomenigocele, 7 Other congenital malformations of encephalus → 4 Holoprosencephaly, 1 Schizencephaly, 1 congenital malformations of corpus callosum, 1 Hydranencephaly, 22 Hidrocephalus, 3 Microcephaly, 1 Macrocephaly, 2 None</td>
<td>0</td>
</tr>
</tbody>
</table>
Diagnostic Method

89 cases

Prenatal Diagnosis
- Ultrasonography: 64%

Postnatal Diagnosis
- Clinical: 28%
- Computed Axial Tomography: 4%
- Magnetic Resonance Imaging: 1%
- Radiography: 1%
- Unknown: 1%
Case Definition of NTDs

- **Anencephaly case:** newborn alive or dead, more than 20 weeks of gestational age, which is detected clinically or by imaging studies, the absence of the skull bones and large part of the brain.

- **Meningocele case:** Individual with one or more saccular defects, broken or intact, at the dorsal midline, in any location: cervical, thoracic, lumbar or sacral, containing inside meninges and CSF, independent of the degree of neurological impairment.

- **Myelomeningocele case:** Individual with one or more saccular defects, broken or intact, at the dorsal midline in cervical, thoracic, lumbar or sacral location, containing inside spinal cord and / or nerve roots, meninges and CSF, independent of the degree of neurological impairment.

- **Encephalocele case:** Individual with presence of meningeal saccular herniation by a defect in the skull bones, which contains brain tissue.