Current research and previous studies
The importance of interdisciplinary studies in health and Usher syndrome

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Outline for the presentation

Previous studies
Health and people with Usher syndrome
Health, work and people with Usher syndrome type 1 and 2

Ongoing studies
Strategies in life for persons with Usher syndrome type 2a
Lived experiences of work and health in persons with Usher syndrome type 2
Health and family life in families where one parent has deafblindness
Health and people with Usher syndrome

Overall aim was to describe health of people with Usher syndrome
The Swedish Usher register

Swedish National Register including individuals with Usher syndrome
All three types included
Audiograms and visual test
Genetic diagnose

The Swedish health questionnaire “Health on equal terms”
Hospital Anxiety and Depression Scale

Reference population
The Public Health Agency of Sweden
A cross section of the Swedish population
Includes individuals with and without impairments
## Participants

<table>
<thead>
<tr>
<th></th>
<th>Reference</th>
<th>USH1</th>
<th>USH2</th>
<th>USH3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number</td>
<td>5738</td>
<td>60</td>
<td>96</td>
<td>15</td>
</tr>
<tr>
<td>Age mean (years)</td>
<td>49</td>
<td>49</td>
<td>55</td>
<td>41</td>
</tr>
<tr>
<td>Age (min-max)</td>
<td>16-84</td>
<td>20-79</td>
<td>18-84</td>
<td>19-71</td>
</tr>
<tr>
<td>Women</td>
<td>56%</td>
<td>60%</td>
<td>53%</td>
<td>71%</td>
</tr>
<tr>
<td>Clinical diagnose</td>
<td>60</td>
<td>96</td>
<td>15</td>
<td></td>
</tr>
<tr>
<td>Genetic diagnose</td>
<td>43%</td>
<td>59%</td>
<td>73%</td>
<td></td>
</tr>
<tr>
<td>Mean HL</td>
<td>99 db</td>
<td>73 db</td>
<td>99 db</td>
<td></td>
</tr>
<tr>
<td>Mean Visual acuity (1,0-0,0)</td>
<td>0,5</td>
<td>0,4</td>
<td>0,7</td>
<td></td>
</tr>
<tr>
<td>Mean Visual field (1-5)</td>
<td>3</td>
<td>4</td>
<td>3</td>
<td></td>
</tr>
</tbody>
</table>
Results

- Poor physical health
- Social trust
- Poor psychological health
- Financial situation
People with USH regardless of clinical type, and in comparison with a cross-section of the Swedish population, report problems with physical and psychological health, lack of social trust and a strained financial situation.

A biopsychosocial perspective is needed when describing the health of people with Usher syndrome otherwise there is a risk of reducing the complexity and the consequences of living with deafblindness.
Work and Health in persons with Usher type 1 and 2

The aim was to describe relations between work and health in persons with Usher syndrome type 1 and 2
Participants

Usher type 2
Working group 34 individuals
Disability pension 33 individuals
No differences in hearing or vision
Slightly more women in both groups

Usher type 1
Working group 23 individuals
Non working 24 individuals
43% women working group and 63% non working group
No differences in hearing or vision
Reference population
Results Usher type 2

Significantly more psychological health problems reported from the individuals with disability pension.

Physical health did not present with the same pattern.
Results Usher type 1

The Usher non working group (and the reference non working group) reported to have more problems with psychological and physical health problems as well as problems with ADL than did the working groups.

Problems with social trust was more pronounced in the Usher type 1 non working group, except for being offended where persons with Usher type 1 who worked reported more problems.

Financial situation
Life strategies and facilitating factors when having deafblindness due to Usher syndrome type 2a

The aim of the study was to explore life strategies in people with Usher syndrome type 2a
Methods

Explorative qualitative study design with focus group interviews of 14 individuals with Usher syndrome type 2a.

Participants where 4 women and 10 men aged 20-64. Moderate to severe hearing loss, range 48-80db (mean 65db).
Visual field range 2-5 (median 4)
Visual acuity range 0,05-1,0 (mean 0,4)

Three focus groups, audio and video recorded.
Preliminary results

• Resolving or preventing challenges

• Comforting oneself
Resolving and preventing challenges

It is about being able to have the imagination to find new ways, not being blind to the fact that there is more than one way…for example you may not need to hammer in nails when you can use a screwdriver. (Person 14)

I can be independent when I have a guide or interpreter or whatever, then I’m independent. But if I don’t have one and want to go shopping then I become dependent on finding some shop assistant who’ll help me. And that’s when I’m not independent. (Person 3)
Comforting oneself

So I feel that you have to do what you are capable of when you are capable of it, then you get the most out of activities you want to do. That’s what I believe anyway. (Person 1)

It is like when I share a bath with my daughter, then we light a lot of candles, which she really loves. And then you relax and sort of feel content and in the here and now. Because then you are blissfully unaware of your problems, even though they still exist. (Person 2)
Being at the helm

The participants are committed agents in a process of striving to live a active life in accordance with their own values.
Conclusion

The participants express a variety of strategies to handle consequences of their deafblindness. By being at the helm the participants express a high psychological flexibility and are active agents in their life's.
Ongoing and upcoming studies

• Lived experience of the relation between health and working life of people with Usher syndrome type 2

• Health and family climate in families where a parent has deafblindness

• Update of the health and people with Usher syndrome

• Experiences of support given in rehabilitation/habilitation settings for adults with deafblindness
Thanks for the attention!

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